Discriminating between Autism and Disinhibited Social Engagement Disorder in a clinical context.

Claire Davidson-Jamieson

Student number: xxxxxxx

PhD thesis by alternative (journal) format

Submitted in fulfilment of the requirements for the degree of Doctor of Philosophy

Institute of Health and Wellbeing

Medical and Veterinary Life Sciences

Abstract

Clinicians are concerned about making accurate differential diagnoses between Autism and Disinhibited Social Engagement Disorder (DSED) because both groups of children may present with social relationship and communication difficulties, yet DSED is associated with maltreatment and Autism is not. The overall objective of the thesis was to identify any skills or behaviours which may help clinicians discriminate between these two diagnostically distinct groups.

This thesis brings together the findings of an-depth case study investigation, discussed across four separate but related papers, each of which addresses a gap in our knowledge regarding DSED, and how it may be differentiated from Autism. Paper 1 is a systematic review assessing the social functioning of children with DSED. Paper 2 directly compares the profiles of children with Autism, children with DSED and children who are typically developing (TD) via current 'gold standard' autism assessment and an unstructured behavioural observation called the Live assessment. Papers 3 and 4 expand on the areas of possible differentiation which were highlighted in papers 1 and 2; language and social communication (paper 3) and sensory processing (paper 4).

Specific differences in skills/behaviours of children with Autism in the case study, compared to the children with DSED, were most apparent within the domain of social communication, suggesting that future research focused on differentiating Autism from DSED should focus on this area. There was also a tentative suggestion that some sensory behaviours may be more 'Autism-specific.' For complex cases, a change in approach from standardised structured Autism assessment to a *holistic neurodevelopmental approach* using unstructured observation, which includes conversational elements, and increases the social challenge may more easily elucidate the differences between core Autism behaviours, DSED-specific behaviours and other co-occurring neurodevelopmental conditions like Attention-Deficit/Hyperactivity Disorder (ADHD) which can complicate the picture. While awareness raising and training may be indicated, we do have tools, like the Live assessment and clinical expertise, such as Speech and Language Therapists (SLTs), available, which can be utilised to support differential diagnosis of the 'hard to assess' cases.

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Acknowledgements

First and foremost, I would like to thank all the families who participated in this doctorate study. Without them, this work would not have been possible, and our understanding of the experiences of children with Autism or children with DSED would not be enriched.

Following from this, I would like to acknowledge the contribution of the funders- Castang Foundation, Gillberg Neuropsychiatry Centre and NHS Greater Glasgow and Clyde. I am extremely fortunate to have worked and enhanced my research skills and knowledge in a field that I am extremely passionate about. I would also like to acknowledge the support of the Scottish Centre for Autism for their wisdom, and training, and my PhD colleagues for their banter. Special mention to Fiona Turner for her enthusiasm and time in undertaking Live assessments with me.

Related to this, I must thank every one of my supervisors, Professor Helen Minnis, Professor Christopher Gillberg and Professor Anja Lowit, for their knowledge, constructive feedback, clarity and ongoing support. When I first began working with Professor Minnis as a research assistant on the predecessor project to this study, we shared a vision regarding the importance of better understanding the overlaps between Autism and DSED, and the desire to investigate further. I am delighted to be able to present the fruits of our labour and look forward to the next steps in our ongoing shared vision.

On a more personal note, I would also like to thank Professor Minnis, especially, for her unwavering belief in me. When I became chronically physically ill, Professor Minnis knew she had to let me stop my work, but always made it clear that the return door was always open, if/when the time was right. That human understanding cannot be underestimated, and it was so amazing to walk back through that open door. I must thank Professor Gillberg and Professor Lowit too for also keeping the door open and enabling us all to pick right back up like time had never passed.

Finally, my husband Neil, he deserves the greatest of acknowledgement for he has been a steadfast rock and an example of unconditional love, and for that I am so grateful. I am also grateful to my family who embody strength and resilience and have taught me so much. My support network would not be complete without my pets, my cats and chickens, who provide so much fun, peace and company. They are perhaps the unsung heroes who allowed me to chill out with them whenever I needed a thesis break.

Thank you to all who choose to read this thesis. I hope you enjoy and learn somethings useful, like I have.

List of papers

Paper 1: Published (in Chapter 2)

Davidson, C., Islam, S., Gillberg, C., Lowit, A., Venturini, E & Minnis.H. (2023) Social competencies of children with Disinhibited Social Engagement Disorder: A systematic review

Paper 2: Published (in Chapter 4)

Davidson, C., Turner, F., Campbell, Gillberg, C., Campbell, S.L., Boyd, S & Minnis.H. (2023). Using the live assessment to discriminate between Autism Spectrum Disorder and Disinhibited Social Engagement Disorder. *Research in Developmental Disabilities.* 134, 1-10

Paper 3: Submitted to journal– under review (in Chapter 5)

Davidson, C., Gillberg, C., McCool, S., Elder, B., Minnis, H & Lowit. A. (2023) Autism and Disinhibited Social Engagement Disorder: Overlaps and differences in language and communication skills.

Paper 4: Submitted to journal– under review (in Chapter 6)

Davidson, C., Gillberg, C., Lowit, A & Minnis.H. (2023). An exploratory comparison of sensory problems in children with Autism and children with Disinhibited Social Engagement Disorder

Contribution to papers

Paper 1: I was responsible for the conceptualisation of the research question, which was discussed and agreed with H, Minnis, C, Gillberg and A, Lowit (supervisors). I was responsible for the methodology, the investigation in collaboration with S, Islam, H, Minnis and E, Venturini and I completed the formal analysis. I was responsible for writing the original draft and the review and edits. All co-authors contributed to the final review/edits and particularly, H, Minnis, C, Gillberg and A, Lowit.

Paper 2: I was responsible for the conceptualisation of the research aims and objectives and methodology, which were discussed and agreed with H, Minnis, C, Gillberg and A, Lowit. S.L, Campbell and S, Boyd contributed to the methodology. I was responsible the investigation and formal analysis. F, Turner contributed to the investigation. I was responsible for writing the original draft and the review and edits. All co-authors contributed to the final review/edits and particularly, H, Minnis, C, Gillberg and A, Lowit.

Paper 3: I was responsible for the conceptualisation of the research aims and objectives and methodology, which were discussed and agreed with H, Minnis, C, Gillberg and A, Lowit. I was responsible the investigation and formal analysis. S, McCool and B, Elder contributed to the formal analysis. I was responsible for writing the original draft and the review and edits. All co-authors contributed to the final review/edits and particularly, H, Minnis, C, Gillberg and A, Lowit.

Paper 4: I was responsible for the conceptualisation of the research aims and objectives and methodology, which were discussed and agreed with H, Minnis, C, Gillberg and A, Lowit. I was responsible the investigation and formal analysis. I was responsible for writing the original draft and the review and edits. H, Minnis, C, Gillberg and A, Lowit contributed to the review and edits.

Key terms with definitions

Autism Spectrum Disorder: is a neurodevelopmental condition characterised by impaired social interaction, social communication and repetitive and restricted behaviours, as described by the Diagnostic Statistical Manual of Psychiatric Disorders-5 (DSM-5) or the International Classification of Disease- 11 (ICD-11).

Autism: is the term used within the thesis to describe individuals who have been diagnosed with Autism Spectrum Disorder. The choice was made to use the term Autism in the main body of the text because individuals with Autism, and many professionals and researchers, are calling for the use of less pathologizing terms (disorder).

Disinhibited Social Engagement Disorder (DSED): is described by DSM-5 and ICD-11 as a trauma and stressor associated disorder. It is characterised by indiscriminate sociability and poor social boundaries and is associated with maltreatment (neglect and/or abuse). DSED is *only* diagnosed where there is known evidence of childhood maltreatment.

Holistic neurodevelopmental assessment: assessing **broadly** across multiple domains of neurodevelopment, anticipating that more than one neurodevelopmental condition may present and identifying **all** the child's needs or traits within their neurodevelopmental profile, including diagnosis (es) where appropriate. In cases of known childhood maltreatment, this could also include identification of DSED.

ESSENCE: stands for Early Symptomatic Syndromes Eliciting Neurodevelopmental Clinical Examination (Gillberg, 2010) and is a framework to support thinking around holistic neurodevelopmental assessment. It proposes that severe problems with language, motor coordination, sleep, mood etc. at age 3-5 years are indicative of one or more neurodevelopmental conditions including, Autism, Attention-Deficit/Hyperactivity Disorder (ADHD), Developmental Coordination Disorder (DCD), Language Disorder and Intellectual Disability (ID). Co-occurrence of neurodevelopmental conditions is expected.

Receptive Vocabulary: is the ability to understand simple vocabulary (single words) when it is heard. It is a fundamental building block to development of more advanced language skills.

Receptive Language: is the ability to understand and make sense of heard vocabulary in combination with grammatical structures to infer meaning from phrases, clauses and sentences. It

often requires understanding how one thing (concept) relates to another, which is known as semantic reasoning.

Expressive Language: is the ability to combine words with correct grammatical structures to form phrases, clauses and sentences to verbally communicate in an interaction.

Non-verbal Language: the use of body language to communicate i.e. through gestures with hands, eye contact, facial expression etc. or changes in tone of voice.

Pragmatic Language: specifically relates to how individuals use context to infer meaning in social interactions. Context can include: the social situation/environment, words and language heard, the 'unwritten' social rules and non-verbal cues; all of which are quickly interpreted and synthesised to make sense of/draw inference about the intentions of the other person in the social interaction, their meaning, how they might next react, and allows the individual to respond to the other appropriately.

List of Abbreviations

ASD	Autism Spectrum Disorder
ADI-R	Autism Diagnostic Interview-Revised
ADOS-2	Autism Diagnostic Observation Schedule -2
ADHD	Attention Deficit Hyperactivity Disorder
BEIP	Bucharest Early Intervention Project
CCC	Child Communication Checklist
CSP	Child Sensory Profile
CELF	Clinical Evaluation of Language Fundamentals
DISCO	Diagnostic Interview of Social Communication and Other Disorders
DSED	Disinhibited Social Engagement Disorder
DSM-5	Diagnostic and Statistical Manual of Mental Disorders – 5th edition
ERA	English and Romanian Adoptees (study)
ESSENCE	Early Symptomatic Syndromes Eliciting Neurodevelopmental Clinical Examination
HFA	High Functioning Autism
ICD-11	International Classification of Diseases – 11th edition
NICE	National Institute for Health and Care Excellence
RAD	Reactive Attachment Disorder
RADA	Reactive Attachment Disorder and Disinhibited Social Engagement Disorder Assessment
RPQ	Relationship Problems Questionnaire
ToM	Theory of Mind
UK	United Kingdom
US	United States (of America)
WRO	Waiting Room Observation

Chapter 1: Introduction

1.1. Overview of section

The shared theme connecting the four papers within this thesis regards the differential diagnosis of Autism from Disinhibited Social Engagement Disorder (DSED). There is ongoing clinical concern that symptoms of Autism may overlap with symptoms of DSED, yet DSED is thought to be caused by maltreatment and Autism is not. Clinicians, therefore, are striving to find greater understanding regarding what differentiates Autism from DSED, and which tools best support the process. This section begins with an overview of the core features related to each disorder, then outlines why differential diagnosis is so important. Focus is then given to the evidence base regarding the Autism-DSED overlap and what is currently known regarding differentiation. This section ends with a summary discussion of the remaining gaps in literature: 1. social functioning of children with DSED, 2. which tools best support discrimination between Autism and DSED, 3. The language and communication skills of children with DSED compared to children with Autism and 4. the sensory processing profiles of children with DSED compared to children with Autism. Each paper addresses one of these gaps and each is presented in the chapters which follow. The section concludes with discussion of the overall research aim and related research questions.

1.2 What is Autism?

Autism is a lifelong neurodevelopmental condition which is present from birth and is characterised by difficulties with social interaction and communication and behaviours/interests which are repetitive, narrow in focus, obsessive and/or inflexible in nature (American Psychiatric Association (APA), DSM-5, 2013; World Health Organisation (WHO), ICD-11, 2019/2021). Yet, the diagnostic classification has undergone changes since the fifth edition of the Diagnostic and Statistical Manual for Mental Disorders (DSM) was released. In DSM-5 (2013), the previous overall umbrella term of Pervasive Developmental Disorder (PDD), was replaced by Autism Spectrum Disorder (ASD) and the Childhood Autism and Aspergers Syndrome sub-classifications removed. This change has been somewhat controversial but was meant to reflect that core symptoms are shared and are on a continuum, rather than categorically separate (Volkmar, 2013). Symptom severity is now rated on three levels, each relating to greater severity of impact on adaptive functioning and need for support. ICD-11, (2019/2021) has taken the same approach. In this thesis, where the term Autism is used, it is in reference to the DSM-5 and ICD-11, but any studies using previous classifications will be discussed related to their diagnostic context.

The second major change within the DSM-5 and ICD-11 criteria is that Autism is now considered as a dyad of impairment, instead of a triad of impairment. Essentially the core symptoms of

impaired social interaction, impaired communication and repetitive and restrictive behaviours, which made up the triad, remain. However, in DSM-5, impaired interaction and communication are regarded as one conjoined problem, largely because the skills and deficits related to each are generally intertwined (Mandy et al., 2012).

The final change in DSM-5 and ICD-11 is that sensory problems are now considered as one of the four possible areas of difficulty that encompass the second part of the dyad: repetitive and restrictive behaviours. This addition was a consequence of the body of evidence which has demonstrated that children with Autism invariably present with one or more sensory processing problems (Tomchek and Dunn, 2007; Tomchek et al., 2014), and additionally that repetitive sensory behaviours may discriminate Autism from other disorders such as Intellectual Disability (ID) (Adrien, 1987; Dahlgren and Gillberg, 1989; Rapin 1996).

1.2.1 Cognitive theories underpinning Autism.

The DSM-5 identifies specific behaviours which are associated with each domain of the dyad. Regarding social interaction and communication, core difficulties are: 1. reduced social-emotional reciprocity, (lack of back and forth in conversation, limited attempts to initiate or respond to interaction, limited sharing of interests or emotions, inappropriate social approach and/or limited awareness of personal boundaries). 2. Deficits in nonverbal communicative behaviours used for social interaction, (eye contact, use of/understanding of facial expressions, gestures, body language and integration of these with verbal communication) and 3. Deficits in developing, understanding and maintaining relationships, (lack of adjustment to different social environments, difficulties making friends, limited imagination skills in social play, and/or lack of interest in peers) (DSM-5).

One prominent cognitive theory which aims to explain the social-emotional difficulties of children with Autism is that of impaired Theory of Mind (ToM) (Baron-Cohen, 1985; 2000); that is, one's ability to mentalize or understand others' cognitions, intentions, desires and emotional states. Studies with both children and adults appear to support the proposition that theory of mind skills are impaired in Autism (Happe, 1994; Happe, 1994; Colle et al., 2007; Yirmiya, Erel, Shaked & Solomonica-Levi, 1998; Tager-Flusberg, 2007). For example, meta-analysis has found significantly greater ToM problems in children with Autism compared to individuals with ID, (Yirmiya, Erel, Shaked & Solomonica-Levi, 1998). Greater ToM problems have been reported even when language demands were reduced to a minimum, (Colle et al., 2007) and in a recent review of the research, Tager-Flusberg, (2007) concluded that the current research does provide evidence to suggest that in Autism, theory of mind skills are impaired, but in addition to problems with social-affective information processing. The main criticism of theory of mind as a possible underpinning of Autism is the evidence that some individuals do pass the commonly used false

belief task, (Happe,1994). However, limitations in measurement may account for some of these findings (Livingston, Carr & Shah, 2019; Baron-Cohen, 2000); for example, Happe, (1994) found that individuals who passed first, or even second order, ToM tasks still showed significant impairment when assessed using more naturalistic and complex stories.

Similar to the ToM deficit, is that of impaired social imagination, or 'social instinct,' (Wing, Gould, & Gillberg, 2011). Here, impaired social imagination is described as 'decreased capacity to think about and predict the consequences of one's own actions for oneself and for other people.' In other words, children with Autism may find it difficult to intuitively know their role in any given social interaction and to predict what may come next, or what they should say or do next. The authors give the example that individuals may be able to show *sympathy* in situations where they can perceive the overt distress of an individual, and respond to it, but they may still lack empathy for it (imagine themselves in the other's situation) and further will likely have difficulties understanding more subtle signs of emotional upset, including those caused by their own behaviour (Wing, Gould and Gillberg, 2011). Wing et al, go as far as to argue that social imagination is a core autistic characteristic which is detrimentally missed in the DSM-5 diagnostic criteria (Wing, Gould and Gillberg, 2011).

Weak Central Coherence (WCC) is the other prominent cognitive theory and is a difficulty incorporating different levels of information (details) and integrating that information into a coherent whole (bigger picture) (Frith, 1989). In other words, children with Autism are more likely to show skill, pay attention to and/or focus in on the fine details and may have problems seeing the gestalt. It seems to fit with one area of social communication which is thought to be universally impaired in individuals with Autism, that of pragmatic language (Rhea, 2007). Pragmatic Language relates to the ability to use the linguistic context (words, language, intonation/tone of voice) to inform meaning i.e., to draw inference or make sense of ambiguities by integrating the language provided with their knowledge from previous experience (Norbury, 2014) and the available contexts (social situation, nonverbal communication). Impaired WCC, could explain why children may pick up key facts but miss the gist of the story, and it could impact what they notice and respond to, or what they are interested in/focused on. This in turn could negatively impact narrative discourse which is another prominent feature of impaired pragmatic skills (Adams, 2002). One study found that in a clinic referred sample described as High Functioning Autism (HFA, pre-DSM-5) (n=31), children who failed the ToM task were impaired in their description of common events in a general fashion and in describing activities in the time-order that they'd normally occur, once verbal IQ was controlled for. Yet, children who passed the ToM task, still demonstrated difficulties with narrative of familiar events, as they focused on detail rather than a global sense and demonstrated inflexibility in their expectations regarding events (Burnette et al., 2005). Happe, (1995) argues that WCC is somewhat related to

ToM, as mentalising skills may be hampered because it is more difficult to understand others' emotions or perspective if the relevant details and social context are not accounted for.

These cognitive theories, together, do appear to make some sense of the social interaction and communication difficulties often seen in Autism. With regards to the second part of the dyad of impairment, repetitive and restricted behaviours, it could be argued that a need for sameness, predictability and routine is compensatory or even protective if difficulties with understanding one's role in social interactions and taking account of others' thoughts and emotions are difficult. Furthermore, if there is a bias to focus in on the specifics rather than the whole, this may also fit with the pattern of specific interests and rigid attention to these interests, often seen in Autism. The diagnostic criteria states that individuals must demonstrate one of the following: 1. Stereotyped or repetitive motor movements, use of objects or speech, 2. Insistence on sameness, inflexible adherences to routines or ritualized patterns of speech and behaviour, 3. Highly restricted, fixated interests that are abnormal in intensity or focus and 4. hyper -or-hypo reactivity to sensory input or an unusual interest in sensory aspects of the environment (DSM-5).

Some researchers are beginning to challenge to the posited cognitive theories and are seeking to define the core social difficulties and attentional bias in children with Autism as related to poorly integrated sensory processing. For example, Milton, (2017) argues that for all individuals, attention is a scarce resource and one must strategize what they attend to by balancing sensory information with their beliefs and previous knowledge. But, for individuals with Autism, overwhelm of sensory input can make this much harder. Therefore, he argues, it is supportive to filter out what they cannot attend to, be specific regarding what they do give attention to, or be absorbed only by what is of most interest. Given the recent acknowledgement of sensory processing problems as part of the core symptomology, it seems reasonable that such thinking is further researched.

1.2.2. Aetiology

The estimated population prevalence for Autism in the UK and internationally is 1-1.9%, Rydzewska et al., 2019; WHO, 2023). While the diagnosis of Autism has increased in recent years, robust investigations suggest that this is not necessarily due to an increase in Autism symptoms, which have remained stable in the general population (Lundstrom et al., 2015; Russell et al., 2022), but more likely due to greater awareness among clinicians and in schools, increased awareness regarding Autism in adults and in females (Russell et al., 2022), which can present more subtly, media attention and to some degree, broader diagnostic criteria with the DSM-5 re-classification (Lundstrom et al., 2015; Volkmar, 2013).

Research has gone some way to understanding causes of Autism, as there is evidence to suggest Autism runs in families and in most cases there is a strong genetic component (Delorme et al.,

2013). For example, a large birth cohort of combined data from 5 countries (n= >2 million) found that inherited genetic factors were associated with 81.2% of the variance in Autism occurrence, (Bai et al., 2019) and a meta-analysis estimated heritability at 64%-91% following analysis of all heritability studies of Autism (Tick et al., 2016). Yet, the picture is not clear-cut because in many cases, causes often remain unknown and environmental factors can play a role. There is increased risk of Autism in children born pre-term, particularly where there is a very low birth weight (Limperopoulos, 2009) and some teratogens increase the risk of development of Autism. For example, foetal exposure to valproic acid, a common epileptic drug, also used in the treatment of bipolar disorder and schizophrenia (Moore et al., 2000; Christensen et al., 2013; Rasalam et al., 2005; Williams et al., 2001) and, in cases of alcohol exposure, Foetal Alcohol Syndrome (Aronson, Hagberg & Gillberg., 1997; Landgren et al., 2010; Harris, McKay & Osborn., 1995). In addition, Autism has been found to co-occur in neurological conditions, (Ryland et al., 2012) including Cerebral Palsy, (Bjorgass et al., 2014).

One environmental theory that has been emphatically discredited is that cold or harsh parenting causes Autism. This theory was known as the 'refrigerator mother theory of autism' (Bettelheim, 1967). Both Rutter et al and Rimland, in their separate studies, demonstrated that the parenting of children with Autism and the parenting of non-autistic controls did not differ (Rimland, 1964; Rutter, 1968). Furthermore, the English and Romanian Adoptees (ERA) study investigated the possibility of links between environmental adversity and Autism. The ERA studies were conducted by a group of UK based researchers who longitudinally followed children adopted within the UK who had been institutionalised, usually from birth, in social-emotionally and environmentally deprived Romanian institutions (Sonuga-Barke et al., 2017). In this severely deprived population (n = 111), when additional risk factors such as premature birth were accounted for, the prevalence of Autism was 1.8%, (Rutter et al, 1999) which is similar to the estimated UK prevalence of Autism in the general population (up to 1.9%, Rydzewska et al., 2019).

Children with neurodevelopmental conditions or Intellectual Disability (ID) are known to be at higher risk of being maltreated (McDonnel et al., 2019), however, a recent study which followed a large cohort of Swedish twins from the general population (n= 8,192 9-year-old twins), investigated whether maltreatment was a risk factor for increased neurodevelopmental problems. Although findings suggested that maltreated children had a greater number of neurodevelopmental problems than non-maltreated children, monozygotic twins discordant for maltreatment did not significantly differ in number of neurodevelopmental disorders. Common genetic effects explained most of the covariance of maltreatment with neurodevelopmental problems (Dinkler et al., 2017). There appears to be no *direct* link between maltreatment and

development of Autism, and this is crucial to keep in mind as we now turn attention to the discussion of Disinhibited Social Engagement Disorder (DSED).

1.3. What is Disinhibited Social Engagement Disorder (DSED)?

DSED is a disorder of social relatedness (Zeanah and Gleason, 2015) which is associated with maltreatment (abuse or neglect) (DSM-5) but is usually diagnosed only when there is known evidence of maltreatment. Maltreatment can be defined as exposure to threat, which would include physical, sexual and emotional abuse, or severe deprivation from expected inputs, for example physical, emotional and communicational neglect (McLaughlin, 2016; Humphreys and Zeanah, 2015). Within DSM-5, DSED is described as a 'trauma and stressor' related disorder, characterised by *a pattern of behaviour in which the child actively approaches and interacts with unfamiliar adults.* To meet the DSM-5 diagnostic criteria, in addition to maltreatment, two of the following behaviours must be present: overly familiar verbal or physical behaviour (that is not consistent with culturally sanctioned and age-appropriate social boundaries); diminished or absent checking back with a caregiver after venturing away, or willingness to go off with an unfamiliar adult with minimal hesitation (DSM-5). These behaviours are considered to be inappropriate, but social in nature and not the result of impulsivity, as might be seen in ADHD. DSM-5 also recommends that Autism is ruled out before making a diagnosis of DSED. This is, in part, why clinicians are concerned about correctly identifying DSED, but more so because the lack of social boundaries described above can be found in some children with Autism. Other core features of Autism such as lack of social reciprocity, empathy and poor awareness of social cues were associated with DSED in the ERA studies (Rutter et al., 1999) and in clinic referred community samples of children in the US and in Europe, (Pears et al., 2010; Mukaddes, Bilge, Alyanak, & Kora, 2000). Two recent studies from the Bucharest Early Intervention Project (BEIP) have also found that children with DSED have poorer social competencies than peers (Guyon-Harris et al., 2019). The BEIP was another seminal study in this field, conducted by a team of US led researchers who investigated the longitudinal outcomes of a foster care intervention for severely deprived institutionalised children in Romania. BEIP was a randomised control study including 6 institutions and 56 foster homes. Children (n=136) were randomly allocated to care as usual (the Romanian institution) or the foster care intervention which was specifically set up for the project within the infrastructure of the Romanian community and was overseen by social workers who were specially trained by the research team, and supervised by US based psychologists, to provide care within a nurturing framework (Zeanah, Fox & Nelson, 2012).

Reactive Attachment Disorder (RAD) is the other 'trauma and stressor related' disorder within DSM-5, which shares the aetiology of childhood maltreatment. The difference between RAD and DSED is that children with RAD tend to present with internalising behaviours (withdrawn and

anxiety related behaviours) rather than some of the externalising behaviours (e.g. impulsivity and attention seeking) seen in DSED, (Lehmann, 2016) but both RAD and DSED are associated with additional conduct problems (Mayes et al., 2017; Seim et al., 2022). The other difference is that RAD is primarily associated with disturbed attachment behaviour (Zeanah et al., 2016), for example, failure to seek comfort from primary caregivers (Lehmann et al., 2016) whereas DSED is not primarily associated with attachment. For this reason, in DSM-5, DSED was re-classified as a distinct disorder from RAD, rather than a sub-type of RAD as in previous classifications (DSM-IV; ICD-10). For example, in both the ERA and BEIP studies, and in clinic referred community samples of children in the UK and Norway, RAD has been found to dissipate once children are placed within a nurturing environment and attachment relationships have been formed (Rutter et al., 2010; Zeanah and Gleason, 2015; Zeanah et al., 2016; Bruce et al., 2019; Turner et al.,2022), whereas DSED tends to persist (O'Connor et al., 2003; Zeanah et al., 2005; Minnis et al., 2007; Lyons-Ruth et al., 2009; Gleason et al., 2011; Kennedy et al., 2017; Guyon-Harris et al., 2019 and Seim et al., 2022). In the ERA study (n=111), although indiscriminate behaviours were associated with insecure attachments (insecure plus indiscriminate, n=12), in some cases, these indiscriminate behaviours were also apparent in children with secure attachments (secure plus indiscriminate n=3) (O'Connor et al., 2003). In BEIP (n=136), it was found that in toddlers, DSED symptoms were not associated with attachment classification (secure/insecure etc) in almost half the sample, although at 42 months, in some cases, there was a moderate reduction in DSED symptoms as secure attachment increased. As in the ERA study, there were children with secure attachments who demonstrated high levels of indiscriminate behaviours (Gleason et al., 2011). Guyon-Harris et al, (2019), (BEIP) later demonstrated that symptoms of DSED can be identified in adolescence in post-institutionalised adoptees and Seim et al, (2022), investigating in Norway (n=31), and Moran et al, (2023), investigating within the UK (n=110), demonstrated persistence into adolescence in residential and prison samples respectively. Kennedy et al, (2017) reported persistence of DSED in young adults followed up from ERA.

Despite potentially having a shared aetiology, DSED and RAD also present with distinct symptom clusters to each other (Gleason et al., 2011; Lehmann et al., 2016) and from other forms of psychopathology (Lehmann et al., 2016), although co-occurrence is also common. New evidence has also suggested that the symptom 'failure to check back in with caregivers', which is also an attachment-related behaviour, but is considered in DSM-5 as a symptom of DSED, may factor better with RAD symptoms than DSED symptoms (Monette et al., 2022).

The DSM-5 re-classification brought about the change in nomenclature from disinhibited-RAD (DSM-4)/Disinhibited Attachment Disorder (ICD-10) to Disinhibited Social Engagement Disorder, (DSED) but it is of note that changes occurred only in classification and terminology, and not regarding core symptoms. This means that the population within studies using the old

terminology still represent the population now described as DSED. The slightly messier issue of terminology arises where studies pre-dating DSM-5 used only the umbrella term RAD, rather than differentiating between the sub-types, disinhibited-RAD and inhibited-RAD. However, we now know that DSM-5 RAD (previously the inhibited sub-type) is rare, on its own, within the population, (Minnis et al., 2013; Zeanah et al., 2000) and, as mentioned above, it tends to dissipate with nurturing care in contrast to DSED which tends to be persistent. For example, Minnis et al screened 1646 6-7 years old children in the general population and only found one case of RAD (Minnis et al., 2013). Thus, we use the moniker $DSED^{RAD}$ in reference to studies that used the historical umbrella term RAD, as it is likely that the population referred to actually consists largely of children with DSED or some mixed DSED & RAD.

1.3.1. Possible underpinnings of DSED.

Typically, selective attachments with primary caregivers tend to develop around 7-8 months old and in the general population around 30-40% of individuals are expected to present with insecure attachment, (Ainsworth, 1979), but this is not the same as the lack of stranger danger present in children with DSED. One of the first studies to describe children with DSED was a seminal study by Tizard and Rees (Tizard & Rees, 1975). Twenty-four children adopted from a British institution and a comparison group of home reared children (n=20) participated. The institutional environment offered improved physical care and toys but was emotionally neglectful; the matrons were encouraged not to form bonds with the children. The adopted children were described as exceptionally affectionate and friendlier towards strangers than the home-reared children, and about one third of adoptive parents reported overfriendliness, despite describing their children as being attached to them. 'Overfriendliness' was confirmed by observation. Significant research has since shown that indiscriminate behaviours are common among children adopted from severely deprived international institutions ranging in geography from Eastern Europe to South America, (Chisholm, 1998; Groark, McCall, & Fish, 2011; O'Connor & Rutter, 2000; Rutter et al., 2007; Smyke, Dumitrescu, & Zeanah, 2002; Zeanah, Smyke, & Dumitrescu, 2002). Although the pre-adoption environmental circumstances experienced by the post-institutionalised adoptees was extreme in nature, which limits the generalisability of the findings, the idea that DSED occurs in the context of maltreatment is further supported by findings with community samples of maltreated children.

Prevalence of DSED in community based populations: Oosterman and Schuengel, (2007) reported that of 60 children in the Netherlands in foster care, 7 children met criteria for DSED when assessed via the Disturbances of Attachment Interview and a further 2 had co-occurring DSED and RAD. Minnis et al, (2013) conducted a population study of 1646 children in one deprived UK urban area and reported prevalence of DSED and RAD combined to be around 1.4%, but this

figure included suspected and borderline cases. Twelve cases were diagnosed with DSED specifically, giving a prevalence of just less than 1% (0.72). One study measured DSED symptoms in 153 looked after adolescents in the UK, in comparison to a non-looked after sample, using the Development and Wellbeing Assessment and of 153 participants, 80 met criteria designated for 'caseness' of DSED (Kay and Green, 2013). Kay and Green (2016) found that of 60 children adopted from UK-out of home care, 49% demonstrated symptoms of DSED and Moran et al., (2023) found that 30% of 110 young adults (n=30) (16-23 years) in a youth offenders institute in the UK presented with DSED, while an additional 6 cases had symptoms of co-occurring DSED and RAD.

More recently there has been debate among researchers in relation to dimensions of maltreatment i.e., threat versus deprivation or harshness versus unpredictability, with some proposing models that suggest each 'type' of maltreatment may have a different measurable outcome regarding neurodevelopment (Ellis et al., 2022; Sheridan and McLaughlin, 2014). Threat is considered to impact the developing nervous system via rapid response fight/flight reactions (Sheridan and McLaughlin, 2014; Hiles Howard et al., 2020), whereas deprivation impacts the developing nervous system through lack of experience of adequate stimulation (Perry, 2001; Hiles Howard et al., 2020). Preliminary research suggests that deprivation may be associated with reduced cognitive ability, language skills, associative and implicit learning and impaired executive functioning skills, in addition to atypical patterns of cortical and white matter development which are areas of the brain associated with learning, (McLaughlin et al., 2017). Threat, in comparison, is associated with neurobiological changes such as activation of the amygdala and heightened limbic system and stress pathways (McLaughlin et al., 2017), although one systematic review suggested that maltreated children may demonstrate two types of post-maltreatment responses - blunted cardiovascular and sympathetic nervous system responses i.e., less of a stress response than might be typically expected during challenging situations, or hyper-responsiveness, as described above by McLaughlin et al, (Young Southward et al., 2020). It is interesting to note that deprivation, rather than threat, is associated with impaired cognitive skills such as executive functioning, because there is some evidence to suggest that poor inhibitory control is associated with both pre-school and school-age children with DSED in community samples as well as post-institutionalised adoptees (Bruce et al., 2009; Pears et al., 2010). Furthermore, poor inhibitory control is one of the core features of ADHD and evidence from both the ERA and BEIP longitudinal studies suggests that ADHD, symptoms, in particular, may frequently co-occur with DSED (Bos et al., 2011; Zeanah & Gleason, 2015; Kennedy et al., 2017). Studies conducted in Norway with moderate sized community samples (Seim et al., 2022) and in the UK with small clinically referred samples (Davidson et al., 2023) have also noted co-occurrence of DSED with ADHD. A recent systematic review also suggested that ADHD may, in some cases, be caused by

maltreatment, but in other cases, ADHD may exacerbate vulnerable circumstances where maltreatment occurs (Bali et al., 2023). For these reasons, further research regarding dimensional models of maltreatment could offer potential insights, but a major limitation is that DSED and RAD populations are generally not identified within the wider maltreatment and Adverse Childhood Experiences (ACES) literature. Currently, it is not yet clear whether these dimensional approaches to adversity, including maltreatment, help to elucidate our understanding of the underpinnings of DSED, or the relative impact of co-occurring neurodevelopmental conditions. The available evidence appears to suggest that DSED, or RAD, presents where social and emotional- as well as physical neglect- occur (Zeanah and Gleason, 2015; Oliveira et al., 2012), while the role of physical and emotional abuse is less clear (Zeanah and Gleason, 2015). However, one difficulty is that threat/abuse or deprivation/neglect are not always categorically distinct, often co-occur and/or lack of report or knowledge of one does not negate the presence of the other (Lacey and Minnis, 2020). The DSM-5 DSED criteria does refer to deprivation of care regarding DSED aetiology, but this may, in part, be because much of the early identification of DSED arose through studies of post-institutionalised children where severe early deprivation was identifiable; presence of abuse/threat cannot be excluded in these cases.

1.4. Concerns regarding the overlap between Autism and DSED

There is concern amongst experienced clinicians regarding Autism and DSED, (Davidson et al., 2015) because some of the diagnostic social interaction and communication difficulties associated with Autism, (DSM-5) have been found in post institutionalised adoptees with DSED (ERA) (Rutter et al., 1999) and in clinically referred community samples in the UK, (Sadiq et al., 2012; Davidson et al., 2023). Accurate diagnosis is essential for treatment and case management, but, additionally, adverse media regarding child protection was reported to have heightened the caution of even senior medical practitioners, (Dyer, 2010) because DSED is associated with maltreatment (DSM-5) and Autism is not (Turner et al., 2019). Concerns first arose following publication from the ERA studies that some of the institutionalised children appeared to present with Autism-like symptoms yet did not fully meet the diagnostic criteria for Autism. This phenomenon was described as Quasi-Autism, (Rutter et al., 1999). The following section outlines both the historical and current research evidence regarding the overlap between Autism and DSED and discusses the clinical conundrum in more detail.

Quasi-Autism was the term used to describe a sub-group of 11 post-institutionalised Romanian children adopted within the UK, (from ERA cohort) who when followed up presented with aberrant communication, poor social boundaries and indiscriminate behaviour, (Autistic traits) but whom, with the exception of 1, demonstrated greater social interest and flexibility, than would be typically expected in Autism. The children were assessed via the standardised caregiver report,

the Autism Diagnostic Interview-Revised (ADI-R) and at initial assessment, age 4 years, The ADI-R scores of the children with Quasi-Autism, and no cognitive impairment (n=8), were similar to the comparison group of Autistic children with no maltreatment history (n=14). However, by age 6 all the ADI-R scores in the Quasi-Autistic group were significantly lower than the comparison group. Many of the repetitive and stereotyped behaviours disappeared and significant catch up, including in language and other cognitive skills, was demonstrated. Of the 3 children with cognitive impairment, one child did have Childhood Autism, but Quasi-Autism was exemplified in the others by one child who used her sign language, Makaton, more spontaneously and flexibly than would be expected in Autism (Rutter et al, 1999). In a secondary study, still from the ERA cohort, the original Quasi-Autism group (no cognitive impairment) (n=10) plus children with milder Quasi-Autism symptoms (n=10) and children from the original maltreatment sample who met Autism criteria on the Social Communication Checklist (n= 8), were assessed via the ADI-R and the Autism Diagnostic Observational Schedule (ADOS). Findings suggested that 16 of these children had Quasi-Autism. Three of the 16 were excluded as they presented with additional intellectual disability, leaving 13 children who were then followed up at age 11-12 years. At follow up, 61.5 % (n=8) no longer met criteria for Autism (Rutter et al., 2007). Nevertheless, other social difficulties did persist for the 8 children who no longer appeared Autistic. These were socially indiscriminate behaviours such as poor relationship with the examiner, unsolicited physical contact and an excess of spontaneous comments. It later became apparent that for these children their difficulties might be better explained by DSED (Rutter et al., 2010).

The Quasi-Autism finding raised awareness and apprehension amongst clinicians because clinicians are required to make decisions based on the evidence in front of them, at a specific point in the child's development. Two years follow up is not typically an option when decisions regarding a child's clinical care are needed and definitely not if child protection concerns may be indicated. Clinicians do have the option to wait and see how development continues and then re-assess later i.e. 'watchful waiting,' if there are no immediate child protection concerns, but the problem with watchful waiting is that in most cases, and especially with Autism where environmental changes and supportive communicative strategies are essential, early intervention better supports later health outcomes (Powell and Gheera, 2021). It is for this reason that the National Institute for Health and Care Excellence (NICE) recommends that once behavioural symptoms of Autism have been recognised and accepted for referral, a decision should be made within 3 months (NICE, 2011& 2017). Furthermore, caregivers typically seek referral because they are in distress in that moment and receiving both an 'answer' and appropriate intervention support can reduce parental and child stress (Carter, 2005).

The findings by Sadiq et al., (2012) that pragmatic language difficulties overlapped in children with Autism (average verbal IQ) (n=52) and children with DSED^{RAD} (n=35), in a community sample of children in the UK, were also concerning. Impaired pragmatic language skills is considered a hallmark of Autism (Rhea, 2007). Both the DSED^{RAD} and Autism groups significantly differed from typically developing children (TD) (n=39) in the domains of inappropriate initiation, coherence, stereotyped conversation and social interests, on the well validated Child Communication Checklist, but only children with DSED^{RAD} significantly differed from the TD group regarding rapport. The DSED^{RAD} group also showed *greater* impairment than Autistic children regarding use of language in context, rapport and social relationships. The main limitations, however, were use of caregiver report only and overlapping Autism symptoms in the DSED group when assessed via standardised caregiver autism assessment. The authors acknowledge that they were unable to confirm presence or absence of co-occurring Autism with DSED without further observational assessment (Sadiq et al., 2012).

The following case example demonstrates an example of the mixture of symptoms which might leave clinicians wondering whether difficulties are attributable to Autism, DSED or both. The example is based on a DSED case that was referred to the research study by a CAMHS clinician.

1.4.1. Case study

I met Rosie at her mum's house. As I entered the living room, Rosie walked right up to me, a stranger to her, said hello and placed a kitten in my hand. Rosie is 8 years old and she smiled at me as she told me that her kitten was called Patch. I joked that I couldn't tell why the cat was named Patch and she took my hand and led me to the couch. Look, he's all patchy, she told me. I was there to complete an interview with her mum and as I introduced myself and the plan for the morning, Rosie's mum told her to play with her toys at the other side of the room. Rosie returned immediately and interrupted us. Her mum said that it was always like this. She even gave an example of Rosie telling people on the bus about herself. Every time Rosie spoke to me, she stood a bit too close to me and she often placed her hand on my knee. It felt over-familiar and unusual. Rosie was fidgety and her smile appeared to be a bit forced. I felt like she stared when she spoke with me. Rosie told me that her mum had given away her rabbit and she made a grimace face when I asked how that felt. During my conversation with her mum, Rosie interrupted to tell me pieces of information that were quite personal - her mum wouldn't let her dad see her anymore and her mum worried about money – but she also talked at length about her toys and she told me lots of information about her kitten. I took a developmental history with her mum and found out that Rosie met all her major milestones but has had some difficulties with learning at school. Her mum was most concerned about her 'odd' behaviour which she described as too friendly, pushy, controlling and she was worried that she couldn't keep friends once she made them. She was also

worried by movements that Rosie sometimes made in public with her hands and she felt that she was extremely clingy. Mum mentioned that social work were concerned about her ex-partner, Rosie's dad, and that she was not meant to be seeing him anymore, but she herself was not concerned about him.

This child's clinical presentation includes clear signs of autism such as poor social boundaries, unusual eye contact, unusual hand movements, interrupting, focused interests, friendship problems and difficulties at school. However, she also demonstrates features associated with DSED such as indiscriminate friendliness, and added to the child protection concerns mentioned, a diagnosis of DSED might thus be more likely. Given the importance of differential diagnosis for effectively supporting this child, clinicians clearly need further information regarding the differences between the two disorders, and in particular whether certain types of behavioural symptoms might distinguish them.

1.5. Differences between Autism and DSED.

The Coventry grid, (Moran, 2010) was probably the first response by clinicians, rather than researchers, to proactively react to the Autism-DSED^{RAD} clinical conundrum. The Coventry grid is a matrix of observations regarding the behaviours of children with Autism and children with DSED^{RAD}. It was originally designed by a group of experienced psychologists and describes, in their opinion, the overlaps and possible differences in behaviour between children with Autism compared to children with DSED^{RAD}. The Coventry Grid was published as a shared knowledge opinion piece. It is not an assessment tool, nor was it designed to be, but given the scarcity of research evidence available regarding differentiation between Autism and DSED, clinicians have been using it as guidance (Davidson, 2016, unpublished pre-doctoral qualitative investigation; Flackhill, James, Soppitt and Milton, 2017.) The Coventry Grid consists of 8 functional domains: 1. flexible thinking and behaviour, 2. play, 3. social interaction, 4. mind reading, 5. communication, 6. emotion regulation, 7. executive function and 8. sensory processing. Within each domain, the first column of the grid describes the ways in which children with Autism and children with DSED^{RAD} have been observed to behave similarly. The second column within each domain describes behaviours which are proposed to be 'typical' in Autism and the third, behaviours proposed as 'typical' of DSED^{RAD,} and which may differ from Autism. In the social interaction domain, for example, one sided approach, lack of interest in sharing and lack of awareness of one's role, among others, are described as behaviours indicative of Autism. In DSED^{RAD}, the child is thought to seek interaction but may preferentially choose adults over peers, or they may not share reciprocally but will show awareness that their behaviour is upsetting to others. Furthermore, a 'matter of fact' feel is described regarding the therapeutic relationships with children with Autism, compared to children with DSED^{RAD}. The latter are suggested to show some skill in building relationships, yet were, on occasions, inappropriate and challenged personal

boundaries. To give another example, in the communication domain, one described difference between children with Autism and children with DSED^{RAD} is lack of repair of conversation breakdown; children with Autism are suggested to have much greater difficulty maintaining conversational flow than children with DSED^{RAD} (Moran, 2010). Davidson, Moran and Minnis, (2022) examined the domains of the Coventry Grid and found that there is preliminary evidence to support *some* of the assertions. For example, Davidson et al., (2015) reported that of 58 children with Autism, and no maltreatment history, who had been referred to a UK based clinic for Autism assessment, 38% met core diagnostic criteria for DSED on the standardised caregiver report, the Reactive Attachment Disorder and Disinhibited Social Engagement Disorder Assessment Interview (RADA) (Lehmann et al, 2020). The parent-report RADA did not help to discriminate between Autism and DSED. However, in all but 4 cases, Autism was easily distinguishable from DSED via unstructured observation. The unstructured observation involved a 1:1 conversation between the child and the assessor during a juice break between cognitive tasks. Some of the identified Autism behaviours included, break-down in communication when the assessor did not scaffold the interaction, repetitive questioning, tangents about own interests and unusual prosody. With the exception of unusual prosody, the communication differences found by Davidson et al, in Autism compared to DSED are highlighted as core Autistic difficulties within the Coventry Grid.

Sadiq et al, (as described previously) found that caregivers reported similar pragmatic difficulties between children with Autism and children with DSED^{RAD} but more stereotyped and repetitive language and behaviour in children with Autism. The Coventry Grid authors also report greater stereotyped and repetitive language and behaviour in Autism compared to DSED^{RAD}. However, some caution is warranted in interpretation because repetitive or stereotyped language may be found in some children with specific language impairment (now known as Developmental Language Disorder), who do not have Autism, although difficulties do tend to be more pronounced in Autism (Guerts et al., 2008; Bishop et al., 2014). There are no current published investigations of the expressive language (rather than pragmatic) skills of children with DSED, but maltreated children, generally, are at higher risk of language problems, especially expressive language difficulties (Carr et al., 2020).

Both the Coventry Grid and the available research evidence lack consideration of the wider language abilities of children with DSED and compared to children with Autism. To further our understanding of whether differences between Autism and DSED may lie within social communication, we first need investigation of the receptive and expressive language skills of children with DSED, and in addition to consider their pragmatic language skills in the context of their language ability. As children who have been maltreated are at greater risk of expressive language problems, it is plausible that some of the pragmatic language difficulties reported by caregivers in the Sadiq et al study were impacted by expressive language difficulties rather than

only pragmatic impairment but this is unknown. Without further rigorous research, perhaps also using observational assessment in addition to caregiver report, and by involving experts such as Speech and Language Therapists (SLTs), the current evidence base regarding social communication differences is tentative.

Davidson et al, (2023), was the first study to explore differences between children with Autism and DSED using conversation and observational methods, alongside standardised assessment. All the children were noted to have free flowing language and ability to speak at sentence level (module 3 of the ADOS) but expressive language was not more thoroughly investigated here. In this study, it was possible to discriminate between children with Autism and DSED via the ADOS-2, except in the cases where ADHD overlapped with DSED or Autism overlapped with DSED. These cases were more clearly identified via the unstructured and conversation based observation. This study forms part of the body of this thesis, thus is described in full detail in chapter 4, but it is of relevance to note that the findings both support and contradict elements of the Coventry Grid. Further support for elements of the social interaction and communication domains of the Coventry Grid were found i.e. reduced use of nonverbal communication, limited understanding of humour/literal interpretation and idiomatic language were more apparent in the Autism group than the DSED group, but samples were small (n=10). The contradiction arises because Davidson et al found that the small group of children with DSED (n=8) were better able to understand and actively engage in banter, simple sarcasm and humour (relative to typically developing children) than children with Autism. Yet the Coventry grid suggests that in DSED^{RAD} *gentle teasing may provoke extreme distress (self-esteem seems to be too fragile to cope) – internalise/assume it is about them.* In part, the discrepancy may be related to a different measurement perspective as Davidson et al have examined the behaviour in terms of presence of skill whereas the Coventry Grid is framing the behaviour in terms of emotional reaction. It may also be that the lack of discrimination between DSED and RAD in the Coventry Grid has influenced perceptions as, it is now known, that it is RAD which tends to be associated with internalising behaviours rather than DSED (Gleason et al, 2011). Finally, both studies are based on observations from clinically referred samples, where presentations may vary and sample bias may present.

One other study has investigated differences between Autism and DSED. A sample of 506 children were assessed via a specialist Autism assessment clinic in the US by two experienced psychologists and 486 were diagnosed with Autism only (no maltreatment history), 7 children with DSED^{RAD} and 13 children with DSED^{RAD} + Autism. As part of the assessments, the Checklist for Autism Spectrum Disorder (CASD) screening questionnaire had been completed by caregivers (Mayes et al., 2017). The CASD total scores significantly differed between children with Autism only and children with DSED^{RAD}, but not between Autistic children and children

with DSED^{RAD}+ Autism. Of the 30 items which make up the CASD screening, 9 items were present only in the Autism group and the DSED+Autism group, suggesting that they may be more Autism specific. Seven of the 9 items presented within the domain restricted and repetitive behaviours and were as follows: restricted and obsessive interests; repetitive stereotyped play (e.g. lining up objects); stereotypies (e.g. hand flapping and spinning); craving movement (e.g. excessive running, jumping, and swinging); distress with crowds; fascination with repetitive movements (e.g. fans); picky eater (limited food preferences and/or hypersensitivity to food texture). The other two items were delayed speech milestones and unusual fears (e.g. elevators, tornadoes, and small spaces) (Mayes et al, 2017). Greater repetitive and restricted interests in Autism rather than DSED^{RAD} replicates the finding by Sadiq et al (2012) and the report within the Coventry Grid. Furthermore, both the Coventry Grid and Mayes et al identified hyposensitivity to food textures (picky eating) as a specific difference between the 2 groups. However, as Flackhill and colleagues (2017) highlight, the range of sensory difficulties experienced by children with Autism is vast but the range of sensory experiences considered by the Coventry Grid is actually very limited (Flackhill, James, Soppitt and Milton, 2017). Furthermore, caution is warranted as the generalisability of Mayes et al's findings are limited due to small sample size of the DSED^{RAD} and DSED^{RAD}+ASD groups, lack of standardised assessment of DSED and possible sample bias. Replication with additional samples of children with Autism and DSED are still required to test these findings.

There is preliminary evidence to suggest that differences between Autism and DSED are identifiable (Rutter et al., 1999; Davidson et al., 2015; Mayes et al., 2017; Davidson et al, 2023) and that some elements of the Coventry Grid are replicated by these studies (Davidson, Moran and Minnis, 2022), but it is clear that there is much still to learn. A cautionary approach overall to differential diagnosis of Autism from DSED is required, particularly with regards to the Coventry Grid; best practice guidelines of good clinical judgement and holistic multi-informant assessment is imperative (Flackhill, James, Soppitt and Milton, 2017; Davidson et al, 2023).

1.5.1 Complexity

While the overlap in core symptoms between Autism and DSED appear concerning, in 'straightforward cases' discrimination is possible with standardised Autism observation tools (Davidson et al, 2023). The cases which are more likely to cause concern are cases where the child's developmental history may be unclear, symptoms may be subtle, child protection concerns may have already been raised by social work or neurodevelopmental conditions may overlap (Davidson et al., 2015; Gajwani and Minnis, 2023; Davidson et al, 2023). For example, in the Davidson et al, (2015) study described above of the 4 children with Autism who met diagnostic criteria for DSED but whose symptoms were not easily identified as clearly Autistic in nature, all met Wing's 'active but odd' criteria – socially motivated but nevertheless with core social and

communication difficulties. Also, one was a girl with more subtle Autism symptoms and co-existing ADHD was present in some cases. (Davidson et al., 2015). The latter is probably of most concern because co-morbidity is considered the norm not the exception for both children with Autism (Gillberg, 2010), and children with DSED (Minnis, 2013; Dinkler et al., 2017). Furthermore, problems such as aggression, anxiety, hyperactivity, behavioural problems or social relationships difficulties may be more likely to result in a referral to CAMHS than core symptoms of Autism, (Byrne, 2003; Kantzer, Fernell, Gillberg, & Miniscalco, 2013). Clinicians concerns regarding some 'hard to assess' cases being misdiagnosed are legitimate.

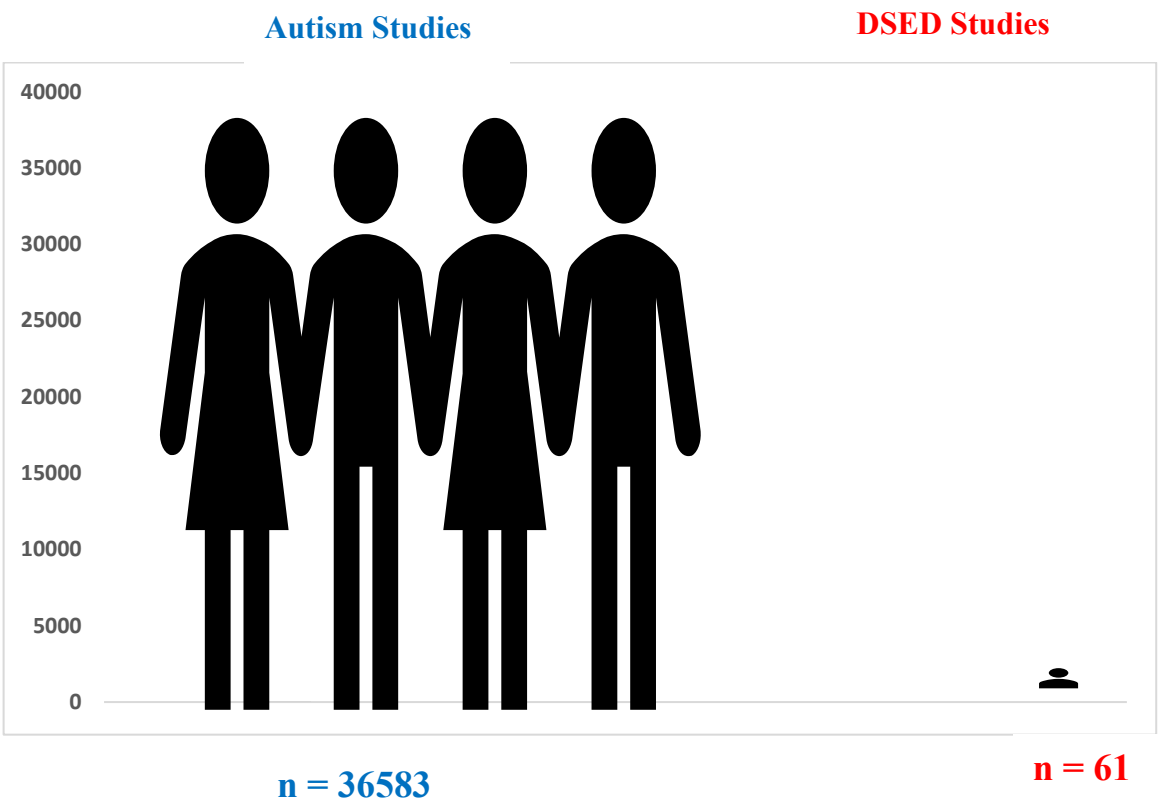
As neurodevelopmental overlap is complex but not at all uncommon, Gillberg (2010) strongly advocated for a new approach to assessment, using a framework coined ESSENCE -Early Symptomatic Syndromes Eliciting Neurodevelopmental Clinical Examination. An ESSENCE approach recognises that children presenting in clinic before ages 3-5 years with impairment in any of the following markers: general development, communication, social problems, motor-coordination, attention, activity, behavioural, mood or sleep are likely to present with one or more lifelong neurodevelopmental conditions. Therefore, it emphasises assessing broadly, and jointly by multi-disciplinary professionals, rather than the current practice of diagnosis specific assessment, which can see families referred from pillar to post, with one referral diagnosing Autism, for example, and a later referral for communication problems that may end up in a third referral due to severe attentional and hyperactivity. Instead clinicians are encouraged to expect that the ESSENCE markers may be suggestive of one or more of Autism, ADHD, Oppositional Defiant Disorder, Specific Language Impairment (now called Developmental Language Disorder (DLD) (DSM-5)), ID, Tics/Tourette's Syndrome, or DSED^{RAD}, (Gillberg, 2013; Miniscalso et al., 2006; Fernell and Gillberg, 2023) and that early symptoms may change or develop over time and therefore re-evaluation is necessary (Fernell and Gillberg, 2023). The goal is not to overly label the child, but to avoid lack of recognition of difficulties which tend to occur together, so that each can be treated appropriately and in a timely manner.

1.6. Where are the gaps for research on discriminating between Autism and DSED?

The existing literature appears to suggest that the biggest gaps in knowledge exist with regards our knowledge of DSED; DSED is rarely identified within the wider maltreatment literature, and the few studies that have investigated DSED have largely focused on prevalence, longitudinal follow up of institutionalised children or identifying overlaps/differences between Autism and DSED. This means that specific information about the skills and behaviours of children with DSED is missing for clinicians. To exemplify the extent of the scarcity of research on DSED, in comparison to Autism, the total number of studies registered with the database pub-med since the advent of DSM-5 until the time of writing was investigated and are demonstrated in figure 1.

Although the DSM-5 was published in May 2013, the time frame searched was 2014-2023 to allow for any delay between study completion and publication and to ensure that any studies pre-DSM-5 were not included.

Figure 1: Total number of studies regarding Autism versus total number of studies regarding DSED in pub-med (2014-2023)



This gap in knowledge regarding DSED exacerbates any uncertainty that clinicians may feel when trying to differentiate Autism from DSED, as they have less knowledge regarding what interaction or communication difficulties to expect in this group. Consequently, paper 1 is a systematic review of the social functioning of children with DSED.

The second gap regards which tools best help to differentiate between Autism and DSED. While the few studies available appear to suggest that unstructured observation is a useful tool, current clinical Autism assessment tends to rely on standardised assessment measures which are structured and manualised (see chapter 3 for more details). No previous studies have directly compared the current gold standard Autism assessments with unstructured observation in the context of differential diagnosis of Autism from DSED, therefore this is addressed by paper 2.

There is a further gap in the literature with regards language and communication skills of children with DSED. Again, DSED is not generally identified within maltreated samples in the

communication literature, but there is evidence to suggest that maltreated children are at greater risk of language problems, particularly expressive language difficulties (Carr et al., 2020) and pragmatic language impairment (McCool & Stevens, 2011; Ciolino et al., 2021). These findings appear to support the outcome of Sadiq et al, that children with DSED present with pragmatic language problems, similar to children with Autism, but the relative impact of expressive language difficulties is unknown. Other studies suggest that skill differences between children with Autism and DSED may be found within the social communication domain (Moran, 2010; Davidson et al., 2015; Davidson et al., 2023). Therefore, paper 3 addresses the gap in literature by investigating the receptive, expressive and pragmatic skills of children with Autism and children with DSED, compared to typically developing children via multi-informant assessment. It also explores these domains in detail via SLT analysis to determine if there are any patterns of differentiation.

Finally, there is preliminary evidence that repetitive and restricted behaviours may differentiate between Autism and DSED (Mayes et al.,2017), and there is perhaps the most evidence available within this domain (Davidson et al., 2022). However, sensory processing is now included within the DSM-5 Autism diagnostic criteria, yet this is an area largely un-investigated, both within literature investigating DSED and in the reviewed studies regarding differential diagnosis from Autism. This is despite our understanding that both post-institutionalised children and community samples of maltreated children demonstrated broad sensory problems, including sensory avoidance, auditory problems which are particularly associated with Autism (Greenspan and Weider, 1993; Gillberg and Coleman, 1996; Rogers et al., 2003; Tomchek et al., 2007; Schoen et al., 2009) and sensory seeking behaviours (Cermak & Daunhauer, 1997; Cermak and Groza, 1998; Atchison, 2017). Paper 4 meets this literature gap and provides the first comparison of the sensory profiles of children with Autism compared to children with DSED, and in comparison to the DSM-5 ASD sensory processing criteria.

Aim: The overall aim of the thesis was to explore the profiles of a case study sample of children with Autism and DSED in the context of differential diagnosis of Autism from DSED. Four research question were identified to address the gaps in knowledge and support the overall aim of the thesis. There were as follows:

1. What are the broad social functioning difficulties, if any, of children with DSED? (Paper 1).
2. Do current multi-informant 'gold standard' Autism diagnostic tools support differential diagnosis of Autism from DSED, and how does the structured ADOS-2 assessment compare to an unstructured behavioural observation, in this context? (paper 2)

3. What is the profile of children with DSED regarding receptive, expressive and pragmatic language, and can any differences be identified compared to children with Autism? (paper 3)

4. What are the sensory processing profiles of children with DSED, compared to children with Autism?

1.7. References

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Chapter 2: Social competencies of children with Disinhibited Social Engagement Disorder: A systematic review

Paper 1 is a systematic review of the literature regarding the social functioning of children with DSED. The aim was to determine whether children with DSED present with social difficulties as this will improve our knowledge as to whether social relationships difficulties associated with Autism overlap with DSED, or not.

Paper 1:

Davidson, C., Islam, S., Gillberg, C., Lowit, A., Venturini, E & Minnis. H. (2023) Social competencies of children with Disinhibited Social Engagement Disorder: A systematic review. *Journal of Child Psychology and Psychiatry Advances,* e12226. https://doi.org/10.1002/jcv2.12226

Received: 6 July 2023 Accepted: 15 December 2023

DOI: 10.1002/jcv2.12226

RESEARCH REVIEW

JCPP Advances

Social competencies of children with disinhibited social engagement disorder: A systematic review

Claire Davidson[1] | Shahela Islam[1] | Enrico Venturini[1] | Anja Lowit[2] | Christopher Gillberg[3] | Helen Minnis[1]

[1]University of Glasgow, Academic CAMHS, West Glasgow Ambulatory Care Hospital, Glasgow, UK

[2]University of Strathclyde, Speech and Language Therapy, Glasgow, UK

[3]Gillberg Neuropsychiatry Service, University of Gothenburg, Gothenburg, Sweden

Correspondence

Claire Davidson, University of Glasgow, Academic CAMHS, West Glasgow Ambulatory Care Hospital, Glasgow, G3 8SJ, UK.
Email: claire.davidson@glasgow.ac.uk

Funding information

Gillberg Neuropsychiatry Centre; NHS Greater Glasgow and Clyde; Castang Foundation, Grant/Award Number: LAY1-WS_LEGAL.FID1730709

Abstract

Background: Children with Disinhibited Social Engagement Disorder (DSED) have specific difficulties with indiscriminate sociability, yet little is known about their broader social competencies as DSED tends not to be identified within samples in the wider 'maltreatment literature.'

Aim: To systematically review the literature to determine the social competencies of children with DSED.

Methods: A comprehensive search following PRISMA guidelines was undertaken using PsycINFO, Medline, Embase, and Cumulative Index to Nursing & Allied Health. **Results:** From a total of 553 articles, 16 studies were selected and critically evaluated. Children with DSED were consistently reported to have poorer social competencies than non-maltreated peers and environmental controls. Greater peer problems were consistently found, and they may present with poor self-esteem/ concept related to social acceptance. Findings regarding social interaction/ communication skills were mixed.

Limitations: 50% of studies were of moderate quality due to sampling and possible confounding variables.

Conclusion: Children with DSED present with social relationship problems, beyond the core symptoms of the disorder, but the relative impact of co-occurring neuro-developmental conditions is not yet clear. In addition, pragmatic language and communication skills require further research.

INTRODUCTION

Childhood maltreatment (abuse or neglect) is associated with social relationship difficulties and/or poorer quality relationships across the lifespan (Doyle & Cicchetti, 2017; Flynn et al., 2014; Goemans et al., 2023; Shonk & Cicchetti, 2001). Yet the mechanisms under-pinning such problems are complex. Attachment relationships play a crucial role in the early development of social relationships through the dyadic process of infant signalling and parental sensitivity to child behaviours (Ainsworth et al., 1974; Bowlby, 1982, 1988). This 'serve and return' like interaction helps build neural pathways in the developing brain (Ilyka et al., 2021), and supports development of early skills such as joint attention, which are important for later language and reciprocal social interaction (Bottema-Beutel, 2016; Carpenter et al., 1998; Markus et al., 2000).

As a multi-faceted concept social functioning is difficult to measure but in childhood, social competency tends to relate to peer acceptance/rejection, pro-social skills/skill deficits, self-regulation and ability to navigate social conflict (John, 2001).

Children who have experienced early maltreatment, however, are more susceptible to developing insecure and disorganised attachments (Bowlby, 1973, 1982; Pickreign Stronach, et al., 2011), or attachment disorders (Minnis, 2013; Zeanah et al., 2016). Social skill deficits such as poor play and joint attention, language delay, poorer identification of non-verbal cues and deficits in facial recognition of emotions have been reported (Carr et al., 2020; Culp et al., 1991; Law & Conway, 1992; Sheaffer, Golden, & Averett, 2009; Sheaffer, Golden, & Averett, 2009). Greater peer conflicts, bullying or victimisation are associated with maltreatment (Yoon et al., 2021; Goemans et al., 2023; Humphrey's et al., 2018; Guyon-Harris, Humphreys, Fox, et al., 2019), as well as lower self-esteem (Cederbaum et al., 2020; Seim et al., 2022), risk of mental health problems and risky or 'problem' behaviour (Carr et al., 2020; Humphrey's et al., 2018).

McCrory et al. (2022) hypothesised that maltreated children may be more susceptible to cumulative stress because of factors discussed above, and, additionally, because social networks of maltreated children may diminish due to poorer social competencies and missed opportunities to build social relationships. Gajwani and Min- nis (2023) argue that an important element may be the interaction of co-occurring neurodevelopmental conditions, as maltreated children are at higher risk of presenting with one or more neurodevelopmental conditions and/or maltreatment associated disorders (Dinkler et al., 2022; Minnis, 2013).

We are particularly interested in the social competencies of children with the maltreatment-associated disorder, DSED, as these children have specific relational problems, inherent to the diagnosis of the disorder. The core symptoms of DSED are indiscriminate sociability and poor social boundaries, which occur in the context of maltreatment (DSM-5, American Psychiatric Association, 2013). The term DSED is a relatively recent change to the nomencla- ture, which occurred with the advent of DSM-5. Within previous diagnostic classifications, DSED was known as the disinhibited sub- type of Reactive Attachment Disorder (d-RAD) (DSM-IV, American Psychiatric Association, 2000), or as Disinhibited Attachment Dis-order (DAD) in the European equivalent, ICD-10 (World Health Organisation, 1993). At that time, the presumed aetiology and lack of preferential selection of primary caregivers suggested that DSED may be a disorder of attachment. This changed as a body of evidence demonstrated that core features of DSED persisted, despite children developing secure attachments once placed with foster/adoptive families (Lyons-Ruth et al., 2009; Minnis et al., 2007; O'Connor et al., 2003; Zeanah et al., 2005). In DSM-5, the name DSED was introduced to better reflect the core problems of social-relatedness and DSED is now a *separate* disorder to Reactive Attachment Disorder, (DSM-5) (N.B. despite change of name, the core *symptoms* of DSED have not changed and remain as described under previous nomenclature).

Overview of Disinhibited Social Engagement Disorder

Disinhibited Social Engagement Disorder behaviours were first re-ported among children adopted from severely deprived international institutions (Chisholm, 1998; O'Connor et al., 2000; Rutter et al., 2007; Smyke et al., 2002; Tizard & Rees, 1975; Zeanah et al., 2002), and the Bucharest Early Intervention Project (Smyke et al., 2009, 2012) and the English and Romanian Adoptees Study (O'Connor et al., 2000; Rutter et al., 2007; Sonuga-Barke et al., 2017) were inspirational in demonstrating the childrens' needs, the possi-bility of positive developmental growth, and set the scene for better understanding of DSED.

Disinhibited Social Engagement Disorder has since been reported in community samples of maltreated children (Kay et al., 2016; Kay & Green, 2013; Minnis et al., 2013; Scheper et al., 2019; Seim et al., 2021). In one population study of 1646 children in a deprived UK urban area, 12 cases were diagnosed with DSED, suggesting an estimated preva-lence of just less than 1% (0.72) (Minnis et al., 2013). Scheper et al. (2019) found that in a community sample of 124 children, 38% (n = 47) presented with DSED and symptoms were still present 4 years later in 57% of those children. Of note, when associations with neu-rodevelopmental conditions and environmental factors were investi-gated, Attention-deficit/hyperactivity Disorder, (ADHD) was the only variable associated with persistence of DSED. This latter finding is interesting, given the recent preliminary research which suggests that different dimensions of maltreatment that is, threat versus deprivation may have different effects on neurodevelopmental outcomes (Ellis et al., 2022; Sheridan & McLaughlin, 2014). Disinhibited Social Engagement Disorder is thought to be associated with severe neglect, (social-emotional, in particular) (Zeanah & Gleason, 2015; DSM-5, 2013; Oliveira et al., 2012) and there is some evidence to suggest that severe deprivation, as opposed to threat, is associated with changes in cortical and white matter in the brain, reduced cognitive ability and negative effects on executive functioning (McLaughlin et al., 2017). While, DSED has been found to overlap with other neuro- developmental conditions, such as Autism (Dinkler et al., 2017; Mayes et al., 2017; Rutter et al., 1999), it is ADHD, or symptoms of ADHD, which appear more prevalent (Bruce et al., 2009; Pears et al., 2010; Bos et al., 2011; Seim et al., 2022). Even in early adulthood, when both Autism and ADHD were found to co-occur with persistent DSED, it was the interplay with ADHD which was associated with poorer functional outcomes (Kennedy et al., 2017)

Key points

- What's known:

Maltreated children may be more susceptible to social relationship problems, but Disinhibited Social Engagement Disorder (DSED) is often not identified within these samples, despite maltreatment being considered part of the disorder's aetiology.

- What's new:

Children with DSED demonstrate poorer social competencies and greater relational conflicts than typically developing peers, and in some cases, environmental controls. Possible pragmatic language deficits also require further investigation.

- What's relevant:

All professionals working with maltreated children should consider assessment for DSED *and* consider impact of impaired social functioning, especially as DSED is persistent over time. Co-occurring neurodevelopmental conditions are also frequently reported and requires further research regarding social functioning and later outcomes.

Nevertheless, the scarcity of research with children with DSED is still a concern (Zeanah et al., 2016). In other social impairment disorders, such as Autism, our wealth of knowledge (Carter et al., 2005) gives parents and clinicians better understanding about the difficulties that children experience. Such knowledge is crucial in assessment and case management, in reducing stress, supporting relationships and school functioning. Furthermore, there is ongoing concern regarding differential diagnosis of DSED from Autism (Davidson et al., 2015; Davidson, Minnis and Moran, 2022; Mayes et al., 2017; Moran, 2010), yet lack of knowledge about DSED and broader social problems makes it *even* more difficult for clinicians to untangle possible overlaps.

METHODS

Aims & research question

A scoping search of the literature revealed no synthesis of data regarding the social relationships of children with DSED. To address the gap in knowledge, we aimed to systematically review the literature to assess the social competencies (interpersonal relations, social skills, conflicts and perceptions of self) of children with DSED. We proposed the following research question:

1. Do children with DSED demonstrate impaired social competencies, beyond the core problem of indiscriminate behaviours?

Search strategy

Following the Preferred Method of Reporting of Systematic Reviews guidelines (PRISMA) (Liberati et al., 2009) four electronic databases were searched: PsycINFO (1872-present), Embase (Ovid, 1947-present update daily), Cumulative Index to Nursing & Allied Health (1973-present) and Medline (Ovid, 1946 to January Week 3 2017 & OVID 1946 to March 28, 2023). Studies were limited to English.
The search shown below exemplifies the search strategy:
Example phase 1 search of PsycINFO using subject headings and key words.

1. DSED (major concept) OR Attachment Disorder (explode).
2. Key words: Disinhibited Social Engagement OR DSED OR Attachment Disorder OR Indiscriminate friendl* OR Overfriendl* OR Over friendl* OR Indiscriminate Sociability
3. Combine 1 & 2 using "AND."
4. Social competence (major concept) OR Interpersonal relationships (major concept) OR relationship quality (major concept) OR interpersonal interaction OR social adjustment (major concept), OR social interaction (major concept), OR social skills (major concept) or social communication (major concept),
5. Social relationship* OR social interaction OR interpersonal relations* OR social skills OR interpersonal skills OR interpersonal interactions OR social communication, OR interpersonal communication OR pragmatic language.
6. Combine 4 & 5 using "AND."
7. Combine 3&6

Phase 2: We examined DSED synonym keyword searches individually by title and abstract, as some seminal studies have a broader focus for example, prognosis post-institutionalisation, yet still have relevance. Findings were appraised by title and abstracts. Selected studies were read in full. CD and SI reviewed a third of abstracts jointly to calibrate the process then both individuals reviewed half each of the remaining articles. Uncertainties were discussed at conference until agreement was reached.

Inclusion and Exclusion Criteria

Inclusion Criteria:

1. Study sample included children up to 18 years with a diagnosis of either DSED, RAD (DSM-IV) or dRAD (DSM-IV), or DAD (ICD-10) or core symptoms that is, indiscriminate friendliness with strangers in the context of maltreatment.
2. Paper discussed social competencies or impact on social relationships, social skills (verbal/nonverbal) or concepts such as self-esteem in relation to social functioning.
3. Studies used a standardised tool, observation or qualitative methods.

Exclusion Criteria:

1. Sample included maltreated children but did not identify DSED symptoms.
2. The study was about attachment patterns that is, secure/insecure attachments.
3. Thesis abstract, case study only or review.
4. Not available in English.

Due to the change in nomenclature, it was necessary to include studies in which the population was defined using the previous DSM- IV terminology (RAD, disinhibited RAD, indiscriminate friendliness)/ ICD-10 (Disinhibited Attachment Disorder (DAD)). However, this has no impact on the integrity of the results as the key symptoms of the disorder did not change with the DSM-5 re-classification. In some cases, the authors did not directly discriminate between sub-types of RAD (DSM-IV), but these studies were not excluded to ensure relevant data was not missed. This is methodologically justified for two reasons, 1. RAD (DSM-5) (the inhibited form) on its own has been demonstrated to be rare in the population (Minnis et al., 2013; Zeanah, 2000); it is DSED that is persistent, 2. RAD is a separate disorder to DSED (Gleason et al., 2011; Zeanah & Gleason, 2015;

with separate symptomology, despite its shared aetiology. Thus, if RAD symptoms were present, these would be considered as an additional but co-existing problem. In discussion of the results, we have used the term DSED^{RAD} to identify these older studies which likely contain mainly DSED cases but may include some cases of RAD. Otherwise, the use of terms DSED and RAD refer to the current DSM-5 diagnostic criteria. The Crowe Critical Appraisal Tool, V1.4 (Crowe et al., 2011) was used to rate study quality because it can be applied to both quantitative and qualitative methodologies. A score of <20 is considered low quality, 20-29 moderate quality, and 30-40 high quality. CD, SI and HM rated a third of the included full text articles independently and discussed findings jointly.

RESULTS

A total of 553 abstracts were identified, 496 removed and 57 studies read in full, of which, 41 were ineligible (see Figure 1).

Sixteen studies were included (50% of high quality and 50% of moderate quality) (see Table 1). Two studies were included where the maximum age was 2 years above the maximum inclusion criteria (18 years) but due to the scarcity of research in DSED, this was justified. Due to heterogeneity of study methods, we present the findings as a narrative synthesis. The four emergent sub-themes were: 1. social competence (general); 2. peer relationships, 3. self-esteem/ self-concept related to social functioning and 4. social interaction/ communication skills.

Social competence (general)

Two studies of moderate quality, investigated differences in total scores on standardised measures of problem and pro-social behaviour. Children with DSED^{RAD} scored significantly more poorly regarding social behaviours, (conflicts and pro-social) than typically developing children (Millward et al., 2006; Pritchett et al., 2013). In both of these studies, the authors did not discriminate between DSED and RAD and Pritchett et al., recognise that as UK norms were not available the study is limited by comparison to normative data of American children.

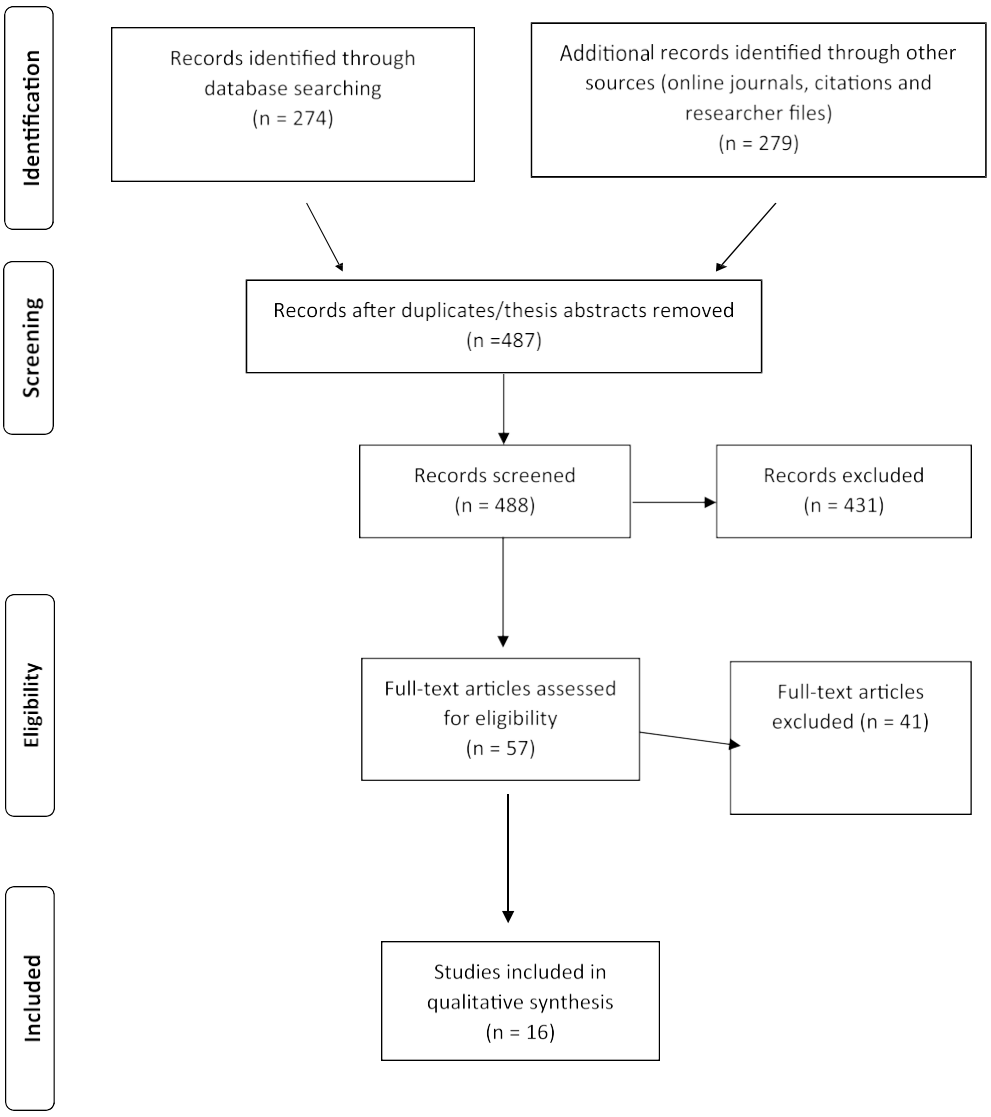

FIGURE 1 Preferred Method of Reporting of Systematic Reviews guidelines (PRISMA) flow diagram.

Another moderate quality study found that children with DSED who had mild intellectual difficulties performed significantly worse on the socialisation domain of the Vineland assessment compared to intellectually similar controls (Giltaij et al., 2016). The comparison group of children with similar intelligence quotient (IQ), but without DSED, is a relative strength, but the sample size of children with DSED (n = 7) was small and the authors recognised that some had mixed DSED and RAD.

In the final study, social competence was measured in 136 post-institutionalised children at age 12 years. To meet threshold for social competency the child had to be competent in 6 of the following 7 domains: family relationships, peer relationships, physical health, mental health, academic performance, substance misuse and risky behaviour. Overall, children with symptoms of DSED were significantly less socially competent than those without (Guyon-Harris, Humphreys, Fox, et al., 2019). When DSED symptoms were measured dimensionally ('never'-no symptoms, 'early'- symptoms before 54 months, 'late'- symptoms at 12 years and 'persistent'- symptoms before 54 months and at 12 years), 57% of the 'never' group met competency threshold, compared to just 28% in the 'early group, '33%' in the late group and of most note, 0% in the 'persistent' group. This was a high quality study, which expanded on measurements of social functioning in DSED, and adds weight to the consistent reports of this theme; children with DSED appear to have poorer general social competence than typically developing peers.

Peer relationships

A high-quality qualitative study by Bennett et al. (2009) utilised rigorous Interpretative Phenomenological Analysis following a story task with a strong sample of eight indiscriminately friendly children, who had experienced childhood maltreatment. The inclusion of opinions of children with DSED was a strength, and demonstrated, lack of understanding about friendships, feelings of social exclusion and a perceived need for acceptance.

Kay and Green (2013) investigated DSED in a high-quality case-control study of a non-institutionalised community sample of 153 high risk adolescents. Eighty-nine met 'caseness' via a standardised measure, for what they termed Disinhibited Indiscriminate Symp- toms. On the Health of Nations Outcome Scales, which was double- rated blindly, the Disinhibited Indiscriminate Symptoms factor made an independent prediction of greater peer problems.

Raaska et al. (2012) included a large, sequentially sampled group of 364 adopted children with DSED^{RAD} compared to large-scale register data. Twenty percent of children with DSED^{RAD} experienced victimisation, 8% bullied others and both bullying and victimisation were present independent of learning and language skills. Lack of social skills was also associated with victimisation. The large sample and consideration of possible confounding factors were strengths but due to lack of discrimination between DSED and RAD, it must be considered moderate quality.

Seim et al. (2022) also examined victimisation, bullying and aggression in a reasonable foster care home sample, (n = 31) and the findings support those of Raaska et al. This was a study of high quality which separately identified DSED and RAD.

A second high quality study by Guyon-Harris, Humphreys, Fox, et al. (2019) compared a sample of post institutionalised foster care children (n = 55) to a sample of post-institutionalised children in care as usual (n = 55) and a group of children from the local community (n = 50). Although symptoms of both DSED and RAD were assessed, it was not clear the total number of children who met criteria for DSED. Bearing this in mind, symptoms of DSED, and not RAD, were associated with greater caregiver perceptions of victimisation (rejected/bullied), and children with DSED were perceived to have greater conflicts in peer relationships. There was no significant association between symptoms of DSED, victimisation and the teachers' perceptions.

Self esteem/self concept in relation to social functioning

A case-control study on self-concept, (worthiness as a person and acceptance by peers), found that in 33 school-aged children with Disinhibited RAD, their perceptions of self-concept were higher than typically developing peers (n = 101) (Vervoort et al., 2014). This study was considered of moderate quality due to possible sampling bias and possible confounding variables such as co-occurring neurodevelopmental conditions.

In contrast, a high-quality study by Vacaru et al. (2018), found that DSED and RAD was associated with poorer self-concept, (cognitive competence, physical competence and peer acceptance) in a reasonable sized sample of post-institutionalised children (n = 33). However, self-perception ratings of physical competence were greater than the teacher ratings. The main limitation was that results were not discussed as to how they relate to DSED and RAD individually.

A high-quality study by Seim et al. (2021) also found that, in a community sample, children with DSED, (n = 26) demonstrated lower self-esteem regarding social acceptance compared to children with RAD (n = 28), environmental controls, (n=) and typically developing children (stratified sample of 10,480 school children). The authors acknowledge that the lack of standardised measures of DSED for the age group was a limitation but used as close to age measures as possible and then stringently applied DSM-5 diagnostic criteria.

Both high quality studies suggest that self-esteem regarding social acceptance generally may be lower in children with DSED, but Vacaru and Vervoort's studies together perhaps suggest that in specific circumstances, children with DSED may perceive themselves as more competent than significant others see them.

Social interaction skills

Groark et al. (2011) measured dyadic interaction between caregivers and indiscriminately friendly children (n = 123) in an institution using

TABLE 1 Study details.

Authors/Date	Study design	Investigative focus of study	Participants (*n*, age range, recruitment)	Measures (related to social functioning only	Outcomes (pertaining to social relationships/social function only)	CCAT score
1. Social competence						
Millward et al.	Quantitative cross sectional	Investigate symptoms of DSED[RAD] in children and related outcomes	82 families of children in foster care, residential care or special education schools, (age range 4- 16 years). Control group: 125 families from local nurseries and schools and 231 families recruited from local general practices, within a similar socio-economic status, (age range 5-16 years), matched on age and gender. DSED[RAD] symptomology established via the standardised reactive attachment disorders questionnaire (caregiver report).	Strengths and difficulties questionnaire (SDQ) via parent report.	Children with DSED[RAD] symptoms were likely to score highly on all sub-scales of the SDQ and significantly more poorly than controls.	29 Moderate
Pritchett et al. (2013)	Quantitative	To describe the characteristic of children with DSED[RAD]	22 children (6-8 years old) with definite or suspected DSED[RAD] from population screening of 1600 children. Symptoms of DSED[RAD] were identified via triangulation of standardised measures of caregiver report (relationships problems questionnaire, child and adolescent psychiatric assessment -RAD and development and wellbeing assessment) and child observation (waiting room observation). Diagnoses were made using DSM-IV criteria by clinicians in the research team.	Strengths and difficulties questionnaire (SDQ), completed by care givers and teachers. Social skills improvement system which assesses social skills, problem behaviours and academic competence.	SDQ: 75% of children with definite or suspected DSED[RAD] scored within the abnormal range. SSIS: 10/22 children scored below average, compared to American norms.	28 Moderate
Giltaij et al. (2016) cross sectional	Quantitative	Tested whether children with RAD or DSED had lower adaptive functioning (which included socialisation) than peers without RAD or DSED.	Total sample of 55 children with intellectual disabilities, mean age 10 years, mean IQ 72. 1/55 had DSED symptoms; 6/55 children mixed RAD/DSED. Comparison group, *n* = 45/55	Vineland screener 0-12. Developmental Behaviour Checklist	Vineland: DSED and mixed DSED/RAD scored significantly lower than peers for socialisation DBC parent: Mixed DSED/RAD & DSED only group more disruptive and antii-social behaviour than peers. DBC Teacher: more emotional disturbance in DSED	28 Moderate

TABLE 1 (Continued)

Authors/Date	Study design	Investigative focus of study	Participants (*n*, age range, recruitment)	Measures (related to social functioning only	Outcomes (pertaining to social relationships/social function only)	CCAT score
Guyon-Harris, Humphreys, Fox, et al. (2019)	Quantitative, prospective cohort study	Investigated the association between symptoms of DSED in early childhood and social competency in adolescence, across multiple domains. Caregiver reports examined at 4 time points (30, 42 and 54 months and 12 years).	DSED & RAD symptoms were based on the list of behavioural signs of disturbed attachment in young children following observation of parent-child interactions. 136 Romanian children from the Bucharest Early Intervention Project (BEIP).	The authors created a composite of competent functioning based on 7 domains (family relationships, peer relationships, academic performance, physical health, mental health, substance use and risky behaviour); information was gathered via items from the following standardised measures: The social skills rating system. DSED symptoms were investigated via the standardised disturbances of attachment interview at all 4 time points, and stranger at the door observation (age 54 months only) Youth risk behaviour survey. MacArthur health and behaviour questionnaire (HBQ).	Children with *more* symptoms of DSED were significantly less likely to meet threshold for social competency at age 12 years. Children who received diagnosis of DSED (before 54 months) were significantly less likely to be classified as socially competent. Social competence at 12 years: 'Never' group (no DSED symptoms at any time): 58% were socially competent. 'Early' (DSED diagnosis before 54 months): 28% of children were socially competent. 'Late' group (DSED symptoms at 12 years only): 33% of children were socially competent. 'Persistent' group (DSED symptoms at all time points): 0% were socially competent.	34 High
2. Peer relationships						
Bennet et al. (2009)	Qualitative	Social experiences of disinhibited children.	8 indiscriminate friendly children (aged 9–14 years, mean 11.5 years), with suspected/ confirmed maltreatment history, recruited from clinical services and voluntary organisation for adoptive parents. DSED symptoms were assessed using the standardised relationship problems questionnaire.	Semi-structured interview with indiscriminate friendliness social scenarios (IPA analysis).	IPA themes: 1. difficulty with concepts of friendships. 2. Exclusion from peer friendships. 3. Need for trust in relationships. 4. parental attempts to instil stranger danger and 5. Kindness as a response from others (seeking kindness and acceptance).	32 High

TABLE 1 (Continued)

Authors/Date	Study design	Investigative focus of study	Participants (*n*, age range, recruitment)	Measures (related to social functioning only	Outcomes (pertaining to social relationships/social function only)	CCAT score
Raaska et al. (2012)	Quantitative cross sectional	Bullying or victimisation in DSED[RAD]	364 international adoptees in Finland, ages 9-15 years (mean, 11.6 years) and comparison data of 146,767 children was derived from a large data set from Finnish schools.	Five to fifteen questionnaire	Children with mild DSED[RAD] more likely to report victimisation. Children with severe DSED[RAD] more likely to be both victims and bullies.	29 moderate
			Assessments conducted via postal survey (response-rate 49.4%) Symptoms of DSED[RAD] were measured via FINADO questionnaire, designed for use within the study.	Olweus bully/Victim questionnaire (OBVQ).	Lack of social skills was associated with victimization but not independently from bullying.	
Kay and Green (2013)	Quantitative cross sectional	Assess DSED behaviours and associated functional impairment. in non-institutionalised adolescents exposed to early maltreatment or neglect.	153 adolescents, at high risk of placement breakdown, referred by social workers, were assessed. The mean age was 174 months.	Health of the nation outcome scales for children and adolescents (HoNOSCA).	High risk group: Demonstrated significantly higher total and scale DAWBA-RAD scores than the low risk group.	34 High
			Control group was a low risk community sample in a deprived area, recruited via schools and local youth clubs and mean age was 168 months.		The disinhibited indiscriminate scale (DAWBA-RAD) was a significant predictor of impaired peer relationships on the HoNOSCA. The superficial relationships item showed the most association with functional impairment.	
			DSED symptoms measured via the standardised development and wellbeing assessment–reactive attachment disorder (DAWBA-RAD).			
Guyon-Harris, Humphreys, Fox, et al. (2019)	Quantitative cross sectional	Examine associations between signs of DSED and RAD and social functioning in early adolescence.	Post institutionalised children, randomised into a high quality foster care intervention (n = 55) compared to post-institutionalised children in care as usual (n = 55) and 50 never institutionalised children from the local community. Participants assessed at 12 years. Signs of DSED were Measured using the standardised Disturbances of Attachment Interview-Early adolescence.	Peer conflict scale (PCS).	Symptoms of DSED (and not RAD) were associated with greater caregiver perceptions of the child being victimised and were perceived to have greater conflicts in peer relationships.	35 High
				McArthur Health and Behaviour questionnaire (HBA)	DSED (and RAD) associated with lower social competency, independent of placement disruption or time in institutional care.	

TABLE 1 (Continued)

Authors/Date	Study design	Investigative focus of study	Participants (*n*, age range, recruitment)	Measures (related to social functioning only	Outcomes (pertaining to social relationships/social function only)	CCAT score
Seim et al. (2022)	Quantitative cross sectional	To assess possible co-occurrence of psychopathology and/or psycho- social problems in children with DSED, or RAD.	A total sample of 381 adolescents (mean age, 16.7, range 12-20 years) in a group residential setting were assessed and 31 presented with DSED. DSED was assessed using the preschool age psychiatric assessment (PAPA) (caregiver report) due to lack of available age appropriate measure and then DSM-5 criteria applied stringently to determine if symptoms met diagnostic criteria.	Psycho-social difficulties were assessed via the child behaviour checklist for ages 12-18 years, In addition, the child and adolescent psychiatric assessment was utilised to assess for 'exposure to bullying' (and other psychiatric problems, not relevant to this review).	Children with DSED had significantly greater number of associated psycho-social problems than children without DSED, (mean 4.04) and were more likely to be bullied.	High 32
3. Self-esteem/Self-concept						
Vervoort et al. (2014)	Cross sectional quantitative	To compare indiscriminately friendly children with controls regarding their perceptions of self, reliability trust in significant others and perceptions of child-teacher relationship.	33 likely cases for disinhibited reactive attachment disorder (d-RAD, DSM-IV) from special education for children with emotional and behavioural disorders (mean age, 8.52) and 33 controls from general education (mean age, 8.42) (matched by age, sex and socio-economic status). DSED symptoms were assessed using the standardised relationship problems questionnaire	Self-description Questionnaire-I (SDQ-I) and the following 2 scales were used: The general-self scale and the peer relations scale. Three domains are assessed: Cognitive competence, physical competence and peer acceptance.	Perceptions of self-concept were higher in the d-RAD group than those of the control. The d-RAD group also reported more trust in the reliability of significant others but greater conflict with their teachers, despite dependency on the teacher-child relationship.	28 Moderate
Vacaru et al. (2018)	Quantitative cross sectional	To investigate possible associations between disturbed attachment, (DSED & RAD) and self-concept.	Thirty-three institutionalised children (Mean age, 9.75, range 4-12 years) participated along with staff working at the institute (caregivers, social workers and teachers). DSED was assessed via the disturbances of attachment interview (caregiver report) and the behavioural signs of disturbed attachment (observational measure)	Self-concept was assessed via the validated measure, the pictorial scale of perceived competence and social acceptance in young children.	DSED, and RAD, were generally associated with negative perceptions of self-competence, but self-perceptions of physical competence were higher than the teachers' perception of their physical competence.	High 33

(Continues)

TABLE 1 (Continued)

Authors/Date	Study design	Investigative focus of study	Participants (*n*, age range, recruitment)	Measures (related to social functioning only	Outcomes (pertaining to social relationships/social function only)	CCAT score
Seim et al. (2021)	Cross sectional quantitative	Investigate whether global and domain-specific self-esteem among adolescents living in group residential care differs between those with a RAD diagnosis, a DSED diagnosis, or neither RAD nor DSED diagnoses, and with adolescents in the general population.	306 individuals living in youth residential care in Norway, (mean age, 16.8 years, range, 12-20 years) of which 26 met diagnostic criteria for DSED and 28 RAD. DSED/RAD symptoms were assessed via preschool age psychiatric assessment (PAPA) (caregiver report) due to lack of available age appropriate measure and then DSM-5 criteria applied stringently to determine if symptoms met diagnostic criteria.	Self-perception profile for adolescents (SPPA) measuring global self- worth and domain-specific elements of scholastic competence, social acceptance, athletic competence, physical appearance, romantic appeal, and close friends.	Children with DSED demonstrated lower self-esteem regarding social acceptance compared to both children with RAD and the environmental control, as well as the 'typical development' group	37 High

4. Social interaction						
Groark et al. (2011)	Quantitative cross Sectional	Assessed caregiver characteristics and child-caregiver interactions in Latin american institutions.	3 institutions with 120 children, many of whom were indiscriminately friendly, were included. Age range, from birth to 7 years old (wards had average 8–23 children with some larger).	The caregiver—-child Emotional/Relationship rating scale (CCSERRS): Designed for purposes of measuring child-carer interactions in institutions. Children's problem behaviour scale (CPBS).	CCSERRS: Low levels of carer responsiveness and availability to the child were rated, although interactions were more positive during free play and physical caring. Children demonstrated poor responsiveness /anticipation, child directed behaviours and relationship with the caregiver. CPBS: High levels of indiscriminate friendliness, non-compliance, provocative interpersonal behaviour and aggression, but little stereotyped self-stimulation or withdrawn behaviours.	moderate 27
Sadiq et al. (2012)	Cross sectional quantitative	Pragmatic language deficits in children with DSED[RAD] compared to children with ASD and typically developing children.	35 children with DSED[RAD] (mean age, 6.7, range, 5–8 years); 52 children with autism (mean age, 6.4, range, 5–8 years) and 39 with typical development (TD) (mean age, 6.5, range 5–8 years) verbal IQ was within 'normal' range for all 3 groups. DSED[RAD] group recruited from clinical and social work services, the TD group recruited via their general practitioners, during a previous study. The autism group were recruited from a specialist tier 4 clinic.	Children's communication checklist (CCC).	CCC-2: The DSED[RAD] group performed most poorly in domains use of language in context, rapport and social relationships. Only the DSED[RAD] differed from the TD group on rapport	moderate 28

TABLE 1 (Continued)

Authors/Date	Study design	Investigative focus of study	Participants (*n*, age range, recruitment)	Measures (related to social functioning only	Outcomes (pertaining to social relationships/social function only)	CCAT scor
Davidson et al. (2023)	Mixed method cross sectional	To determine the outcomes of children with autism, in comparison to children with DSED, on standardised autism measures and a socially challenging behavioural observational assessment called live.	10 children with autism (no maltreatment history), 8 children with confirmed symptoms of DSED who were either caregiver referred or referred by local mental health service, and 10 typically developing children who were caregiver referred (age range, 5-11 years and groups were matched by age). DSED symptoms were confirmed via caregiver reports using standardised relationship problems questionnaire and reactive attachment disorder and disinhibited social engagement disorder assessment interview and via observation using the standardised waiting room observation.	Diagnostic interview for social and communication disorders (DISCO): Autism diagnostic observational Schedule-2 (ADOS-2) Live assessment: An unstructured dynamic assessment (2 assessors) designed to increase unpredictability and greater social challenge.	Disco: Almost all the children with DSED met core criteria for autism on the parent interview. ADOS-2: 62.5% of children with DSED did *not* meet diagnostic criteria for autism. But false positive outcomes were found in 3 cases of DSED, who had additional moderate to severe symptoms of ADHD. LIVE: The DSED group were more able than children with autism to engage in complex humour, their play was more creative and spontaneous, and children with DSED tended to involve the assessors, even if their interactions were not entirely appropriate. Controlling and/or obsessive behaviours and lack of empathy were noted in both groups.	30 High
Sheaffer, Golden, and Averett (2009)	Cross sectional quantitative	Decoding of emotions from facial expressions and paralanguage (intonation patterns) in DSED^{RAD}	17 children with DSED^{RAD} 15 children in foster care, without DSED^{RAD} recruited by social services, private clinicians and university psychology department; 31 typically developing children recruited via an afterschool organisation and university psychology department (age range 5-19 years).	Diagnostic analysis of nonverbal accuracy (DANVA2): 2 sub-tests relating to facial expressions, 1 sub-test relating to child paralanguage and 1 to adult paralanguage.	No significant differences found between groups regarding ability to decode emotions from facial expressions or paralanguage.	26 Moderate

Abbreviations: CCAT, Crowe Critical Appraisal Tool; DSED, Disinhibited Social Engagement Disorder; IQ, intelligence quotient; RAD, Reactive Attachment Disorder.

a measure designed for this environment. The institutionalised children showed little anticipation of caregiver interactions and tended not to signal or direct interactions. These findings are considered in relation to caregiver behaviour which lacked empathy and showed low responsiveness to child initiations. Due to the unusual caregiving environment, findings have limited generalisability.

Sadiq et al. (2012) used the standardised parent report Child Communication Checklist to investigate the pragmatic language (use of social language in context) of a community-based sample of children with DSED^{RAD} (n = 35), compared to children with Autism (n = 52) (average verbal IQ) and typically developing children (TD) (n = 39). The DSED^{RAD} and Autism groups significantly differed from the TD group in domains of, inappropriate initiation, coherence, stereotyped conversation and social interests, but *only* the DSED^{RAD} group significantly differed regarding rapport. Surprisingly, the DSED^{RAD} group showed *greater* impairment than children with Autism regarding use of language in context, rapport and social relationships. However, this is a study of moderate quality because the authors did not discriminate between DSED and RAD and unconfirmed co-occurring Autism, based on parent report only, was a possible confounding variable.

In contrast, a high-quality qualitative study reported that children with DSED (n = 8), compared to children with Autism (n = 10), demonstrated more engagement in complex humour, more creativity and spontaneous play and more often involved the assessors, even if their interactions were not entirely appropriate, during unstructured clinical observation (as opposed to caregiver report, as above.) Controlling and/or obsessive behaviours and lack of empathy were observed in both groups (Davidson et al., 2023). ADHD was co-existing in some of the children with DSED and impacted social behaviour on the standardised ADOS-2 assessment, but social skills were less effected by ADHD during the unstructured observation. The main limitation was the smaller sample size.

Finally, Sheaffer, Golden, and Averett (2009) investigated ability to decode emotions from facial expressions and paralanguage in children with DSED^{RAD} (n = 17) compared to a foster care group (n = 15), without DSED^{RAD}, and a typically developing group (n = 31) and found no group differences on the standardised measures. This study was considered of moderate quality because they did not discriminate between DSED and RAD, the samples were small and the DSED^{RAD} group were receiving therapy, which could inadvertently influence results.

The findings within this theme are mixed. Each study is measuring slightly different aspects of social interaction/communication which may account for some differences, and it appears that type of measurement (caregiver vs. observation) may be important.

DISCUSSION

This systematic review aimed to address the gap in knowledge regarding the social functioning of children with DSED. It is recognised that children who have experienced early childhood maltreatment are at higher risk of social relationship and communication difficulties (Cicchetti, 2016), and it seems that children with DSED are no exception. Regarding general social competencies, reports were consistent; children with DSED may present with greater social functioning difficulties than peers (Giltaij et al., 2016; Guyon-Harris, Humphreys, Fox, et al., 2019; Guyon-Harris et al., 2019, 2019; Millward et al., 2006; Pritchett et al., 2013), which supports the reconceptualization of DSED as a disorder of social-relatedness, separate from RAD (DSM-5). It is perhaps unsurprising then that children with DSED appear to be at higher risk of peer victimisation and conflicts in peer relationships (Guyon-Harris, Humphreys, Fox, et al., 2019; Kay & Green, 2013; Raaska et al., 2012; Seim et al., 2022). Only one study included child report, as opposed to caregiver report, but this qualitative study demonstrated that lack of understanding of friendships (Bennett et al., 2009), may be a key area for further investigation. For example, are peer problems reflective of cognitive deficits, as associated with Autism, or is lack of understanding of these relationships arising from missed opportunities and stressful experiences, as proposed by McCrory et al. (2022)? The findings regarding the social interactions/communication of children with DSED were mixed, but differences in measurement stood out as an important factor. However, one interesting point of convergence between the findings of Sadiq et al. (2012) and Davidson et al. (2023) regarded the initiations of children with DSED, which were not always appropriate, even if Davidson et al. did not perceive them as autistic in nature. Moreover, Davidson et al. found that controlling behaviours and lack of empathy overlapped between DSED and Autism. These latter behaviours, in addition to inappropriate initiation, are likely to impact rapport, one of the key areas of difficulty reported by Sadiq et al. It is conceivable that subtle pragmatic language/interaction skills are negatively impacted by both core symptoms of DSED and/or these additional behaviours, which have been reported in other studies (Mukaddes, et al., 2000; Pears et al., 2010; Rutter et al., 1999). Future research is required, with larger samples, and would benefit from both caregiver report *and* observation, perhaps involving relevant professionals like Speech and Language Therapists to complete targeted investigation of pragmatic language. It is also vital that observational studies, such as Davidson et al.'s, be repeated, but in comparison to typically developing children. The interactional skills of the children with DSED may be 'improved' compared to the children with Autism, but it is unclear if the skills of the DSED group were developmentally appropriate.

Finally, most of the included studies suggest that children with DSED may have poorer self-estem/self concept with regards to social acceptance (Seim et al., 2021; Vacaru et al., 2018), but one study found that children with DSED had higher perceptions of self-concept than typically developing peers (Vervoort et al., 2014). As participants in the Seim et al. study were older (mean age, 16.5 years), it is plausible that by late adolescence, when peer relationships have even greater salience in identity formation and social behaviour (Merritt & Snyder, 2015; Reitz et al., 2014) participants were more acutely aware of their difficulties. Yet, Vacaru et al. (2018) did find that in one specific domain, physical competence, children with DSED perceived themselves as more competent than significant others perceived them. The authors argue that a self- perception bias may have some benefit for socialisation in certain settings (Vacaru, et al., 2018), and this seems worthy of further exploration. For example, other groups of children with neuro- developmental conditions, such as ADHD, have been found to

present with self-perception bias regarding competencies, which have been considered self-protective (Ohan and Johnston (2002)). As 40% of Vervoort's sample were found to have co-occurring neuro-developmental conditions, it would be useful to better understand whether sample bias is accounting for Vervoort's findings or whether, in some areas of socialisation, self-perception bias may also be protective for children with DSED.

Neurodevelopmental complexity and future research

Both Autism and ADHD are associated with poorer social functioning and poorer peer relationships, however, symptoms of both were found to overlap in some of the included studies (Davidson et al., 2023; Sadiq et al., 2012; Vervoort et al., 2014). Although there appears to be no aetiological reason why DSED and Autism cannot co-exist (Mayes et al., 2017; Minnis et al., 2020), there are now some studies suggesting that core symptoms of DSED are discriminable from Autism (Davidson et al., 2015, 2023; Davidson, Minnis and Moran, 2022; Rutter et al., 1999). In contrast, core symptoms of ADHD, such as poor inhibitory control appear to be associated with core symptoms of DSED (Bruce et al., 2009; Pears et al., 2010). Preliminary research suggests that neglect is associated with negative impact on higher cognitive skills (McLaughlin, 2017), therefore further research with children with DSED may help to elucidate understanding regarding possible pathways into DSED and why DSED is more persistent in some children with DSED than others (Scheper et al., 2019). In this review, Guyon- Harris, Humphreys, Fox, et al. (2019) found that children with *persistent* DSED had the greatest social difficulties (0% were socially competent), but it is unknown whether there were any differences within the 'persistent' DSED group, regarding neurodevelopmental complexity. Further understanding of the inter-play with ADHD symptoms is important for case management as adolescents with DSED in residential care were 2.5 times more likely to have additional ADHD (Seim et al., 2022) and longitudinal data from the English-Romanian Adoptees study demonstrated that at 15 and 25 years old, DSED behaviours were still present, with some overlap with Autism and/or ADHD (Kennedy et al., 2017; Sonuga-Barke et al., 2017), but at 25 years, functional problems, like employment issues, were related to the inter-play with ADHD (Kennedy et al., 2017).

Gajwani and Minnis (2023) argued that children with DSED, or RAD, may experience 'double jeopardy' regarding mental health outcomes due to interplay of co-occurring neurodevelopmental conditions. Our findings appear to suggest that children with DSED are at higher risk of social problems, therefore in cases of childhood maltreatment, *both* DSED and possible overlapping neuro- developmental conditions must be considered alongside impaired social function to provide a fuller picture for health and social care management. Furthermore, social problems need to be considered in the *early years*, and as a preventative approach to later mental health difficulties, especially given the persistent nature of DSED.

Limitations

Half of the studies were considered of moderate quality due to small samples, possible confounding variables and, in some cases, lack of discrimination between DSED and RAD. Thus we have been careful in our discussion to present only hypotheses and suggest some caution in interpretation of findings. It was also noted that one relevant study (Vacaru et al., 2018), was initially missed out. This occurred because the abstract referred to disturbed attachment and exploratory behaviours, thus we wrongly assumed that the study was about attachment *pat- terns* rather than DSED. On noting this error, the study was read in full and subsequently included. Due to the scarcity of social relationship literature regarding DSED, we took a top-down approach to the search, focusing on broad relational terms and considered possible themes as findings emerged. This meant the search did not include the key words, 'self esteem'/'self concept' and inclusion of these terms within future investigations may yield an even more inclusive picture.

Conclusion

Bearing in mind the limitations, the evidence consistently suggests that children with DSED present with poorer social competencies than peers, have greater peer difficulties and may have poor self esteem/self concept. Further research in specific areas such as pragmatic language and regarding the interplay with other co-occurring neurodevelopmental conditions is required. However, re- searchers and clinicians need to consider the presence of DSED, in maltreated children, possible neurodevelopmental overlap *and* relative impact on social functioning to better support this underrepresented group of children and their families.

AUTHOR CONTRIBUTIONS

Claire Davidson: Conceptualisation, methodology, investigation, analysis, writing, writing-review and funding acquisition. **Shahela Islam:** Investigation, analysis and writing-review. **Enrico Venturini**: Investigation and writing-review. **Anja Lowit:** Conceptualisation, methodology, supervision, review of writing. **Christopher Gillberg**: Conceptualisation, methodology, supervision, review of writing. **Helen Minnis**: Conceptualisation, methodology, supervision, review of writing.

ACKNOWLEDGEMENTS

Funding for this project was provided by the Castang Foundation (UK) [LAY1-WS_LEGAL.FID1730709, Gillberg Neuropsychiatry Centre, and NHS Greater Glasgow and Clyde to complete this work as part of a wider research study.

CONFLICT OF INTEREST STATEMENT

The authors have declared they have no competing or potential conflicts of interest.

ETHICS STATEMENT

Ethical approval was not required for this article.

DATA AVAILABILITY STATEMENT

Data derived from public domain resources.

ORCID

Claire Davidson https://orcid.org/0000-0001-5414-0402
Helen Minnis https://orcid.org/0000-0002-2377-8945

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How to cite this article: Davidson, C., Islam, S., Venturini, E., Lowit, A., Gillberg, C., & Minnis, H. (2024). Social competencies of children with disinhibited social engagement disorder: A systematic review. *JCPP Advances*, e12226. https://doi.org/10.1002/jcv2.12226

Reflections on Paper 1

After completion of our systematic review investigating the social functioning of children with DSED, the following points appear salient.

- Children with DSED may present with poorer social competencies than typically developing children and, from what research is available, it is suggested that they are more likely to have problems with peers, conflicts in relationships and may have poorer self-esteem/self-concept related to social relationships.

- As children with Autism also have greater social relationships difficulties which may manifest as peer problems, this domain appears to be an area where clinicians can expect general overlapping difficulties.

- One of the specific relational difficulties in Autism is thought to be impaired social reciprocity, but social reciprocity was not investigated in any of the studies within the systematic review. Furthermore, findings highlighted a reliance on caregiver report of problems. One of the requirements of the Autism clinical guidelines in determining lack of social reciprocity is the inclusion of direct child observation. Further research is required to better understand the overlap in the context of the diagnostic symptoms of Autism, and at the level of clinical assessment using observational measures.

- Some problems with social communication in DSED were identified but findings were inconclusive and may relate to methodological differences i.e. parent report v clinical observation.

- Related to the above is the lack of investigation of the views of young people, which were specifically examined by one study only. Further qualitative research including children's perspectives of their social interactions is warranted.

- The findings also highlighted the existence of co-occurring neurodevelopmental conditions such as ADHD in DSED and this raises an important question regarding the relative impact of DSED+ADHD on social functioning.

In the next study, we explore the social relationship and communication skills of children with Autism and children with DSED in relation to the Autism diagnostic criteria. We also take an ESSENCE approach, identifying co-occurring neurodevelopmental conditions and discuss the findings in this context.

Chapter 3: Methods

This chapter provides an overview of the methods and the recruitment process that the remaining three papers are based on. The section begins by discussing the methods and where relevant the challenges which impacted the design and implementation of the methods. Regarding procedures, this chapter expands on issues common to all the papers, however, specific elements such as procedures of analysis which would normally be in the methods are specified in each paper individually.

3.1 Research design

Design at conception of the study: The original proposed design was quantitative in nature using iterative processes to identify key symptoms or skills which may differ between children with Autism and children with DSED on standardised assessments. It had been planned to recruit a phase 1 sample of 30 children, (10 children with Autism and no maltreatment history, 10 children with DSED (established maltreatment history) and 10 typically developing children), complete holistic neurodevelopmental assessment and then recruit a 2nd sample of the same size and same groups. The phase 1 sample would be like a 'training set' and then the phase 2 sample, a test of replication of findings. The combined larger sample would enable statistical testing of discriminatory functions of possible differentiating symptoms or behaviours.

Actual design implemented: Due to unforeseen delays to the time schedule, some of which were out with control, it was not possible to complete the planned phase 2 stage of recruitment. Instead, adjustments were made to achieve the most clinically useful and research-rigorous standards with the phase 1 data, while acknowledging the limitations. One adjustment included adapting a more descriptive behavioural clinical analysis of the data in the form of a case study series, as opposed to statistical analysis, with the benefit of richer more in-depth outcomes. While one limitation of the change to a case series was a smaller sample size, there was good argument for this design because the population in question, children with DSED, are a grossly under investigated population (see Figure 1, p 27) thus a smaller in-depth approach was a beneficial way to obtain a thorough

understanding of the strengths and challenges faced by the children within the specific sample and uncover areas worthy of future investigation. The case study design also allowed an iterative approach; each study built on both the learnings and the knowledge gaps identified from the prior investigation. For example, study 1 (paper 1) identified that children with DSED may have broadly similar social relationship difficulties but there was a distinct lack of qualitative methodology exploring child experiences and mixed reports regarding social communication differences in Autism compared to DSED. Drawing from these findings, study 2 (paper 2) explored the role of standardised diagnostic Autism assessments compared to an unstructured conversational-observational approach and found that differences between Autism and DSED could be descriptively identified during the unstructured approach and did appear to lie within the social communication domain. Following on, study 3 (paper 3) focused down on the communication skills of the sample of children with DSED compared to the sample of children with Autism by exploring the still unknown area of receptive and expressive language skills. Social communication was further explored through clinical analysis of pragmatic language, a core component of social communication which is thought to be universally impaired in children with Autism (Rhea, 2007).

The second adjustment was to utilise existing data to investigate, with larger samples, fundamental concepts that were previously unexplored. This involved investigation of receptive vocabulary in children with DSED compared to children with Autism. Receptive vocabulary is a foundational building block to understanding and using language, but it has never been investigated in DSED. In study 3 an existing data set of 104 children (43 with Autism and no maltreatment history, 24 with DSED (established maltreatment history) and 37 typically developing children) was used to examine receptive vocabulary as the starting point for understanding the language and communication skills of children with DSED compared to children with Autism.

Before discussing the participants and procedures of the case series, the following section briefly describes the challenges faced which limited the opportunity to carry out the original quantitative design. It is important that these challenges are recognised as they are not unique to this doctorate study but raise important questions for researchers going

forward regarding the ways in which research governance can support, rather than restrict, research with vulnerable individuals. The challenges of recruiting from clinical services are also discussed.

Barriers to recruitment

Obtaining Ethics: The first barrier was a much longer than expected time to obtain ethical approval. It was readily accepted that we needed to recruit children with *symptoms* of DSED, rather than a diagnosis, (it is not routinely diagnosed yet) and then confirm that symptoms met the diagnostic criteria using diagnostic tools but the sticking point was our decision to disclose a DSED diagnosis on a case by case basis. As discussed in the previous section, giving a diagnosis of DSED to a foster carer could be of benefit in their understanding of the child, but it may be detrimental for a biological parent, who has had their child returned and is working with services to develop their relationship with their child. Here, the maltreatment aetiology inherent to a diagnosis of DSED could serve as a reminder of blame/self-blame and be harmful to the developing relationship. We proposed, in this instance, to discuss the DSED *behaviours* in the context of the child's early experience without formally labelling the diagnosis. The ethics committee, being unfamiliar with the disorder and its ramifications, understandably argued that this withholding of information was not ethical. Several back-forth discussions were required over six months to explain that all the clinical information would be supplied to families by the research team and/or referring clinician, but the presentation of the information (diagnostic label versus discussion of core behaviours) would be tailored on a case by case basis, because it was in the best interests of the child and their family. Eventually, the ethics committee agreed and ethics approval for the full study was granted by the West of Scotland Research Ethics Committee. Nevertheless, the complex nature of the clinical sample and the related delay in achieving ethics, interrupted the start of recruitment by several months.

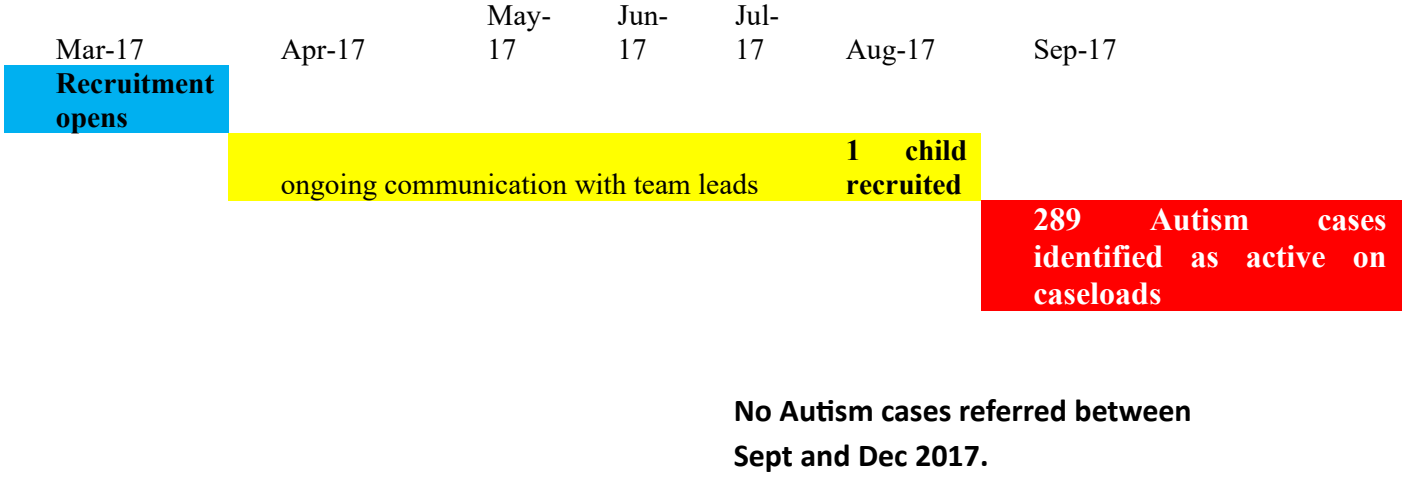
Recruitment from Clinical Services: Other recruitment barriers were related to the broad difficulties of recruitment of clinical samples from clinical services. For the Autism group, recruitment from clinical services was limited because locally it is the Community

Autism Teams, not CAMHS, who make the diagnosis but as they do not hold the cases they were less keen to refer children to the research who no longer had input from their service. Children with Autism tend only to be seen in CAMHS as a result of additional mental health problems and for these cases, some gatekeeping was apparent. Gatekeeping is a phenomenon where clinicians demonstrate reluctance to offer all eligible participants the chance to participate in research and instead approach only those who *they* believe the research to be suitable for (Fletcher et al., 2012, Bucci et al., 2014). A few clinicians raised concerns at the 'introduction to the research' meeting that such *"complex cases were not suitable for research."* Regarding children with DSED, the current lack of identification of DSED within clinical practice was a barrier. Much of this was likely due to limited awareness and the fact that it is a disorder which is extremely under researched in comparison to other neurodevelopmental conditions (Zeanah et al., 2016; Minnis, 2013). However, due to the maltreatment aetiology some clinicians are reluctant to include DSED within their formulation as maltreatment can be difficult to conclusively evidence, (Dyer, 2010) and some clinicians might perhaps feel on safer ground considering behaviours instead within a trauma framework, without making the diagnosis.

Despite some of the above difficulties, it is likely that, for the vast majority of clinicians, the intentions to engage with the research were diluted by the daily competing pressures on clinician's time and resources (Bucci et al., 2014). Directly related to pressure on clinician time is the value that clinical work is of greater priority than research (Fletcher et al., 2012). While this is understandable, it can also result from limited resources or lack of clinician interest in the research question; the latter of which has been shown to impact on recruitment (Ross et al., 1999). The fact that the responsibility for research tends to sit with universities and the primary responsibility of direct patient care sits with clinicians has been raised internationally as a barrier to recruitment (Allison et al., 2017). Despite the good intentions of clinicians to support recruitment, it was apparent that another source was required, due to the time constraints of this being a doctorate study and lack of additional funding to support a longer period of recruitment.

Ethics amendment: in response to the lack of recruitment from CAMHS, an ethics amendment was requested. The request was twofold: 1. Recruitment via advertisements on Autism and Fostering/Adoption charity websites was requested, in addition to the previously agreed recruitment from clinical services. This recruitment method had been used successfully by colleagues in the past, (Kocovska et al., 2012). 2. I requested permission to access the electronic health records in order to identify possible eligible cases using diagnostic key words (Aspergers, Attachment Disorder) (the clinical system used pre-DSM-5 terminology) and then approach clinicians to ask if they felt the identified cases may be suitable for the research. An email survey with CAMHS clinicians suggested that they preferred this method as it took some of the burden off them. The ethics amendment was approved without issue, but inevitably it still impacted recruitment time. Permission to access the electronic records was granted by September 2017. As figure 1 demonstrates, a substantial number of Autism cases were held on CAMHS caseloads, yet only 1 Autism referral occurred in the whole recruitment period.

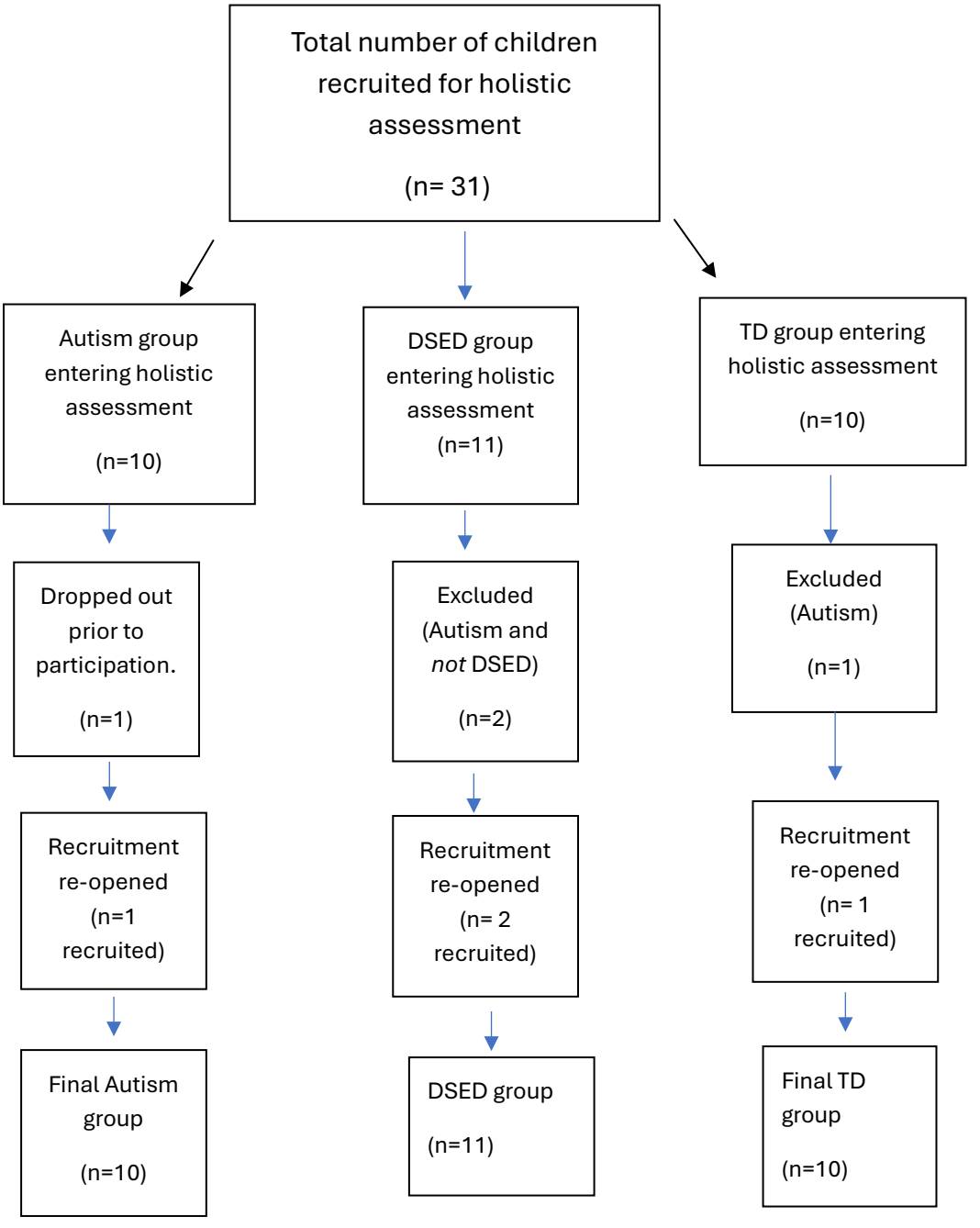
Figure 1: recruitment difficulties during first year of the doctorate study



In contrast, referrals were readily received from families following advertisements being placed on the various charity websites. By the end of the recruitment phase, 9 of the 10 children with Autism were self-referred by their families and, in the DSED group, 7 of the 11 were self-referred.

Neurodevelopmental complexity: One final recruitment challenge arose once the holistic assessments with the children with DSED began. While all the children recruited to the DSED group met core criteria for DSED on the DSED multi-informant package, it became apparent following the clinical observations that 2 of the recruits were children with Autism, who had a maltreatment history, but did *not* have DSED. These 2 children were excluded, and recruitment of an additional 2 cases was then required. Of note both these children were CAMHS referrals. Similarly, 1 child who was recruited to the typically developing group appeared to meet criteria for Autism. This case was discussed with the Scottish Centre for Autism as well as HM to get a multi-disciplinary perspective and it was agreed that this child should be excluded from the typically developing group. A discussion was undertaken with the child's parent regarding our formulation and subsequently another child had to be recruited to the TD group. Figure 2 below demonstrates the flow of recruitment.

Figure 2: Recruitment breakdown.



3.2. Case Series Sample:

Participants

The case study sample in the case series were 3 diagnostically distinct groups of children: 10 children with Autism and no maltreatment history (7 males: 3 females), 11 children with DSED (established maltreatment history) (8 males: 3 females) and 10 typically developing children (8 males: 2 females). All children were between the ages of 5-11 years and were in mainstream primary school. The Scottish Index of Multiple Deprivation (SIMD) was used, in addition to age, to group match as closely as possible, by socio-economic status. SIMD data represents areas of deprivation based on postcode; the areas are split into deciles, and the higher the decile the greater the proportion of areas within which are considered as deprived. Table 1 demonstrates the demographic data of the 3 groups of children.

Table 1: demographics

Group	*Gender (male: female)*	*Mean age (range)*	*Mean SIMD decile (range)*	*Co-occurring diagnosis*
Autism (n = 10)	7M: 3 F	7.4 years (5-11)	5 (1-10)	Two children had existing diagnosis of ADHD
DSED (n=11)	6M: 5 F	7.5 years (5-11)	6.3 (1-10)	Two children identified with ADHD Two children identified with co-occurring Autism
TD (n=10)	8M: 2 F	9.67 years (8-12)	6 (2-9)	N/A

Two of the children in the Autism group had an existing diagnosis of ADHD which was being supported via medication. Following holistic neurodevelopmental assessment it became apparent that two children in the DSED group also had additional, previously unidentified, ADHD and onward referrals to CAMHS for ADHD support were made for these children. The decision was made not to exclude children with co-occurring neurodevelopmental conditions because overlap is now known to be the norm, not the exception, (Gillberg, 2010). While lack of a 'pure' sample could introduce some sample bias, and additional complexity in analysis, the benefit was a sample more reflective of 'real world' clinical cases which are more likely to be seen in CAMHS. Given the clinical nature of the overall research aim/individual research questions and the in-depth design of the case series, which allows for analysis of complexity, it seemed reasonable to include these cases with transparency about the strengths and weakness within each individual study.

During the holistic assessments, it also became apparent that another two children with DSED also had co-occurring Autism. This was more problematic as the primary aim was to differentiate between Autism and DSED. By this point, there was not enough time to recruit additional DSED only cases, so the decision was made to include these cases of DSED + Autism in paper 2 as the findings actually provided useful information regarding the discriminatory abilities of the observational assessment tools. However, it was necessary to exclude these cases from the analysis of papers 3 and 4; the impact of which was reduction of the sample size for these studies.

Measures

One strength of this case series design was the use of holistic neurodevelopmental measures to investigate a broad range of symptoms (related to diagnostic criteria), skills (social interaction and communication, imagination, receptive, expressive and pragmatic language), and behaviours (sensory processing) of children with DSED, in comparison to children with Autism. Another was the multi-informant approach and the third was the comparison of standardised measures with clinical expertise/judgement via conversational based unstructured observation. The measures used to answer the research questions specific to each study vary and are described within each paper, but common to all the studies was the

initial assessment and these measures were as follows (N.B discussion of the psychometric properties and justification for choice of assessment are described in the subsequent sections):

Measures to confirm DSED symptoms met diagnostic criteria for DSED (DSED group only).

Reactive Attachment Disorder and Disinhibited Social Engagement Disorder Assessment Interview (RADA) (Lehmann et al., 2020) is a standardised semi-structured interview for the diagnosis of DSED and is completed by parents/caregivers.

The Teacher Relationship Problems Questionnaire (Teacher RPQ) (Minnis et al., 2007) is a 10-item screening questionnaire for symptoms of DSED completed by teachers.

Waiting Room Observation (WRO) (McLaughlin et al., 2010): is a structured 19-item observation of child behaviour with parents/carers and a 'stranger' carried out in a clinic waiting room. The role of the stranger, a trained rater in the WRO, is to record the interactions of the child with their primary caregiver, the child interactions with the clinician on greeting them, and interaction or social approach by the child to the stranger. The stranger was a research colleague who was otherwise uninvolved with the study and unknown to the participants.

Standardised diagnostic multi-informant assessment of Autism (All 3 participant groups).

Diagnostic Interview for Social and Communication Disorders (DISCO) (Wing et al.,2002) is a standardised in-depth interview for the diagnosis of Autism and is completed with primary caregivers.

Autism Diagnostic Observational Schedule-2 (ADOS-2) (Lord et al., 2000) is a standardised observational tool used to diagnose Autism across all ages and developmental stages. The aim is to assess absence or presence, and severity, of the core features of the dyad of impairment.

Autism Spectrum Screening Questionnaire (ASSQ) (Ehlers, Gillberg and Wing, 1999,) is a standardised screening tool for use with caregivers or teachers. In this study the ASSQ was sent to teachers of all participants for completion with a S.A.E.

Unstructured conversational based observation

Live Assessment: The Live assessment is a triadic (2 assessors and the child) unstructured assessment designed to increase unpredictability and greater social challenge. Two assessors interact with the child individually and together fjust as one might during group interaction. The assessors use informal conversation, games, banter (playful teasing), direct questioning and create social scenarios for the child to respond to. The outcomes are analysed descriptively and using clinical judgement.

Additional information on possible co-occurring neurodevelopmental conditions

Autism - Tics, AD/HD and other Comorbidities (A-TAC) (Anckarsater et al., 2007) is a well validatd screening questionnaire for use in research for assessment of child neurodevelopment. It is completed bv primary caregivers. When making decisions regarding co-occurring neurodevelopmental conditions the A-TAC was examined in addition to all the other multi-informant data and the video data of the observational assessments in case conference with HM, a Consultant Psychiatrist expert in maltreatment associated problems and neurodevelopment. On occasion this also involved CG, a Consultant Psychiatrist and expert on child neurodevelopmental conditions and co-occurrence.

Procedure

Prior to specific investigations as described in papers 3 and 4, the core assessments as described above were undertaken as follows:

DSED group only: The RADA was completed via telephone interview with primary caregivers of children referred to the DSED group. Additional information regarding disinhibited behaviour was gathered via the Teacher-RPQ which was posted and returned via S.A.E. The Waiting Room Observation was conducted in the research clinic prior to the first meeting. Only children whose DSED symptoms met diagnostic criteria were admitted to the study.

All groups: caregivers were invited to complete the DISCO assessment at a home visit. For most cases the DISCO was completed by the doctoral researcher (CD-J), who is a Speech and Language Therapist and trained to administer both DISCO and ADOS; two DISCOs were administered by another trained Speech and Language Therapist. Within 1–4 weeks of the DISCO assessment, the child was invited to the clinic for ADOS assessment (no child in any of the groups had participated in an ADOS assessment within 6 months of the study, as per the requirement for re-assessment with the ADOS (Lord et al., 2000). CD-J was still a stranger to the child at this point. Within 1–4 weeks of the ADOS, participants were invited back to complete the Live. The Live assessments were administered by CD-J (assessor 1) and her colleague, a Health Psychologist with both lived experience as a parent of a child with Autism and a research interest in neurodevelopmental conditions and maltreatment associated problems (assessor 2). Both assessors were trained to use the Live by the Scottish Centre for Autism (see section below). The A-TAC was completed by caregivers in the waiting room. Table 2 demonstrates the assessments undertaken with each group, and further details are presented in paper 2.

Table 2. *Assessments undertaken, per group, (reported in paper 2).*

	Assessment prior to participation	*Maltreatment check*	*Assessment undertaken during study participation*
Autism Group	None (referred with pre-existing diagnosis)	Electronic health records checked to confirm **lack** of maltreatment history.	• DISCO (caregiver interview). • ADOS-2 (structured child observation). • Live assessment (dynamic unstructured child observation). • A-TAC
DSED group	• RADA (caregiver interview). • Teacher RPQ (teacher questionnaire). • WRO (observation in clinic).	Maltreatment history provided either by referring clinician or foster carer/adoptive family	• DISCO (caregiver interview). • ADOS-2 (structured child observation). • Live assessment (dynamic unstructured child observation). • A-TAC
Typically Developing Group	None	**lack** of maltreatment history confirmed via electronic health records.	• DISCO (caregiver interview) • ADOS-2 (structured child observation) • Live assessment (dynamic unstructured child observation). • A-TAC

In addition to the assessments demonstrated in table 2, the children also participated in an assessment of receptive and expressive language ability and the caregivers completed a standardised screening of language and pragmatic skills and a standardised assessment of sensory processing. These assessments are discussed in detail in paper 3, (chapter 5) and paper 4, (chapter 6) respectively.

3.3. Diagnostic processes and decision making for study 2 (paper 2)

Paper 2 explored the role of assessment tools in supporting differential diagnosis of Autism from DSED. For the remainder of this chapter, the context for paper 2 is set through discussion of the current best practice guidelines for assessment along with justification of standardised diagnostic tools chosen for inclusion. Autism is addressed first and then DSED.

Current Autism assessment practices

The tools used during clinical assessment of Autism may vary across different health boards within the UK, but assessment procedures are informed by the minimum requirements within the national guidelines; Scottish Intercollegiate Guidelines Network (SIGN): Assessment (2016) (SIGN, 2016) and the National Institute for Health and Care Excellence (NICE, 2011; 2017).

Both the SIGN and NICE guidelines are clear that the minimal information to be gathered to provide a 'gold' standard assessment is a) parent/caregiver report which includes a developmental history, b) child observation and c) reports from other environments, such as school. These should be discussed in a multi-disciplinary context.

The clinical guidelines recommend 4 diagnostic tools for possible use; the Autism Diagnostic Interview-Revised (ADI-R) (caregiver report) (Le Couter, Lord and Rutter, 2003); The Diagnostic Interview of Social Communication Problems and other (DISCO) (caregiver report) (Wing et al., 2002); The Developmental, Dimensional and Diagnostic Interview (3di) (caregiver report) (Skuse et al., 2004) and the Autism Diagnostic Observation Schedule-2 (ADOS-2) (child observation) (Lord et al., 2012). Yet, caregiver diagnostic tools tend not to be used in clinical practice because they are lengthy and therefore too burdensome in a busy clinic. Instead, health boards tend to develop their own

agreed caregiver and developmental history interview and then use the diagnostic observation tool, the ADOS-2, in direct child assessment.

In contrast, caregiver diagnostic tools are widely used in research, although which tool is dependent on the study aims and is at the discretion of the research team. The ADOS has also been widely used in research, with good reliability and validity (Lord et al., 2000) and along with its popularity in clinic, it has become thought of as the 'gold standard' observational assessment for Autism.

Justification for choice of Autism diagnostic tools for paper 2

The objective was to select diagnostic tools which have the best rigour, but most relevancy to the research aims, without overburdening participants, while adhering to the clinical assessment guidelines.

Caregiver report

3DI: The 3DI (Skuse et al., 2004) is considered a reliable diagnostic instrument (SIGN, 2016) but, compared to the ADI-R and DISCO assessments it has a limited evidence base, and thus was excluded from consideration.

ADI-R: The ADI-R is a structured parent/caregiver interview which comprises 93 items about past and present behaviours and covers early development, language and communication, reciprocal social interaction, repetitive and restricted behaviours and other more general behaviours associated with Autism. During the studies of reliability (n=20) and validity (n=30), conducted in Canada with clinic referred children (Equal numbers of children with Autism and children with ID), ADI-Rs were rated and videoed by assessors blinded to the diagnoses. Almost all items had good inter-rater reliability of .75 and above (Lord, Rutter and Le Couter, 1994). The diagnostic algorithm also achieved good inter-rater reliability (100% agreement of not Autism for non-autistic group and 94% agreement of Autism for the Autism group). During revisions some questions were amended to target more Autism specific behaviour and this improved differentiation of Autism from ID, (Charman and Gotham, 2013). Furthermore, good inter-rater reliability and validity has been found across research studies

with North American clinic referred participants (Lord et al.,1994; Cicchetti et al., 2008; Risi et al., 2006).

DISCO: Like the ADI-R, the DISCO is a structured interview for caregivers. Ratings are made regarding both current presentation and whether symptoms have ever been present, allowing change to be assessed over time. It encompasses 362 items, covering the core domains of social interaction, communication, imagination and repetitive and restricted behaviours, and additional non-Autism specific domains such as early development, daily living skills, sensory issues, sleep problems, attention difficulties, mood and other psychiatric or forensic problems. The DISCO is more broad ranging because it was designed to be used for clinical purposes to provide a detailed profile of the individual's pattern of development, behaviour and needs (Wing et al., 2002). The inter-rater reliability of the DISCO was tested during assessment of 36 children with a diagnosis of ASD (18 high functioning: 18 low functioning), 17 children with ID, 14 participants with specific language disorder and 15 typically developing children. Participants were recruited from clinics or special educational needs schools in one major city in the UK. All assessments were rated by 2 assessors, blinded to the diagnosis. Agreement was high for 90.5% of developmental items for the school-age children and 83.4% for the untypical behaviour items. For the pre-school children the levels were 89.0% and 83.7% respectively (Leekam et al., 2002). The DISCO has also been used successfully in European research, with good inter-rater agreement and validity (Nygren et al., 2009; Leekam et al., 2002; Billstedt, Gillberg and Gillberg, 2007), but like the ADI-R, prior to its revision, some items did overlap with ID. Given the very broad nature of the DISCO it is not surprising that it has high sensitivity and lower specificity (Nygren et al., 2009).

It is clear that both the ADI-R and DISCO have good psychometric properties, but the DISCO was chosen for paper 2 because it has been validated with UK clinic samples and primarily because it's broader nature better fitted with the research aims. These were: to determine which behaviours, if any, stood out as possible areas of discrimination between Autism and DSED, and used as indicators in clinical practice. At this stage, it was not known whether possible areas of differentiation would be related to core Autism difficulties (dyad of impairment) or a wider range of associated difficulties. Charman and Gotham, (2013) argued in the review of Autism diagnostic tools that the DISCO was one of the best instruments for

providing an overall profile of skills and abilities, across multiple domains, and it was this strength which made it the instrument of choice.

Child observation

ADOS-2: The only available standardised observational tool is the ADOS-2, which is evidenced to have good discriminate validity (SIGN, 2016). The ADOS was updated to the ADOS-2 to account for the changes to the social interaction and communication domain in DSM-5 and there have since been mixed reports (Dorlak, 2018), particularly relating to specificity (Kamp-Becker et al., 2013; Medda et al., 2019) and rater- reliability. For example, in one recent study, large variation in coding of behaviours was found among 100 trained clinicians, when reviewing assessments of children referred to specialist Autism outpatient clinics in Germany (Kamp-Becker, 2018). Nevertheless, the ADOS-2 is considered the 'gold standard' and was therefore included in the assessment battery. Although the ADOS-2 is designed to be administered and coded by one assessor only, across UK NHS health boards clinicians tend to complete the ADOS-2 in pairs, (one assessor and one note taker, usually in mixed professional pairs). In this research the ADOS-2 was administered by 1 assessor, as per the training guidelines, but was video-recorded to support record taking.

Live Assessment: In addition to the ADOS-2, we chose to include a 2nd child observational assessment called the Live assessment. The Live is a behavioural assessment designed and used by the Scottish Centre for Autism (SCA), for 30 years, for assessment of 2nd opinion and complex cases. The Live was chosen as a contrast to the ADOS-2 because, unlike the ADOS-2, the Live is unstructured, free flowing and is administered by 2 assessors who interact both individually and jointly with the child to create a 'group' dynamic. It was important to include an unstructured observation because a previous study by our research group, (Davidson et al., 2015) suggested that differences in social interaction and communication may be best identified via unstructured and conversational observation. The free-flowing nature of the Live assessment also enables assessors to ask the child directly about their experiences using a conversational approach. One example might be chatting with the young person about their friends/activities they do with friends. Another is their sensory preferences/differences. This differs from standardised assessment approaches which tend to rely on carergiver report only,

or test devised structured questions as is the case with the ADOS-2. The Live takes approximately 30-45 minutes to administer, which is similar or less time than the ADOS-2 (45-60 mins), and while it may appear more burdensome on service resources because of the requirement of 2 assessors, in reality it is not because in current clinical practice, the ADOS-2 is routinely being administered and scored by 2 clinicians at a time.

Teacher report

A large number of questionnaires are available to screen for symptoms of Autism but the validity of many of these are unknown; like diagnostic tools, screening tools tend to be tested with referred clinical samples which may be biased by sample characteristics but may be less rigorously validated. For both screening and diagnostic tools population studies are lacking (Charman & Gotham, 2013). We chose to include the Autism Spectrum Screening Questionnaire (ASSQ) (Ehlers, Gillberg and Wing, 1999) because it is one of the few which has established good reliability and validity (Ehlers, Gillberg and Wing, 1999; Posserud et al., 2008) and has been used in Scandinavian research effectively with clinic referred samples as a parent or teacher screening (Bilenberg et al.,2005; Mattila et al., 2007; Posserud et al., 2006).

Current DSED assessment practices

In contrast, to Autism, the research evidence regarding DSED is still in in its infancy, therefore much less information is available to clinicians with regards assessment practices (NICE, 2015). Furthermore, DSED is still not routinely assessed for in clinic settings. This may be in part because of lack of awareness and knowledge, and in part because clinicians are wary given the diagnosis assumes the aetiology of maltreatment and that can be categorically difficult to evidence in practice.

Justification for choice of DSED diagnostic assessments in paper 2

There are many questionnaires and observations which purport to assess maltreatment-associated behaviours but most lack rigorous testing and they have not been updated to reflect the DSM-5 classification (Monette et al., 2022). The exception is the Early TRAuma-Related Disorders Questionnaire (ETRADQ) which demonstrated good convergent validity

with the well-validated Relationship Problems Questionnaire and the Reactive Attachment Disorder and Disinhibited Social Engagement Disorder Assessment Interview, good internal consistency (.88-.95 for all the sub-scales), good re-test reliability (.83-.91) and it can discriminate between children with and without DSED (Monette et al, 2022). However, we chose not to use this tool as it was designed not as a diagnostic tool but as a screening tool, primarily to increase time efficiency for busy clinicians, who would then use a follow up diagnostic tool if indicated.

There are few diagnostic tools for assessment of DSED (Lehmann et al, 2018), but probably the tool most frequently used in research is the Disturbances of Attachment Interview (DAI) (Smyke et al., 2002). The DAI was developed during the Bucharest Early Intervention Project, which was a US led randomised control trial of nurturing foster care versus institutionalised care as usual in communities in Romania. The DAI has good internal consistency (0.80-0.83), inter-rater reliability in one study was shown to be strong (0.88) (Smyke et al., 2002) and it has been demonstrated to discriminate between children with and without DSED, or RAD (Zeanah et al., 2005; Smyke et al., 2002). However, the DAI was developed for and tested with an institutionalised sample. It is possible that due to the unique nature of the caregiving environments sample biases were present. Furthermore, this tool relies on caregiver report only therefore behaviours across environments cannot be measured.

The Reactive Attachment Disorder and Disinhibited Social Engagement Disorder Assessment Interview (RADA) (Caregiver report) (Lehmann et al., 2020) is the one diagnostic tool which has been updated to reflect DSM-5 criteria, it has good internal validity for assessment of DSED (.88) and it is part of the only published multi-informant assessment package, developed in the UK over time, with community samples of children with DSED and RAD. Along with the RADA, Minnis and colleagues have developed the Relationship Problems Questionnaire- teacher (RPQ-teacher) (Teacher screening) (Minnis et al., 2007) and the Waiting Room Observation (WRO) (child observation) (McLaughlin et al., 2010). These assessments have been used individually and together in clinical practice and in research with community based clinically referred and caregiver referred participants in the UK and Norway, with good reliability and validity (Minnis et al., 2007; Lehmann et

al., 2020; MacLaughlin et al., 2010). We chose to utilise this multi-informant assessment battery in paper 2, to rigorously check that the children referred with symptoms of DSED met DSED diagnostic criteria prior to participation in the study. Table 1 demonstrates the assessments undertaken with each group, and further details are presented in paper 2.

3.4 Training Requirements

CD-J completed the appropriate training courses regarding the DISCO (2 day course), ADOS-2 (5 day research reliability course) and RADA/WRO (1 day course). The SCA provided *in vivo* training on the LIVE, with CD-J, and her colleague, observing and then participating in Live assessments.

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Chapter 4: Using the live assessment to discriminate between Autism Spectrum Disorder and Disinhibited Social Engagement Disorder

Having successfully recruited and rigorously assessed a sample of 10 children with Autism, no maltreatment history, 11 children with DSED and 10 typically developing children the remaining research questions were investigated, (NB. The sample size may vary slightly within each individual paper due to specific requirements of the analysis). Specific to this chapter, the data of 1 child with DSED is not reported due to problems with the video recording rendering analysis unfeasible.

The focus of this chapter is on research question 2: Do current multi-informant 'gold standard' Autism diagnostic tools support differential diagnosis of Autism from DSED, and how does the structured ADOS-2 assessment compare to an unstructured behavioural observation, in this context?

Paper 2:

Davidson, C., Turner, F., Campbell, Gillberg, C., Campbell, S.L., Boyd, S & Minnis.H. (2023). Using the live assessment to discriminate between Autism Spectrum Disorder and Disinhibited Social Engagement Disorder. *Research in Developmental Disabilities.* 134, 1-10

Research in Developmental Disabilities
134(2023) 104415

Contents lists available at ScienceDirect

Research in Developmental Disabilities

Using the live assessment to discriminate between Autism Spectrum Disorder and Disinhibited Social Engagement Disorder

Claire Davidson [a,*], Fiona Turner [a], Christopher Gillberg [a,b], Sharon.L. Campbell [c],

Sheila Boyd [c], Helen Minnis [a]

[a] *University of Glasgow, Academic CAMHS, West Glasgow Ambulatory Care Hospital, Glasgow G3 8SJ, United Kingdom*

[b] *Gillberg Neuropsychiatry Service, University of Gothenburg, Gothenburg, Sweden*

[c] *NHS Greater Glasgow and Clyde, SCA, West Glasgow Ambulatory Care Hospital, Glasgow G3 8SJ, United Kingdom*

A R T I C L E I N F O

Keywords:

Autism Spectrum Disorder

Disinhibited Social Engagement Disorder Observation

Assessment Differential diagnosis

A B S T R A C T

Background: Children with Autism Spectrum Disorder (ASD) and children with Disinhibited Social Engagement Disorder (DSED) may present with similar social problems, despite differing aetiologies, resulting in diagnostic conundrums.

Methods: Thirty children: 10 with ASD, no maltreatment history, 10 with DSED and 10 typically developing children were assessed via 'gold standard' ASD assessments, including the Autism Diagnostic

Observational Schedule (ADOS) and a unique unstructured observation known as the Live assessment. Live utilises a triadic interaction (2 assessors and child), playful teasing and social 'stress' scenarios to increase the social challenge.

Results: The ADOS supported discrimination of DSED from ASD to a degree. Where additional neurodevelopmental problems created ambiguity, the Live assessment was more supportive than the ADOS for unpicking the underlying nature of the social problems

Conclusion: Live supported differentiation between ASD, DSED and other neurodevelopmental problems. The greater social challenge presented by Live exacerbated core problems of ASD and, in DSED, core social skills stood out.

What this paper adds

A unique unstructured observational assessment called 'Live' which offers greater social challenge and therefore may better support discrimination between ASD and DSED.

* Corresponding author.

E-mail addresses: claire.davidson@glasgow.ac.uk (C. Davidson), fiona.turner@glasgow.ac.uk (F. Turner), christopher.gillberg@gnc.gu.se (C. Gillberg), sharon.campbell2@ggc.scot.nhs.uk (Sharon.L. Campbell), sheila.boyd@ggc.scot.nhs.uk (S. Boyd), helen.minnis@glasgow.ac.uk (H. Minnis).

https://doi.org/10.1016/j.ridd.2022.104415

Received 29 August 2022; Received in revised form 21 November 2022; Accepted 29 December 2022

1. Introduction

Autism Spectrum Disorder (ASD) is a neurodevelopmental condition characterised by a "dyad" of impairment, which refers to deficits in a) social communication and social interactions, including impaired social-emotional reciprocity and b) restricted and repetitive patterns of behaviour, including sensory processing problems (DSM-5, American Psychiatric Association, 2013). The UK national clinical guidelines for ASD assessment require evidence from multi-informant assessment, across different environments, and at least one element should involve direct observation of child behaviour (NICE, &, 2011, 2017; SIGN, 2016). The observational tool recommended in the NICE guidelines is the Autism Diagnostic Observational Schedule (ADOS) (Lord et al., 2000). The ADOS can be used with all ages and developmental ability and is designed to be administered by one ADOS trained clinician (Lord et al., 2012). The 2nd edition of the ADOS has been updated to reflect the changes to the diagnostic criteria brought about by DSM-5, and as a tool it is widely used in clinical research, with good reliability and validity for ASD identification (Lord et al., 2000). Consequently, it is generally thought of as the 'gold standard' observational measure for ASD and when combined with thorough care giver report and feedback from school/nursery, in most cases, this seems to be an effective format for completing ASD assessment. However, there are still circumstances in which clinicians are left scratching their heads, asking "is this child presenting with ASD or is there another reason for this child's problems with social relatedness?" (Moran, 2010).

Children with Disinhibited Social Engagement Disorder (DSED) are one group of children which might raise uncertainty amongst clinicians, due to some overlap in diagnostic symptoms (Rutter et al., 1999; Mukaddes, Bilge, Alyanak, & Kora, 2000; Pears, Bruce, Fisher, & Kim, 2010; Moran, 2010; Sadiq et al., 2012; Davidson et al., 2015; Mayes, Calhoun, Waschbusch, & Baweja, 2017).

DSED is one of two DSM-5 disorders believed to be caused by maltreatment (i.e., abuse and neglect), the other being Reactive Attachment Disorder (RAD).

DSED is characterised by indiscriminate behaviours i.e., overfriendliness towards strangers, poor social boundaries and poor relational responses but, unlike ASD, it is considered only in the context of severe maltreatment (American Psychiatric Association, 2013; Turner et al., 2019). The research on DSED is still in its infancy, but current best practice for assessing DSED is a combination of parent report, teacher report and clinician observation. For the observation, an environment such as a clinic waiting room is ideal as the child can be observed in the presence of both their caregiver and strangers. This combination of multi-informant data is used to support clinical judgement regarding symptoms of disinhibition in different environments (Lehmann et al., 2020; McLaughlin, Espie, & Minnis, 2010; Minnis et al., 2007).

Concern regarding overlapping symptoms between ASD and DSED was first highlighted in the 1990s, when Rutter described "Quasi Autism": over-friendliness and lack of social boundaries in children who had experienced severe neglect in Romanian institutions. Yet, all but a few showed marked improvement as they matured, in areas such as language development and cognitive skills post adoption (Rutter et al., 2007; 2010). Quasi Autism began to look more and more like DSED because the children continued to struggle over time with poor social boundaries, despite skills improvement in many other areas.

Sadiq et al. (2012) later found other areas of symptom overlap between ASD and DSED, reporting that children with DSED and/or RAD had social communication problems of severity similar to the ASD group in their use of language in context, rapport and social relationships when assessed via caregiver report on standardised measures of social communication. Mayes et al. (2017), also found that children with ASD and children with DSED had similar problems with social relationships, but they noted repetitive and restricted behaviours were more prevalent in children with ASD (Mayes et al., 2017). Mayes et al., also reported that some children presented with both ASD and DSED. Children who have psychiatric problems in the context of maltreatment have been found to be much more likely to present with complex overlapping problems; they are a group not only at higher risk of having DSED, but a group at much increased risk having neurodevelopmental problems

too (Minnis, 2013; pp. 2, 1268; Dinkler et al., 2017; pp. 2, 1268). It is unsurprising therefore, that ASD and DSED sometimes co-occur (Davidson, Moran, & Minnis, 2022).

Clinicians have driven the need to better understand the interplay between ASD and DSED and a good example is development of the Coventry Grid (Moran, 2010). The Coventry grid is a matrix of social behaviours, demonstrating where overlaps between ASD and DSED may occur, and where differences may be found. The research of Davidson et al. (2015) support the Coventry Grid and together suggest that there may be subtle qualitative differences in the interactions of children with ASD and DSED. For example, the Coventry Grid suggests that both children with ASD and children with DSED have problems with social play. However, children with ASD may be more inclined to play with their toys on their own, or in parallel with others whereas, children with DSED typically try to use the toys to attract attention or to try to take control. Neither group is participating in sharing or cooperative play, as expected in typically developing children, but there are subtle differences in the quality of the problems (Moran, 2010; Davidson et al., 2022).

Despite some symptomatic overlap between ASD and DSED, Davidson et al. (2015) demonstrated that it is possible to differentiate between children with ASD and children with DSED via unstructured observation. Twenty-two of 58 children with a known diagnosis of ASD (38 %), met criteria for DSED on a standardised caregiver assessment of DSED, but from clinician observation of unstructured conversation during the juice break it was abundantly clear in eighteen cases that their impaired social relatedness was characteristic of ASD and *not* DSED. Only four cases were considered 'ambiguous' (3 boys and 1 girl) but ASD, and not DSED, was confirmed via further independent expert review of the unstructured interaction and case records. Of these four ambiguous cases, overlapping Attention -Deficit/Hyperactivity Disorder (ADHD) was a feature in at least one case, exemplifying how additional neurodevelopmental conditions add complexity to differential diagnosis (Davidson et al., 2015).

The above studies demonstrate clinicians' concerns that, in more complex

cases, overlap in symptoms may create problems regarding accurate diagnosis and appropriate case management (Dyer, 2010). The ADOS usually provides a good base from which to determine whether Autism symptomology is present; however, in more complex cases unstructured observation which provides greater social challenge may have a crucial role in supporting accurate differential diagnosis (Davidson et al., 2015, 2022).

One way to increase the social challenge in clinical observation is to decrease the predictability of the assessment through unstructured interaction (Davidson et al., 2015). Another is to mirror more complex everyday social interactions like a 'group dynamic' using two assessors and the third is to introduce 'social stressors' (Davidson et al., 2022). To our knowledge, no assessment of this nature has been used in making differential diagnosis of ASD and DSED.

The purpose of our current study was to investigate the use of an assessment called 'Live' in the context of ASD and DSED diagnostic assessment. The Live is a non-standardised behavioural observation assessment designed and used by the Scottish Centre for Autism (SCA) in all their diagnostic assessments of ASD. We chose to investigate the utility of the Live in differentiating ASD from DSED because it is unstructured, it involves triadic interaction (2 assessors and a child) and assessors introduce 'social scenarios;' all of which decrease predictability, increase the social challenge and can introduce a controlled element of social stress. The assessment is called 'Live' because of opportunity to observe interactions unfolding *live*, as they happen.

1.1. Research objectives

Our objectives were 1. to identify the outcomes of standardised ASD assessments in a sample of children with DSED, 2. to determine whether the increased level of social challenge of the Live helps identify *differences* in the social communication and interaction skills of children with ASD and children with DSED and 3. To determine if the Live assessment better supports differential diagnosis between ASD and DSED.

2. Methods

2.1. Design

We have employed a mixed methods design using cross-sectional data. Due to sample size, we addressed objective 1 via descriptive categorical reporting of the standardised assessments (DISCO & ADOS) and address objectives 2 and 3 via qualitative report and comprehensive case studies.

2.2. Participants

Participants were thirty children: 10 children (7 males: 3 females), with established diagnosis of ASD and no maltreatment history, 10 children (7 males: 3 females), who met DSM-5 diagnostic criteria for DSED on multi-informant standardised measures of DSED and 10 typically developing children (8 males: 2 females), (TD group) with no known child protection concerns. All participants were primary school age (age range 5–11 years) and were group matched by age.

Unlike in ASD, a diagnosis of DSED is not routinely given. This is because labelling the diagnosis, in some cases, may be more harmful than helpful and decisions are made on a case-by-case basis. For example, the clinician may assess and conclude that the child meets DSED criteria and for a foster carer, knowledge of the diagnosis may help them better understand the child's social problems in the context of their early adverse experiences. However, in the case of a biological parent who has worked hard to rehabilitate and is working towards their child returning home, giving a DSED diagnosis may be detrimental to the parent because of the emphasis the diagnosis places on occurrence of maltreatment. The diagnosis may serve to remind the parent of 'fault' when instead it is much more helpful to describe the child's problems with DSED to the biological parent in the context of behavioural symptoms following their difficult start in life. For this reason, it is difficult to recruit samples of children with a *diagnosis* of DSED, but possible to recruit children with DSED *symptoms* and then check the symptoms via the standardised measures, as we did, to ensure they do indeed meet diagnostic criteria (see measures and procedure).

Children in the ASD and DSED groups were referred either by their Child and Adolescent Mental Health Service (CAMHS) clinician or via caregiver self-referral. A recruitment call was advertised on the websites of the following charities: National Autistic Society, Scottish Autism (ASD group), Adoption UK and Scottish Attachment in Action (DSED group). CAMHS clinicians were given the same referral criteria as the charities. For the ASD group, participants were invited who had an established diagnosis of ASD and who were described by parents/caregivers as 'chatty.' We chose to use the parameter 'chatty' to recruit children who were both verbally fluent and motivated to engage with others, as Davidson et al. (2015) found that it was 'actively engaging' children with ASD who were more likely to raise concerns around differential diagnosis. Nine of the 10 children with ASD were referred by caregivers and 1 child via clinician referral. Lack of child protection concerns were established via health board electronic records. For the DSED group, par- ticipants were invited who were described by parents/caregivers as 'chatty *and* overfriendly with strangers.' The latter being the core feature of DSED (American Psychiatric Association, 2013; Lehmann et al., 2020). A history of childhood maltreatment was established in the DSED group either via referring clinicians or foster carer/adoptive parent report. Six of the ten children in the DSED group were caregiver referred and four via clinician referral. The TD group were caregiver referred from a handout given when they attended for a different study. Lack of child protection was established via health board electronic records.

2.3. Measures

The following measures were used in this study. Please refer to the references for further information regarding validity and reliability data.

Reactive Attachment Disorder and Disinhibited Social Engagement Disorder Assessment Interview (RADA) (Lehmann et al., 2020) is the DSM-5-compliant version of the Child and Adolescent Psychiatric Assessment for Attachment Disorders. It is a semi-structured interview for the diagnosis of DSED and completed by parents/caregivers.

The Teacher Relationship Problems Questionnaire (Teacher RPQ) (Minnis et al., 2007) is a 10-item screening questionnaire for symptoms of DSED completed by teachers.

Waiting Room Observation (WRO) (McLaughlin et al., 2010): is a structured 19-item observation of child behaviour with paren- ts/carers and a 'stranger' carried out in a clinic waiting room. The role of the stranger, a trained rater in the WRO, is to record the interactions of the child with their primary caregiver, the child interactions with the clinician on greeting them, and interaction or social approach by the child to the stranger. The first 6 questions focus on disinhibited behaviour towards the stranger. In this study, the stranger was a research colleague who was otherwise uninvolved with the study and unknown to the participants.

Diagnostic Interview for Social and Communication Disorders (DISCO) (Wing, Leekam, Libby, Gould, & Larcombe, 2002) is a standardised in-depth interview for the diagnosis of ASD, although it touches on symptoms of other neurodevelopmental disorders and can be used across the age span. It is completed with primary caregivers.

Autism Diagnostic Observational Schedule-2 (ADOS-2) (Lord et al., 2000) is a standardised observational tool used to diagnose ASD across all ages and developmental stages. The module administered is dependent on the developmental language skills of the child. If the child is fluent, (as is most relevant to this paper), module 3 is administered which involves fourteen tasks. These are a mix of prop/play based tasks and structured questions. The assessor can use "social presses" throughout to elicit conversation. The aim is to assess absence or presence, and severity, of the core features of the dyad of impairment.

Live Assessment: The Live assessment is a triadic (2 assessors and the child) unstructured assessment designed to increase unpredictability and greater social challenge. Two assessors interact with the child individually and together for a duration of 45–60 min, just as one might during group interaction. For purposes of the research, we shortened the Live to 30 min. The assessors use informal

conversation, games, banter (playful teasing), direct questioning and create social scenarios for the child to respond to. The aim is for the experience of the child to feel natural, so the assessors are free to introduce the stages of the Live spontaneously in response to situations and in whatever order, while gradually working up to the greatest social stress the child can tolerate. Often stages are re- visited to assess consistency and to observe how a change in dynamic can affect the behaviour- for example, the quality of 3-way conversation when the child is calm versus 3-way conversation following an active game. The Live assessment, in contrast to the ADOS, is a non-standardised tool which is not categorically or numerically scored therefore it cannot be quantitatively analysed. As a behavioural observation it is an invitation to simply observe the range of skills and behaviours. The outcomes are analysed descriptively and using clinical judgement. The information gathered in the Live can be used to contribute to a formulation or categorical diagnosis, if required. To demonstrate how one may use the Live, we have provided additional details in Table 1.

Table 1

Stages of the Live Assessment

Stage of the Live	*Methods Used to Elicit Behaviours*
Meeting a stranger	Observation of the child meeting the 2nd assessor, who has not previously met the child. The 2nd assessor will immediately attempt to engage in conversation with the child. The lead assessor (familiar to the child) will sit back, observe and then join in when it feels appropriate.
Informal 3-way Conversation Includes observation of: - Non-verbal communication, body language and gestures. - Rapport - Reciprocation	This begins with discussion about the child's favourite toy or interest (they can bring it along). The assessors will offer information about their own interests, ask questions and introduce new topics; observing the interest the child takes, whether they share additional information and ask questions of their own. *The 3-way conversation occurs throughout the assessment and is brought into games as well, in order to increase attentional demand. It is not the case that once completed the section is over.

- Ability to interact in a 'group' dynamic	
Imagination and creativity	Includes observation of: - Free play and joint play with toys. - Conversation while playing with the toys (to increase attentional demand) - Giving the child some random toys and asking them to create rules for a game for all to play, without warning. - Guess what game (use of an object like a ball to pretend it is something else. The person with the ball gives clues about what they are thinking of. The child and assessors take turns at both guessing and giving clues). - Role play (without warning, the child is asked to be a teacher and the assessors the children. One assessor plays role of a 'good' child and the other a 'naughty' child.) - Assessors tell fantastical stories as a 'wind up.'
Empathy and emotions	- Observation during conversations for use of emotional language or an emotional response. And, asking "how did they feel? Or "how did you feel?" either in conversation or during any of the scenarios below: - Assessor tells a story about hurting herself (e.g., last night I burnt my finger…) allowing pauses for the child to respond/predict what happened next.
Stage of Live	*Methods Used to Elicit Behaviours*
Empathy and emotions continued	- Assessor pretends that they have hurt themselves/lost their voice/foots gone to sleep etc. - Assessor tells a story where others are affected by something e.g., they were at a coffee shop with their dog and their dog ate a child's ice cream. - Assessors have a pretend argument/tease each other. - in the turn taking game (passing the ball to each other), one assessor deliberately leaves the other assessor out of the game and/or the assessors make the child miss a few turns.
Friendships/other relationships	- 3-way conversation. - Assessors asks child directly about friends. - Assessor hints/talks about something exciting involving a friendship/relationship (e.g., an assessor is soon getting married). - Assessor pretends that she has a problem relationship (e.g., assessor is worried that her boss is annoyed at her).
Banter	- Banter is back and forth exchange, involving teasing, sarcasm and 'tongue in cheek humour.' (Adapted for age).

	- Assessors tell fantastical stories as a 'wind up.' - Use of metaphors directly to the child (was she winding you up/pulling your leg?) and asking them to explain.
Turn taking, following rules and adapting to changes and winning/losing.	- Gently throwing a ball to each other in a circle (start with a turn taking rule, then an assessor will 'forget' and break it. Occurring more than once). - Setting up and playing Jenga (assessor will make up new rules and adapt the rules over time, often making an assessor the winner).
Sensory	Directly asking the child about any sensory sensitivities or interests. - One assessor will loudly empty the Jenga blocks on the ground, unexpectedly, while the other assessor is chatting to the child. - Observation throughout.

2.4. Procedure

Research ethics approval was obtained from the West of Scotland Research Ethics Committee and the lead author achieved funding from the Castang Foundation, (UK), [LAY1-WS_LEGAL.FID1730709] to complete the research.

DSED assessment: The RADA was completed via telephone interview by the lead researcher with primary caregivers of children referred to the DSED group. Additional information regarding disinhibited behaviour was gathered via the Teacher-RPQ which was posted and returned via S.A.E. The Waiting Room Observation was conducted in the research clinic prior to the first meeting. Only children who's DSED symptoms met diagnostic criteria were admitted to the study.

Following admission to the appropriate group, caregivers were invited to complete the DISCO assessment at a home visit. For most cases the DISCO was completed by the lead researcher, who is a Speech and Language Therapist and trained to administer both DISCO and ADOS; two DISCOs were administered by another trained Speech and Language Therapist. Within 1–4 weeks of the DISCO assessment, the child was invited to the clinic for ADOS assessment (no child in any of the groups had participated in an ADOS assessment within 6 months of the study). The lead researcher was still a stranger to the child at this

point. Within 1–4 weeks of the ADOS, participants were invited back to complete the Live. The Live assessments were administered by the lead researcher (assessor 1) and her colleague, a Health Psychologist with both lived experience as a parent of a child with ASD and a research interest in neurodevelopmental conditions (assessor 2). Both assessors were trained to use the Live by the SCA.

As the lead researcher completed all the standardised assessments and the Live, she was not blind to the diagnosis of the participant groups. For the Live, this was beneficial as the lead researcher was able to 'test out' problem areas in a different assessment dynamic. In contrast, the 2nd assessor of the Live assessment was blinded to the diagnosis of the groups, which helped bring a fresh perspective.

All child assessments were video recorded and after the Live, assessor 2 gave immediate feedback to the lead researcher. Assessor 2 also separately watched 50 % of the video recordings to support analysis of the Live. All diagnostic decisions, including identification of additional neurodevelopmental conditions, were made collaboratively by the lead researcher and the research supervisor, a Consultant Psychiatrist with extensive expertise in attachment disorders and neuro- developmental conditions, using *all* the multi-informant study data.

3. Results

3.1. Neurodevelopmental complexity

On completion of the assessments, we were confident that the TD group did not present with any undiagnosed neurodevelopmental problems. In the ASD group, two of the ten children had a pre-existing diagnosis of ADHD, and both were being treated via ADHD medication. We found significant *undiagnosed* neurodevelopmental complexity in the DSED group, and we referred two cases to CAMHS for a full ADHD assessment, as we were confident their symptoms were significant enough to meet diagnostic criteria. However, ADHD traits (fidgetiness, hyperactivity or impulsivity) were reported by caregivers in *all* DSED cases and were observed, to varying

degrees. Two children with DSED also warranted diagnosis of co-occurring ASD. The final DSED group could be described as six children with DSED only (all of whom had traits of ADHD), two children with DSED+ADHD and two children with DSED+ASD.

Objective 1: outcomes of ASD assessments

TD group: None of the TD cases met diagnostic threshold for ASD on the DISCO or ADOS. Appropriate social communication, interaction, understanding and use of humour and imagination were demonstrated in the Live assessment.

ASD group: All children in the ASD group, who already had an established diagnosis of ASD, met diagnostic threshold for ASD on *both* the DISCO and ADOS. Core deficits in social communication, reciprocal interaction, imagination and inability to engage in subtle humour were observed in the Live.

DSED group: For clarity, we have described the outcomes of the ASD assessments in this group in Table 2 below.

Table 2

Outcomes of ASD Assessments in the 10 DSED Cases

	DISCO (parent/caregiver diagnostic interview)	*ADOS* (structured clinical observation)	*Live* (unstructured clinical observation)
DSED only (With traits of ADHD) (n=6)	5/6 cases met diagnostic criteria for ASD	1/6 cases met diagnostic threshold for ASD (See case study 2).	All 6 cases presented with core communication and interaction skills, in addition to their disinhibited behaviour, suggesting DSED and not ASD.
DSED+ADHD (n=2)	Both cases met diagnostic criteria for ASD	Both cases met diagnostic threshold for ASD.	Both cases presented with core communication and interaction skills, suggesting DSED and not ASD and ADHD severity that warranted diagnosis.

DSED+ASD (n=2)	Both cases met diagnostic criteria for ASD	Both cases met diagnostic threshold for ASD.	Both cases presented with impaired communication, imagination and social problems indicative of ASD and presented with disinhibited symptoms that were unusual in ASD and indicative of DSED

Seven of the eight children *without* ASD in the DSED group, met diagnostic threshold for ASD on the DISCO. Poor eye contact and gestures, friendship and empathy/relational problems, need for routines and sensory issues were the main overlapping concerns reported. The two children with DSED+ASD also met diagnostic threshold for ASD on DISCO.

The ADOS helped to differentiate DSED from ASD in 'clear-cut' cases. ASD negative results were found in 5/6 DSED only cases. But ASD positive results were found in both DSED+ADHD cases, and in a DSED only case with more apparent ADHD traits. The problem areas on the ADOS were rapport, social overtures and either sensory behaviours or repetitive themes in their communication.

Both cases of DSED+ASD scored positively for ASD on the ADOS.

3.2. Objectives 2 and 3: live assessment case examples

The qualitative information and the level of detail offered by the Live assessment were extremely supportive in understanding similarities and, more importantly, differences in the social communication and interactional skills of children with ASD and children with DSED. The greater social challenge of the Live better helped to discriminate between ASD and DSED because it more clearly elucidated the presence or lack of communication and interactional skills and more easily demonstrated the relative impact of additional neurodevelopmental conditions like ADHD (see case studies). Following the Live assessment, we felt more confident in our formulation that some of the children in the DSED group were experiencing additional social interactional problems due to undiagnosed ADHD (2 cases).

We present two comprehensive case studies below to demonstrate a) the differences in social communication and interaction between children with ASD and DSED (objective 2) and b) how the greater social challenge and unpredictability of the Live better supported discrimination between ASD and DSED (objective 3), including via clearer understanding of the impact of neuro- developmental complexity.

We have described the Live assessment in the "sections" presented in the methods (Table 1), but, in clinical practice, the order may differ as assessors introduce the elements in a fluid way, responding to arising opportunities.

Case Example 1*: Molly** (*all names are pseudonyms). Molly had an established diagnosis of Asperger's Syndrome and no child protection concerns. We picked this case from the ASD group because her symptoms presented more subtly.

Live section 1: initial entry to room and meeting stranger(s).

Molly enters the room with the lead researcher (assessor 1), who she has met during the ADOS assessment. When assessor 2 enters the room, Molly says hi and starts chatting, showing no shyness or reticence.

Live section 2: informal three-way conversation.

Molly answers questions and spontaneously offers additional and, for the most part, relevant information, but tends to relate it back to her or her brother's experience. Molly's responses do not form a natural flow that keeps the conversation going when the con- versation relates to things separate to her. The feel of the rapport was relatively easy, but the conversation could become stilted if the assessors didn't support Molly with questions. Molly used an unusual/formal tone or phrasing many times throughout the assessment, particularly when she was feeling uncertain. For example, when asked how she felt about her teacher leaving: "I do not know what she is doing, so please do not ask."

Molly rarely used gestures other than occasionally nodding. She did not use

pointing or descriptive gestures while in conversation. When spoken to, she made eye contact but tended to drop her head, not maintaining it, and never at any time during conversation did Molly show the subtle skill of shifting eye gaze from person to person.

Live section 3: imagination and creativity.

With increasing attentional demand through the introduction of toys and shifting positions of the assessors, Molly's ability to use eye contact greatly reduced. She tended to only look at one assessor when speaking, as opposed to turning to face the other even when they spoke to her. She also found it difficult to play and chat at the same time. In free play, Molly did not spontaneously create imaginative play, but followed the assessors' lead. When assessor 1 presented a few toy animals and a ball and (with no prior warning) asked Molly what the rules of the game were (meaning Molly needed to make up a game) Molly really struggled and instead employed a strategy to avoid the task. Following our persistence and encouragement, she came up with a simple idea that lacked imagination.

Live section 4: empathy and emotions.

Chatting with Molly about her teacher leaving offered an opportunity to assess her understanding of other's emotions and her use of emotional language. Molly uses emotional, but formal, language to describe how she is feeling, "I feel pretty sad, but I also feel happy for her because I think she is going to have another great future." She did not use the same degree of emotional language when discussing others' feelings. She did show empathy for others, "I think she is going to be okay (the teacher) as we gave her lots of encouragement." When responding to scenarios created to explore empathy, Molly tried to solve the issue and tried to relate to the person. Although she seemed to understand what the person might be feeling, she appeared to find it more difficult to express it. Instead, her way of expressing it was to tell the assessors about something that happened to her that raised a similar feeling. For example, during a game of throwing the ball to each other, assessor 1 unexpectedly switched the turn taking and began to only throw the ball to Molly. Assessor 2 immediately

displayed an upset facial expression and body gesture. Molly responded by offering that assessor her turn (problem solving). Assessor 1 asked Molly how assessor 2 was feeling and after a moment of silence, she responded with, "I've felt that before, but even worse," then told the story of what happened to her. It created a feeling that everything resorts to her (even if that was not her initial intention).

Live Section 5: friendships.

Molly used broad statements to describe her relationships and did not spontaneously give details about friends. For example, she stated that she didn't get to say goodbye to 'everyone' on her last day of school. Assessor 2 asked Molly if she has friends and she replied, "yes, lots," naming a list with little detail and no elaboration of the nature of each friendship. When assessor 1 asked Molly if assessor 2 knew Molly's friends, she went quiet (remember assessor 1 and Molly have discussed friends in the ADOS, but assessor 2 was a stranger to Molly). Molly appeared confused, but then without answering the question, carried on and listed the names. We suspected that she possibly knew that assessor 2 didn't know her friends but didn't know how to respond and was keen to please.

Live section 6: banter.

Molly was not able to join in with banter/playful humour and responded to our fantastical stories as if they were real, even her expression sometimes suggested otherwise. When told, "I'm only joking, or I'm winding you up," Molly replied "oh, ok." She showed subtle signs of stress such as becoming silent and not responding for a second or two. By the assessment end, it was apparent that Molly was very sensitive to things that she could not do and was quick to employ strategies to manage the situation. One strategy was to answer with something on the same topic but that was not a direct response to the actual statement. For example, Molly mentioned being good at animal noises and she did an exemplary impression of a dolphin (a known special interest). Assessor 1 said, "you are better than a real dolphin." Molly did not understand the humour and responded literally, "I could never be a real dolphin." Pushing the

subtle humour a bit further, she followed up Molly's response with, "no but you could be a turkey." Again, Molly didn't understand and went quiet before eventually saying, "if it was a flying competition a turkey would win, but not a farm turkey because they can't fly." Molly was keen to provide an answer, but not understanding the dynamic in that moment, she took the subject of the chat, related it to something that she did know about and thereby re-directed the conversation. Interestingly, Molly could explain the meaning of metaphors like 'I'm winding you up, I'm pulling your leg,' and she spontaneously told a joke. It seemed that Molly could understand simple humour intellectually, or when rehearsed, but she was not intuitively able to join in or share subtle humour, like simple sarcasm, in back-and-forth exchanges.

Live section 7: routine/turn taking/winning and losing.

Molly did not insist on rules and accepted any changes, but passively. For example, in the 'pass the ball' game Molly allowed the assessors to continually switch the turn taking order around and did not protest when they deliberately left her out, yet her facial expression demonstrated something akin to confusion/surprise. During all the games, Molly took turns appropriately, tried to comfort the assessors when they didn't win and was able to adapt when we deliberately changed the rule. During conversation, we learned that Molly does like to obey certain types of rules. When asked her response to someone jumping in the puddles in the playground, she replied, "I would tell the teacher. I think that would actually make it a little bit serious, as parents won't want their children going home wet."

Live section 8: repetitive behaviours and sensory behaviours.

No obvious repetitive motor movements, sensory sensitivities or vocalisations were apparent. When we asked her about sensory issues, she told us about her strong dislike of certain smells, describing how she needs to go to another room if her brother is eating chicken curry.

Live section 9: physical and personal boundaries.

During the whole assessment, Molly maintained appropriate physical boundaries, unlike many of the children with DSED. She did not encroach on

personal space and even asked assessor 1 first, “would you like a hand (holding out her hand)?” before she offered to help the assessor stand up (assessor was pretending her foot had gone to sleep). Molly was not shy or hesitant and quick to offer personal information about herself, but the difference between this and DSED was that she was not overfriendly in a way that felt invasive and her main focus was herself and her own experiences. She did not appear to ‘try to get to know’ the assessor or enquire about the examiner in personal detail. Furthermore, her social communication and interest was somewhat stilted when unprompted and had a learned quality. Finally, she did not demonstrate cloying or clingy behaviour.

Case Example *2:* Liam* is an 8 year old boy in the *DSED only* group whose case was more ambiguous; unlike the others with DSED only, he met the ASD threshold on the ADOS as well as the DISCO.

Live Section 1: initial entry to room and meeting stranger(s).

Liam entered the room with assessor 1. He was confident and showed no hesitation. Assessor 2 entered a few minutes later and initiated conversation with him. He showed no wariness towards her, despite having never met her before.

Live Section 2: informal 3-way conversation.

An easy conversation about Liam’s favourite toy was established. Rapport was easy and the conversation naturally expanded as Liam spontaneously offered information, asked for information about the assessor and was not thrown by our deliberate interruptions. There was a sense of social connection. While chatting, Liam smiled, nodded, used pointing and descriptive gestures in time with his speech. He used appropriate eye contact, including shifting his eye gaze between assessors *while* he was talking to them. Liam’s appropriate use of non-verbal communication remained consistent throughout the whole assessment, even when his concentration broke down and verbal narratives became poorer.

Live Section 3: imagination and creativity.

In the ‘guess what game’ (see Table 1), Liam was quick to suggest that he go first and he spontaneously set rules. He appeared to really enjoy this game. He demonstrated good imagination skills in his ability to pretend, in giving us clues and making appropriate guesses to our clues. When assessor 1 deliberately made a wrong guess of a banana, he picked up some slime, modelled it into the shape of a banana, and held it up saying, with a telling voice, “it would look like this if it was a banana.”

Live Section 4: routines/turn taking/winning and losing.

As we began the ‘guess what’ game, Liam became more excitable and fidgety. He seemed to like being in charge and had a desire to be the one making and changing the rules, doing so to fit better with his agenda. For example, he quickly decided because we had to ‘give in’ that meant that he should take another turn. Assessor 1 did not allow Liam to take another turn, which provoked a DSED type behaviour. Liam approached her, encroaching her space, and began to bat the ball in her hand saying “no,” and repeating his plan to take the turn. Liam’s desire to be in charge had a controlling quality to it and was consistent throughout all the games we played. At a later stage when he did not get his own way, he violated assessor 1’s personal space and thoroughly messed her hair up. It was not overly aggressive in nature but highly inappropriate and had an impulsive or over-stimulated quality. Although Liam preferred to be in charge, he seemed to thrive off our participation, which is different to the feel in ASD.

Live Section 4: empathy and emotions.

Assessor 1 pretended the slime had caused a cut on her finger to sting (uses a clear vocal gesture, ouch, and non-verbal gesture - rubbing finger and making ‘sore’ facial expression). Liam immediately stopped his conversation with the other assessor and looked at assessor 1. His facial expression appropriately changed from smiley to serious, and he glanced towards assessor 2 with a look of uncertainty. He then turned back to assessor 1, “I better take your turn then.” Despite Liam’s initial concern there was a lack of empathy to her “injury” or interest regarding how she had cut her finger. Instead, he’d tried to use the situation, almost in a social way (as if it was of benefit to assessor 1), to get

what he wanted. Assessor 2 asked Liam how assessor 1 would have felt when she cut her finger and he was able to say that she'd felt upset.

Liam was still excitable, restless and fidgety and was constantly moving out of his seat and rolling about the floor; especially when we played games. The more excitable Liam was the more disinhibited he became. For example, towards the end of the assessment, Liam was swinging his legs and began to repeatedly 'kick' assessor 1's feet. When she reacted to it saying, "Liam is playing drums with my feet" he laughed and then hit her feet deliberately with a wide grin that made him look overstimulated. Liam showed no sense that this was inappropriate. Assessor 1 pretended that his next kick hurt and put her face in her hands and acted sad. Liam leaned close into her face and pulled a grin. He once again showed little empathy for the situation and did not apologise or show any remorse.

Live Section 5: banter.

Liam told us a story about his aunt's dog jumping up on him and assessor 2 offered her own story about a bear jumping up on us in her garden. Liam immediately looked at assessor 1 saying, "is that how you got the cut on your face?" (There wasn't a cut). At a later point in the assessment assessor 2 re-visited this by asking, "when we were talking about the bear do you think we were pulling your leg?" to which Liam replied "yeah." Liam later demonstrated that he could use metaphors himself. When describing a boy jumping on a trampoline and landing in a pit of balls at the soft play he said, "landed in a pit of…" and hesitated long enough that assessor 1 was able to jump in and finish the sentence suggesting that the boy landed in a pit of fire. Liam immediately looked at assessor 2, rolled his eyes and cheekily said, "has she lost her mind?" His response and body language meant that he included her in his slightly sarcastic joke.

Live section 6: friendship.

Liam named some friends but by this point in the assessment he was constantly distracted and more fidgety – clearly not engaged at all in telling us about his friends. When we tried to ask him about the birthday party and his friends, his

narrative was confused and unclear, a contrast to his easy chatting at the beginning of the assessment. It appeared that after the first game he became excitable and after the 2nd game, he was unable to 'come down' or sit down and return to chatting. Because of this timing, it is difficult to know whether Liam's disengagement was a direct result of poor concentration and hyperactivity or whether he was feeling stressed about the topic. It could also be a combination of both, although in other situations where Liam seemed stressed, he tended to violate physical boundaries (taking the ball out our hands and throwing it away, messing up the assessor's hair, getting in both assessor's faces, shouting in their face etc.) During the friendship conversation he was up and out his seat constantly and was focused on trying to return the guessing game. It had more of a loss of concentration and restless feel along with a controlling element, especially as he tried to turn the conversation into a game, telling us to guess the names of his friends rather than telling us about them. In moments like this, we could see how Liam's restlessness and hyperactivity interrupted the social rapport and his communication, but when more settled this was not the case.

In addition to the detailed findings given in the case studies we were able to identify from the Live some general trends that helped us to discriminate between children with DSED (including DSED+ ADHD) and children with ASD (including DSED+ASD). These were as follows:

- Children with DSED appear better able to engage in more 'complex' humour compared to children of the same age with ASD.
- The play of children with DSED tends to be more creative, spontaneous and they want you involved compared to children with ASD.
- Children with ASD are generally more object focused or governed by their own experiences.
- Children with ASD and children with DSED demonstrated 'stress' differently. Children with ASD tended to freeze, become unresponsive or verbally express anxiety when coping with the unpredictability and the 2-assessor dynamic. Children with DSED often became disinhibited through physical touch and close physical proximity.

- Children with ASD typically did not show shyness with a stranger (1st meeting/ with 2nd assessor) but children of 7/8 years plus were less likely to touch the assessors or get too physically close.
- Obsessive, controlling and oppositional behaviours, particularly, in the cases of DSED+ADHD and the child with ADHD traits added further complexity, but in the Live core communication and interaction skills could be uncovered.

4. Discussion

Our first objective was to identify the outcomes of standardised measures of ASD with a sample of children with DSED, and we found that via parent/caregiver report (DISCO) children with DSED were described as having similar problems with social relatedness and routines as children with ASD. The ADOS, in comparison, was beneficial in differentiating ASD from DSED, but only in cases where additional neurodevelopmental complexity was not present. Our second objective was to determine whether the unstructured nature and greater social challenge of the Live helped to identify *differences* in social communication and interaction between ASD and DSED and found these factors to be a significant strength in this regard. The free-flowing dynamic of the Live supported developmental social skills, where present, to stand out, *despite* co-occurring neurodevelopmental problems and exacerbated any existing social impairment. We believe an assessment, like the Live, may better support clinicians with accurate differential diagnosis between ASD and DSED.

Our findings are limited by a smallish sample size (n = 10 per group) and most participants with DSED being self-referred (6/10), which may be less representative of the complexities seen in CAMHS. Yet, two of the self-referrals were still ambiguous due to un- diagnosed ADHD in one case and ADHD traits in the other.

An unstructured and socially challenging observation like the Live could also be used alongside the Coventry Grid (Moran, 2010) to support clinicians with differential diagnoses between ASD and DSED. The domains of the Coventry Grid have recently been found to have an evidence base (Davidson et al., 2022) and our findings provide further evidence of *differences* between children with ASD and

DSED in many of the Coventry Grid domains. Furthermore, we found that focusing on core developmental communication, social interaction skills and imagination during the Live, as well as broader relational issues, was key. In fact, we found that observed *use* of social communication skills helped to support discrimination of DSED from ASD. This contrasts to the findings of Sadiq et al., (2012), but their use of parent report only could account for the discrepancy. We did observe some problems with rapport, also reported by Sadiq et al. (2012), but mainly in the children with DSED+ADHD/ several ADHD traits and with greater impact on the ADOS outcome than the Live.

The ADOS assessment was able to discriminate between ASD and DSED in 5 out of 8 of the DSED cases where ASD was not present. Yet, when additional neurodevelopmental conditions were present the ADOS outcome was ambiguous. Our hunch is that the more structured and seated nature of the ADOS exacerbated undiagnosed ADHD problems, which then interfered with core social skills. The false positive ADOS findings perhaps serves as a 'pause for thought' for services which tend to rely *solely* on categorical measures, including ADOS, because neurodevelopmental complexity can be considered 'the norm rather than the exception.' This is true for both children with pre-existing neurodevelopmental conditions (Gillberg, 2010) *and* in children who have a history of maltreatment (Minnis, 2013; pp. 2, 1268; Dinkler et al., 2017).

Finally, with DSM-5 re-defining ASD as a dyad, excluding impaired imagination in the criteria, and the upcoming ICD-11 following suit, our findings are timely. Social communication and relationships are undoubtably inter-related but when combined as 'social affect,' or social-emotional reciprocity, particularly in categorical assessments, perhaps some of the qualitative differences between ASD and other conditions with social relatedness problems become less clear. For clinicians faced with the ASD v DSED conundrum, perhaps shifting focus back to core developmental social communication, imagination, and interactional skills may help ambiguous cases feel less daunting.

5. Conclusion

The findings from the current study adds weight to the small body of evidence

suggesting it is possible to discriminate between ASD and DSED (Moran, 2010, Davidson et al., 2015, Mayes et al., 2017; Davidson et al., 2022). Standing on the shoulders of these studies, our new study findings suggest the benefit of unstructured observational methods, particularly in ambiguous or neurodevelopmentally complex cases. We found that children with DSED or DSED+ADHD demonstrated core developmental communication and interactional skills (eye contact and gaze, back and forth chat and humour, informational and descriptive gestures, spontaneous social interest, imagination/creativity and reciprocation), that were often not apparent in the children with ASD, or DSED+ASD. These skills were apparent even when other relational issues such as poor empathy, controlling behaviour and disinhibition/poor boundaries were apparent. The elements of the Live assessment which best aided differentiation were unpredictability (unstructured and free flowing/spontaneous), greater social challenge (triadic interaction) and the social stressors; for the children with ASD, difficulties with core social communication and interaction were exacerbated by the social demands and therefore more apparent, and for children with DSED, they intuitively relied on their social skills to manage it, which made these skills stand out.

CRediT authorship contribution Statement

Claire Davidson: Conceptualisation, methodology, investigation, analysis, writing, writing- review and funding acquisition. Fiona Turner: Investigation, analysis, review of writing. Christopher Gillberg: Conceptualisation, supervision, review of writing. Sharon Campbell: methodology, review of writing. Sheila Boyd: methodology, review of writing. Helen Minnis: Conceptualisation, meth- odology, supervision, review of writing.

Declarations of Competing Interest: None.
Data Availability: The data that has been used is confidential.

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Reflections on Paper 2

In paper 2 we were able to demonstrate that observation, rather than caregiver report, was more supportive in differentiating Autism from DSED. Furthermore, the unstructured and more socially challenging Live assessment better helped elucidate both the skills and difficulties of the different groups of children when cases were of greater complexity. The following points are of note.

- The standardised caregiver report diagnostic measure of Autism, the DISCO, did not help to differentiate between Autism and DSED.

- The standardised structured diagnostic observation measure, the ADOS-2, helped to discriminate between Autism and DSED, to some degree. Straightforward DSED cases, i.e., children who presented with only DSED did *not* meet diagnostic criteria for Autism on the ADOS-2. However, children who presented with DSED *and* moderate to severe ADHD symptoms scored positively for Autism on the ADOS-2, as did children with DSED and Autism. For these cases, we were not able to discriminate using the current gold standard Autism diagnostic tools. Given the potential of this finding add to the ambiguity for clinicians used to only using ADOS for the clinical observation part of the assessment, this is worthy of further investigation with larger samples of children with DSED.

- We found that the unstructured and more socially challenging Live assessment better supported differential diagnosis between Autism and DSED. Increasing the unpredictability and pushing the child socially meant that children relied on social skills, where present, and so these were more observable. Furthermore, core social communication difficulties inherent to Autism were exacerbated.

- The unstructured and free flowing dynamic also helped to identify core ADHD symptoms. The 'stress' element of the Live further helped to identify the 2 cases of co-occurring DSED and Autism, as the disinhibited responses to stress were more in keeping with DSED than Autism, but Autism symptoms were nevertheless demonstrated throughout.

- It is recognised that the assessors involved in the Live assessment in paper 2 are both very experienced in Autism assessment, in an ESSENCE approach to holistic assessment and have expertise in maltreatment associated problems. The findings reported are still nuanced and not all

clinicians may feel they have the expertise to engage with an unstructured assessment approach like the Live without further guidance and training. Testing the feasibility of the Live assessment in clinical practice for holistic neurodevelopment would be a reasonable next step.

- The findings from paper 2 suggest that differential diagnosis between Autism and DSED is possible, but the differences can be subtle. However, many of the social communication and imagination differences identified between Autism and DSED are areas which are already known as core difficulties in Autism, but overlapping neurodevelopmental conditions can add complexity; this suggests further that involving clinicians with vast and varied experience in Autism and holistic neurodevelopment is perhaps key.

In this paper we have made some progress towards understanding how different forms of assessment tools support differential diagnosis and have gained some understanding regarding possible differences between Autism and DSED, in this case sample. Social communication problems known to be present in Autism were less apparent in DSED but there is still much to learn. For example, receptive and expressive language skills directly relate to how individuals communicate socially but this has not been explored in children with DSED. Furthermore, we know that one study, (Sadiq et al, 2012) found that pragmatic language – a sub-domain of the broader facet of social communication- was as impaired in children with DSED as children with Autism, but this was only assessed via caregiver report.

Chapter 5: Autism and Disinhibited Social Engagement Disorder: Overlaps and Differences in language and communication skills.

The language skills of children with DSED remains an under-researched area and these have not been examined in comparison to children with Autism. In addition, further investigation of the pragmatic language skills of children with Autism compared to children with DSED is required to better understand if there are any key differences which may support differential diagnosis. In the third paper, we use multi-modal methods to investigate all areas of language and communication, in the case study sample, to address the literature gaps and investigate these skills in depth.

Paper 3 (submitted to Research in Autism and is under review)

Davidson, C., Gillberg, C., McCool, S., Elder, B., Minnis, H & Lowit. A. (2023) Autism and Disinhibited Social Engagement Disorder: Overlaps and differences in language and communication skills.

Abstract

Background: Paediatric and child mental health clinicians are concerned about differential diagnosis of autism from Disinhibited Social Engagement Disorder (DSED). Both groups of children present with similar social relationship difficulties, yet DSED is associated with maltreatment and autism is not. Language and social communication problems are common in maltreated children, but DSED is rarely examined within the maltreatment literature and this gap in knowledge exacerbates the clinical dilemma. **Methods:** To determine areas of differentiation, we aimed to explore receptive language skills using standardised assessments in 138 children: 43 children with Autism; 24 children with DSED; 37 typically developing (TD) children and aimed in 30 children (3 groups of 10 - Autism, DSED and TD), to additionally explore expressive and pragmatic language skills, including analysis of conversational speech. **Results:** When verbal IQ was accounted for in autism, receptive vocabulary did not significantly differ between groups, but children with autism had poorer scores when semantic reasoning was required. Children with DSED, however, scored more poorly than those with autism and the TD group regarding expressive language (standardised and conversational assessment). Caregiver screening of pragmatic language suggested similar difficulties in autism and DSED, *but* Speech and Language Therapist (SLT) analysis of pragmatics in conversational speech demonstrated some key differences. **Conclusions:** Maltreated children may present with DSED, and all professionals should be aware of the increased potential for language and communication difficulties in this group. Children with DSED, however, may retain some pragmatic skills which are not present in autism, thus, SLTs have a key role in supporting differential diagnosis of autism from DSED.

1. Introduction

Children who have experienced severe early maltreatment (abuse/neglect) are at higher risk of cognitive, language and social developmental difficulties (Culp et al., 1991; Law et al., 1992; O'Connor et al., 2000; Smyke et al., 2009; Cicchetti et al., 2016), but are also at higher risk of one of two maltreatment associated disorders, Reactive Attachment Disorder (RAD) and Disinhibited Social Engagement Disorder, (DSED) (Minnis, 2013). For this paper, we are interested in Disinhibited Social Engagement Disorder, (DSED), because some of the core diagnostic symptoms of DSED overlap with diagnostic

symptoms of Autism. Yet, unlike DSED, Autism is not associated with maltreatment (Turner et al., 2019).

DSED is characterised by indiscriminate friendliness with strangers, lack of reticence in social approach and poor social boundaries, but it is diagnosed *only* in the context of maltreatment (abuse/neglect) (DSM-5). Pre-DSM-5, some studies only used the umbrella term RAD for both RAD (previously inhibited subtype) and DSED (disinhibited subtype) and did not differentiate between sub-types. For clarity, we use the moniker $DSED^{RAD}$ when referring to these still relevant studies. This is justified because evidence has shown that DSM-5 RAD is rare on its own, within the population, (Minnis et al., 2013; Zeanah et al, 2000) and because DSED, unlike RAD, tends to persist beyond infancy (O'Connor et al., 2003; Zeanah et al., 2005; Minnis et al., 2007; Lyons-Ruth et al., 2009). Therefore, the population referred to as RAD in these studies are likely to be predominately children with DSED, or perhaps mixed DSED/RAD. If the latter, this would be similar to any other co-occurring disorder with DSED.

DSED was first reported in samples of international adoptees post institutionalisation (Tizard and Rees, 1975; O'Connor et al., 2000; Smyke et al., 2009) but has also been found in community samples of maltreated children (Kay and Green, 2013; Minnis et al., 2013; Kay and Green, 2016). Concern has grown among clinicians working in Paediatrics and Child and Adolescent Mental Health Services (CAMHS) because some characteristics of DSED/$DSED^{RAD}$ appear to superficially overlap with Autism (Sadiq et al., 2012; Davidson et al., 2015; Davidson, Moran and Minnis, 2023), yet Autism is not caused by maltreatment (Turner et al., 2019). For example, in $DSED^{RAD}$, lack of social reciprocity, empathy, poor awareness of social cues and pragmatic language difficulties have been reported (Rutter et al., 1999; Pears et al., 2009; Mukaddes et al., 2000), and Guyon-Harris et al., (2019) reported poorer social competencies in children with DSED compared to peers. All these difficulties form parts of the core diagnostic criteria for Autism (DSM-5). In addition, Davidson et al., (2015) found that when children with Autism and no maltreatment history were assessed via standardised caregiver assessment of DSED and RAD, 38% (n=58) met diagnostic criteria for DSED. Inappropriate diagnosis of DSED instead of Autism could have significant child protection implications for families.

Nevertheless, a small number of studies are beginning to demonstrate that it is possible to discriminate between behavioural symptoms of Autism and DSED. For example,

Davidson et al, (2023) found that children with DSED demonstrated better reciprocal interaction, imagination skills, use of nonverbal gestures and understanding of complex humour compared to children with Autism and no maltreatment history. Sadiq et al, (2012) and Mayes et al, (2017), found that repetitive and stereotyped behaviours may be more prevalent in Autism compared to DSED^{RAD} and Moran (2010) developed the Coventry Grid which suggested that, in clinic, children with Autism tend to have a more 'matter of fact' feel to the therapeutic rapport, compared to children with DSED^{RAD} and play tended to be less imaginative.

However, except Davidson et al (2023), none of the above studies directly reported any differences related to social communication and no study investigated the receptive and expressive language skills of either group. Furthermore, DSED tends not to be identified within the communication literature regarding maltreated children, and this gap in knowledge further exacerbates the clinical conundrum of Autism or DSED, as clinician have less information about what to expect regarding DSED and communication.

1.1. *Overview of the literature regarding the communication skills of children with Autism and children with DSED.*

Language skills can vary greatly in children with Autism and is relative to cognitive function. Tager-Flusberg et al, (2005) reported that approximately 80% of children with Autism may have cognitive impairment, children with Autism may develop speech at a slower rate than neurotypical peers, some show language regression and around 30% of children with Autism are estimated to be minimally verbal (Bal et al., 2019). However, some studies suggest that children with Autism without significantly impaired verbal IQ can develop phonology, syntax and morphology comparable to peers of a similar *developmental* age, (Tager-Flusberg et al., 2005; Tek et al., 2014) and vocabulary skills tend to be relatively intact (Rhea, 2007). A recent systematic review demonstrated that the receptive and expressive language of children with Autism under 11 years were poorer compared to age matched neurotypical peers, but that progression followed the 'typical' developmental trajectory (Brignell et al., 2018). However, the hallmark of Autism is impaired social communication (DSM-5), and pragmatic language is believed to be universally impaired (Rhea, 2007). Pragmatics, along with semantics, are usually disproportionally impaired relative to other language skills in Autism (Tager-Flusberg et al., 1995) but structural language difficulties can still present (Eigsti et al., 2007).

In comparison, we found no studies which investigated the receptive or expressive language abilities of children with DSED. Yet, the wider literature appears to suggest that children with a maltreatment history may be at higher risk of language problems, especially expressive language (Carr et al., 2020). One criticism of the literature is that standardised assessments, often relied upon in research, may not be of best value in this population as lower socio-economic deprivation is both associated with poor performance on standardised assessment and with maltreatment (McCool, 2021). However, Eigsti and Cicchetti (2004) found that maltreated pre-schoolers presented with lower language scores compared to non-maltreated peers of a similar socio-economic status. Syntactic development was also reported to be poorer in maltreated children compared to children from lower socio-economic backgrounds, although both groups performed below norms (Beeghly and Cicchetti, 1994). Another area consistently reported to be impaired in maltreated children is narrative production, (Ciolino et al., 2021; Snow et al., 2020), which is related to expressive and pragmatic language skills. Furthermore, one systematic review suggested that the pragmatic skills of maltreated children may be impacted because of difficulties with perspective taking, executive functioning and working memory, with narrative discourse being particularly affected (Hyter, 2021). While some bias may be present as maltreated children are at higher risk of having one or more neurodevelopmental conditions (Dinkler et al., 2017), it is of interest in relation to DSED as difficulties with executive functioning, such as inhibitory control, have been reported (Bruce et al., 2009; Pears et al., 2010). McCool and Stevens (2011) also found in a sample of 30 foster care youths, that 63% presented with differences in social communication via the well validated Child Communication Checklist -2 (CCC). Notably, of those individuals, 42% had a profile suggestive of Autism and the remaining other speech, language and communication difficulties. Although it is not known if any of these young people were Autistic or not, the findings certainly support the concerns around symptom overlap.

Only one study has explored pragmatics specifically with children with DSED. Sadiq et al, (2012) found that both the $DSED^{RAD}$ (n=35), and Autism group (average verbal IQ) (n=52) significantly differed from neurotypical children (n=39) in the domains of inappropriate initiation, coherence, stereotyped conversation and social interests, on the CCC but *only* the $DSED^{RAD}$ group significantly differed regarding rapport. Furthermore, the $DSED^{RAD}$ group showed *greater* impairment than children with Autism regarding use

of language in context, rapport and social relationships. However, 40% of the DSED group also met Autism diagnostic criteria on caregiver report, therefore it remains unknown whether the social communication problems were due to co-existing Autism traits.

To address the gap in literature, the current study aimed to investigate the communication skills of children with Autism compared to children with DSED and children without Autism, DSED or any additional diagnosis (NAD), to determine if any group differences could be established. As there was no previous literature regarding the language skills of children with DSED, we employed an iterative design and began phase 1 with the fundamentals of receptive vocabulary. Drawing on the learnings of phase 1, in phase 2 we investigated receptive skills in more depth; in addition to assessing expressive and pragmatic language skills. We utilised multi-informant standardised measures and analysed conversational speech. This was in part because concerns about the validity of standardised measures for maltreated samples have been expressed in the literature, and partly to explore whether conversational speech yielded similar or different outcomes to standardised assessment/caregiver report.

2. **Methods**

2.1 Design

Receptive vocabulary is a foundational building block to understanding and using language functionally to meet a need and socially to interact with others. When considering where to start in the exploration of the language skills of children with DSED compared to children with Autism, it seemed appropriate to begin with early developmental outcomes such as receptive vocabulary because even this base level of communication has not yet been explored in children with DSED.

We used a cross-sectional mixed methods design involving two separate study groups. For the receptive vocabulary investigation, we utilised an existing dataset to explore outcomes in each of the participant groups (DSED, children with Autism and no maltreatment history and typically developing (TD) children) with a larger sample. Then we used the findings to conduct a more focused investigation of receptive language, expressive language and pragmatic language with a necessarily smaller case study sample, using descriptive methods and Speech and Language Therapist (SLT) clinical analysis.

2.2. Participants

Receptive Vocabulary investigation (existing dataset from previous studies (see Davidson et al., 2015*)*

Participants were 43 children with Autism and no maltreatment history (35 male, 8 female, mean age, 8.07), 24 children with DSED (16 male, 8 female, mean age 6.42) and 37 typically developing children (TD) (24 male, 13 female, mean age, 6.46). In brief, the Autism group had been recruited via clinical services following diagnosis. The recruiting clinician cross checked electronic records with the specialist child protection database to ensure lack of maltreatment, (see Davidson et al, 2015). The DSED group were recruited either from clinical services or adoption charities and DSED symptoms were confirmed via standardised multi-informant assessment for DSED. The TD group were recruited via leaflets distributed by their primary school (See Davidson et al, 2015). All participants attended mainstream primary school.

Exclusions: Cognitive functioning is known to impact language development, therefore we set out to control for IQ using the existent data on global cognitive functioning (Wechsler Abbreviated Scales for Intelligence, Wechsler, 1999) However, we found that the full-scale IQ (FIQ) of 11 Autism cases were deemed unreliable because the discrepancy between the verbal IQ scale (VIQ) and the performance IQ (PIQ) scale was too large (PIQ>12 points above VIQ). In addition, a third of the DSED cases also turned out not to have WASI data available. These outcomes meant that we were unable to statistically control for cognitive functioning. All the children with DSED, with available WASI data, had a verbal IQ within average range, and all attended mainstream primary therefore we chose not to exclude any of the DSED group. However, it is understood that a proportion of children with Autism, even in mainstream primary, are expected to present with cognitive difficulties (Tager-Flusberg et al, 2005) and our data reflected this. We excluded twenty-one children with Autism on the grounds that their verbal IQ fell within the borderline -intellectual disabilities (ID) range, leaving a total sample of 83 children: 22 with Autism, 24 with DSED and 37 typically developing children.

In depth case study investigation of receptive, expressive and pragmatic language

A sample of thirty-one children were recruited: 10 children with Autism, no maltreatment history, 11 children with symptoms of DSED and 10 typically developing children (TD). DSED symptoms were confirmed prior to acceptance into the study using the current best

practice multi-informant standardised assessment for DSED (Minnis et al, 2007; Lehmann et al 2020). This involved caregiver interview, observation and teacher questionnaire. For further details on the diagnostic process, see Davidson et al, (2023). *All* the children were rigorously assessed using multi-informant standardised Autism assessment tools to confirm Autism (Autism & no maltreatment group), to assess and exclude cases of co-occurring Autism (DSED group) (see Davidson et al., 2023 for further info) and exclude any unidentified cases of Autism (TD) group. The multi-informant package used was the Diagnostic Interview for Social and Communication and Other Disorders (DISCO) (Caregiver report) (Wing et al., 2002), the Autism Diagnostic Observation Schedule-2 (ADOS-2) (Lord et al., 2000) and the Live assessment. The Live is an unstructured and dynamic behavioural observation, where skilled assessors manipulate aspects of the social interaction to provide opportunities, prompts and probes (see Davidson et al., 2023).

Exclusions: From the 31 children, we excluded cases of suspected intellectual disability based on communication and adaptive functioning skills, which amounted to one child in the Autism group and one child in the DSED group. A further two cases were excluded from the DSED group due to co-existing Autism, leaving a total sample of 27 children: 9 children with Autism, 8 children with DSED and 8 TD children.

Socio-economic deprivation: We used the Scottish Index of Multiple Deprivation (SIMD) to group match, as closely as possible, by socio-economic status and by age. SIMD data represents areas of deprivation based on postcode; the areas are split into deciles, and the higher the decile the greater the proportion of areas within which are considered as deprived.

Environmental circumstances: We aimed to collect data in the DSED group regarding the length of time the child spent in the maltreatment environment prior to removal into care, as there is some evidence to suggest that older age/length of exposure may negatively impact communication (Maguire et al., 2021). However, it was extremely difficult to obtain accurate information as the foster/adoptive families (50% of the children were adopted and 50% were fostered/in kinship care) often did not have a complete picture and, due to unexpected circumstances, we were unable to obtain objective records via social work services. Most of the sample had lived within the maltreatment environment for at least 3 months and several for 6 months to 1 year prior to being fostered. One child (child 2) was internationally adopted; while she had only ever learned English monolingually,

we recognise that she had to learn English morphology and syntax more quickly at an older age without early receptive input, and this is considered relative to her outcomes.

Co-occurring neurodevelopmental conditions: Two children in the Autism group had co-existing ADHD, which was treated with medication, and two children in the DSED group had a recent diagnosis of co-existing ADHD and were not yet receiving treatment, of any kind.

2.3. Measures

Receptive vocabulary investigation

British Picture Vocabulary Scale II (BPVS) (Atkinson, 1992): The BPVS is standardised measure of receptive (hearing) vocabulary for standard English and can be used to demonstrate the extent of English vocabulary acquisition. It is picture based and does not rely on expressive language.

Wechsler Abbreviated Scale of Intelligence (WASI) (Wechsler, 1999): is a standardised short form measure of cognitive ability. It consists of 4 subtests; 2 of which are a measure of verbal comprehension (Verbal IQ) and 2 of which measure non-verbal reasoning (Performance IQ). The two scales combined provide a reliable index of cognitive function. However, a discrepancy of 12 or greater between VIQ and PIQ will render the full-scale IQ unreliable.

In depth case study investigation of receptive, expressive and pragmatic language

To reduce assessment burden, we selected two sub-tests of the Clinical Evaluation of Language Fundamentals-5 (CELF-5) (Wiig et al., 2013) which have been reliably used together in research because they have good reliability and validity as indicators of receptive and expressive language (Botting and Conti-Ramsden, 2008). These were as follows:

Receptive Language: *Word Classes*, which assesses the child's ability to understand relationships between words based on meaningful features, function, place or time of occurrence. The receptive element requires the child to listen to 4 items presented verbally and then choose which 2 items 'go together.' Word classes is more demanding than the BPVS, as it requires attention and listening and semantic skills.

Expressive Language: *Recalling Sentences* is used to assess ability to recall and reproduce sentences of varying length and syntactic complexity. The child is required to listen to the sentence and immediately repeat it. This test relies on both listening and attention skills and working memory, in addition to an understanding of language structures, to reproduce the sentences accurately.

Child Communication Checklist-2 (CCC-2) (Bishop et al., 2003) is a standardised caregiver report which screens for communication problems, screens for general language abilities, and helps identify possible pragmatic language impairments in children ages 4-12 years.

Assessment of conversational speech: CD-J, an SLT experienced in working with this client group, reviewed the video recordings of an unstructured observational assessment called Live that was conducted as part of the larger study, mentioned above. The Live is completed by 2 assessors (one of whom was CD-J) and involves both informal conversation and play (see Davidson et al., 2023 for further details on LIVE). Approximately 10 minutes of conversation was selected, per participant, and the audio was isolated from the video and transcribed verbatim. Two independent SLTs, blinded to the diagnoses, were asked to complete a structural language analysis based on guidelines by Bowen (2011) which uses Brown's Stages of Syntactical and Morphological Development (Brown, 1973). SLTs identified presence or absence of structures expected to have emerged by age 5 years and the frequency of their use. More complex syntax such as conjunctions (and, because, but, if etc.), connectors (also, however etc,), inverted forms of questions etc. were also recorded, and errors noted. Each SLT analysed 4 Autism cases and 4 DSED cases each (blinded to diagnoses) and CD-J completed the same analysis of all the cases. Almost 100% inter-rater agreement was reached between CD-J and each independent SLT, but helpful discussions occurred regarding where to record items like, imperatives, problems with timing, false starts and interrupting, or around more complex error patterns. Finally, CD-J analysed all the speech samples with a narrower focus on pragmatic language skills, drawing on Peccei, (1999), and the independent SLTs noted any observations regarding pragmatics during their above analysis.

2.4. Procedure

As an existing dataset was utilised for the receptive vocabulary investigation there was no participant recruitment. The following refers to the procedures undertaken in recruiting

and assessing the case study group for the receptive, expressive and pragmatic language investigations.

The children with Autism had a pre-existing diagnosis and almost all cases were referred by parents following advertisement on national Autism Charity websites. One child with Autism was referred from CAMHS. Six of the children with DSED were referred by their foster/adoptive parents in response to a web advert placed by fostering/adoption charities and 2 children were referred by CAMHS. All the TD group were referred by their parents in response to a recruitment leaflet. The DISCO was completed at a home visit by CD-J and caregivers completed the CCC-2 in the waiting room, while their child participated in the Live assessment and completed the sub-tests of the CELF-5. CD-J sent the audio samples to the independent SLTs for analysis once all the assessments were completed.

3. Results

3.1 Receptive vocabulary investigation

Table 1 demonstrates the descriptive statistics based on BPVS standard scores of the children with Autism (average VIQ), DSED and the TD children.

Table 1: descriptives for BPVS standard Score

	Total number	*Mean standardized Score*	*Std. Deviation*	*Min-max range*
Autism	22	94.77	14.77	69-127
DSED	24	97.79	9.05	82-118
TD	37	100.57	9.99	82-135

We conducted a one-way ANOVA to explore any differences in mean BPVS score, and using the Levene statistic, the requirement for homogeneity of variances was met. There were no significant differences between groups, ($F(2) = 1.87$, $p = 0.16$). However, the descriptive statistics suggest some difference within the distribution of BPVS standard scores. There was a much wider range within the Autism group compared to the DSED

and TD groups with noticeably more children performing within moderately low to poor range. Yet, some children with Autism also scored highly, which may have compensated for the low scorers and led to the non-significant result.

3.2. In depth case study investigation of receptive, expressive and pragmatic language

3.2.1. Demographic data: While the TD group were slightly older, the Autism and DSED groups were fairly reasonably matched by age and SIMD data, with the DSED group mean suggesting just slightly higher socio-economic deprivation than the Autism group, despite being in foster care.

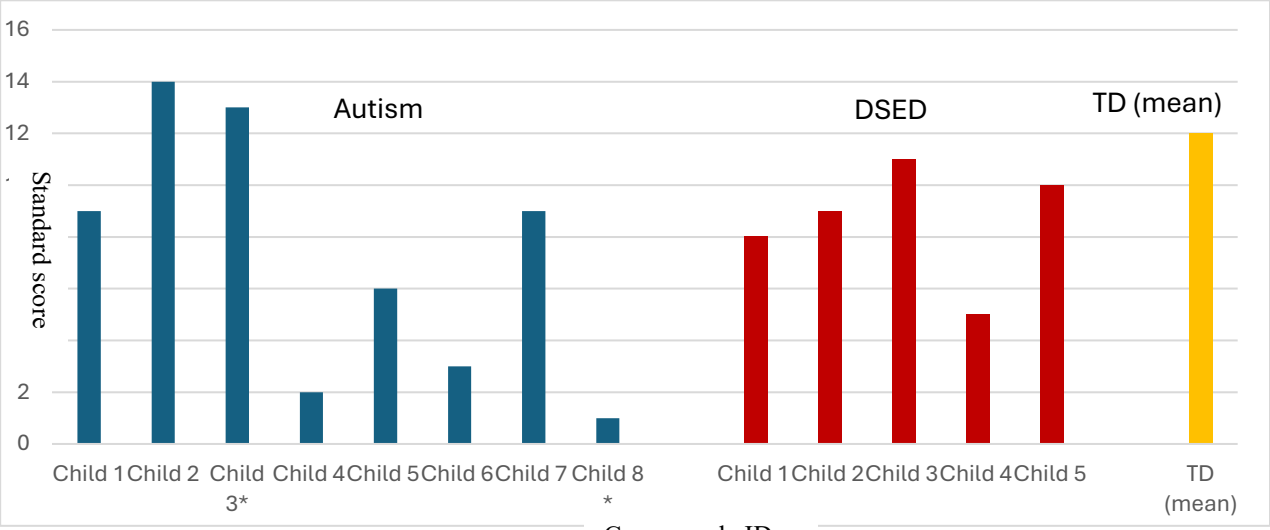
Table 2: demographics of the case study group

Group	*Gender (male: female)*	*Mean age (range)*	*Mean SIMD decile (range)*
Autism	6M: 3 F	7.67 years (5-11)	5.1 (1-10)
DSED	6M: 2 F	7 years (5-10)	7 (1-10)
TD	8M: 2 F	9.67 years (8-12)	6 (2-9)

3.2.2. Receptive language (CELF data)

There is missing data in the Autism group (n=1) and TD group (n=3) due to parental time constraints. In the DSED group, the two children with DSED and ADHD refused to complete either sub-test, both disengaging and presenting as oppositional as soon as the listening demands became apparent. Chart 1 demonstrates the standard scores from the Word Classes receptive language task. In each of the results sections, the asterisks indicate the children with co-occurring ADHD.

Chart 1: Word Classes (WC)



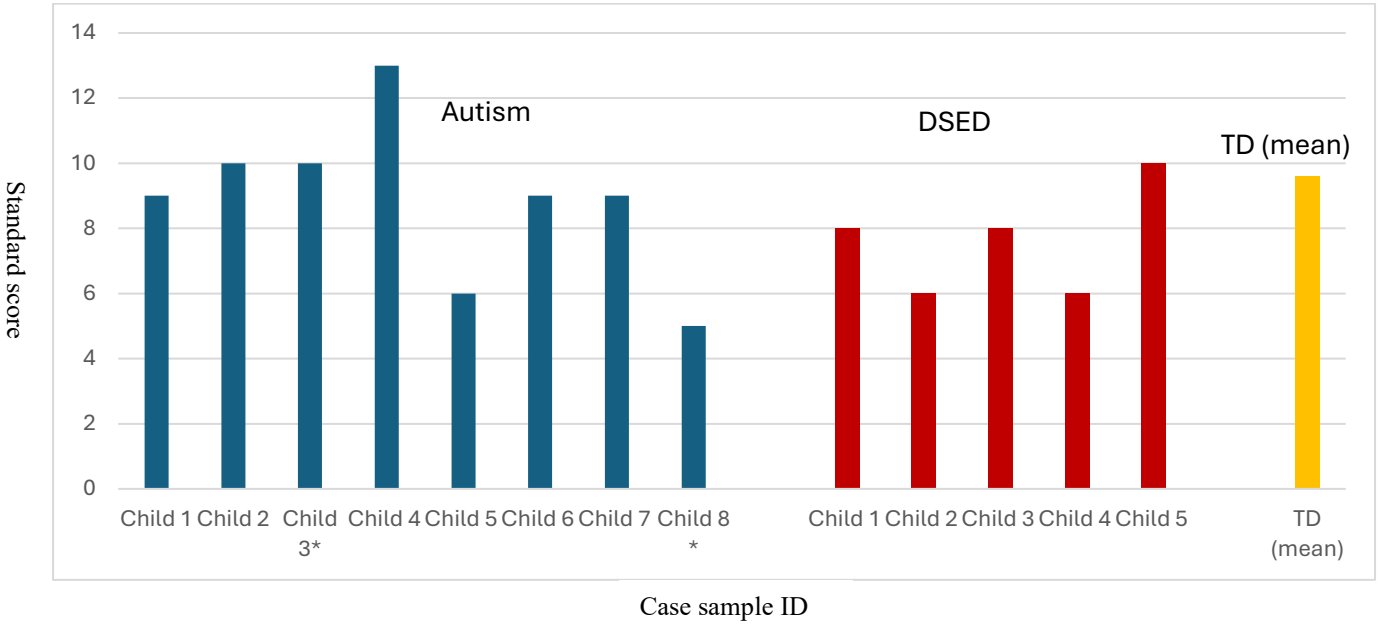
TD Group: All the children demonstrated receptive language skills on the Word Classes task within the average range, and therefore we present the group mean as a point of reference, (mean = 12, range = 9-17).

Autism Group: Half the group scored within average range, or above, on the receptive language sub-task and not dissimilar to the TD group mean. However, the other half, and particularly Child 4, 6 and 8 scored poorly, which again demonstrates the individual variation within Autism, even when verbal IQ is accounted for. For the children who scored poorly, it was apparent that they understood the individual vocabulary but struggled with the more abstract concept of identifying the shared meaning within semantic categories.

DSED: Only one child (child 4) did not score within average range on the receptive language task; this child also understood the vocabulary but was unable to identity shared meaning.

3.2.3. Expressive language (CELF data)

Chart 2: Expressive Language (Recalling Sentences)



TD Group: almost all the children performed within average range, thus we present the group mean for comparison, (mean = 9.6, range = 6-15). There is quite a broad distribution of scores for this task, which demonstrates that even in typical development, language and communication abilities can vary, and one child has performed poorly.

Autism group: Most children with Autism performed within average range on the recalling sentences task, suggesting they had a reasonable grasp of how to use age-appropriate morphology and syntax. Only 2 children scored poorly (child 5 and 8).

DSED group: The scores of the children with DSED are generally lower than most of the children with Autism, except for child 5. The standard scores of child 1 and 3 could be considered 'low average.' Like the Autism group, 2 children performed poorly (child 2 and 4).

Of interest, one of the two children with Autism who performed poorly on the expressive task was a child with Autism+ADHD. This task requires listening and attention skills but also relies on working memory, an executive functioning skill that children with ADHD tend to struggle with. We are unable to compare findings of the children with DSED+ADHD because they disengaged during the task but hypothesise that this is a task which children with ADHD will find much more difficult.

3.2.*4. SLT structural analysis: Autism and DSED groups*

Of the 9 children with Autism, we were unable to complete analysis of one case due to technical problems recording the video of the Live assessment.

Children in both the Autism and DSED group demonstrated use of most of the morphology and syntax expected by age 5 years, as well as examples of more complex speech. Yet, most of the children with Autism used little or zero -wh questions, despite opportunity to do so. This lack of use appeared to be related to pragmatic skills, therefore we discuss this further in section 5. The children with DSED used -wh questions within conversation more often and appropriately.

Table 1 below demonstrates the error patterns identified per child, per group.

Table 1: Error Patterns

Child group+ participant ID	*Age*	*Any additional relevant information*	*Frequency of errors* *few = 0-3* *moderate = 4-7* *frequent = 8-11* *very frequent = 12+* *(no. of errors)*	*SLT input (*= caregiver description)*
Autism Group				
Child 1	8		Moderate (7)	No
Child 2	11		Few (2)	Yes, (in past as part of Autism diagnosis)
Child 3*	5	+ ADHD	Moderate (6)	No
Child 4	7		Moderate (5)	No
Child 5	6		Moderate (5)	No
Child 6	5		Moderate (6)	Yes (in past re: pragmatic language)
Child 7	10		Moderate (5)	Yes (in past re: pragmatic language)
Child 8 *	10	+ ADHD	Moderate (5)	Yes (in past for a stutter)
DSED Group				
Child 1	10		Moderate (6)	No
Child 2	6	International adoptee. Only ever spoken English.	**Frequent (10)**	Yes (recently discharged re: language delay)
Child 3	5		Moderate (4)	No
Child 4	8		Moderate (5)	No
Child 5	6		Moderate (6)	No
Child 6	6		**Frequent (9)**	No
Child 7*	7	+ ADHD	**Frequent (9)**	Yes (in early years re: language delay)
Child 8*	8	+ ADHD	**Very frequent (17)**	No

Autism Group: A similar number of errors were found across the group and errors were distributed across clause, phrase and morphological levels. Morphological errors tended to be -ed overgeneralisations and/or use of wrong pronoun and at phrase and clause

level, errors tended to be omissions of structures (pronouns) or tense agreement errors. The error patterns perhaps reflect some immature language development, more so in the older children (child 1, 7 & 8), and some like -ed overgeneralisation are patterns found in typically developing children at the early stages of language acquisition.

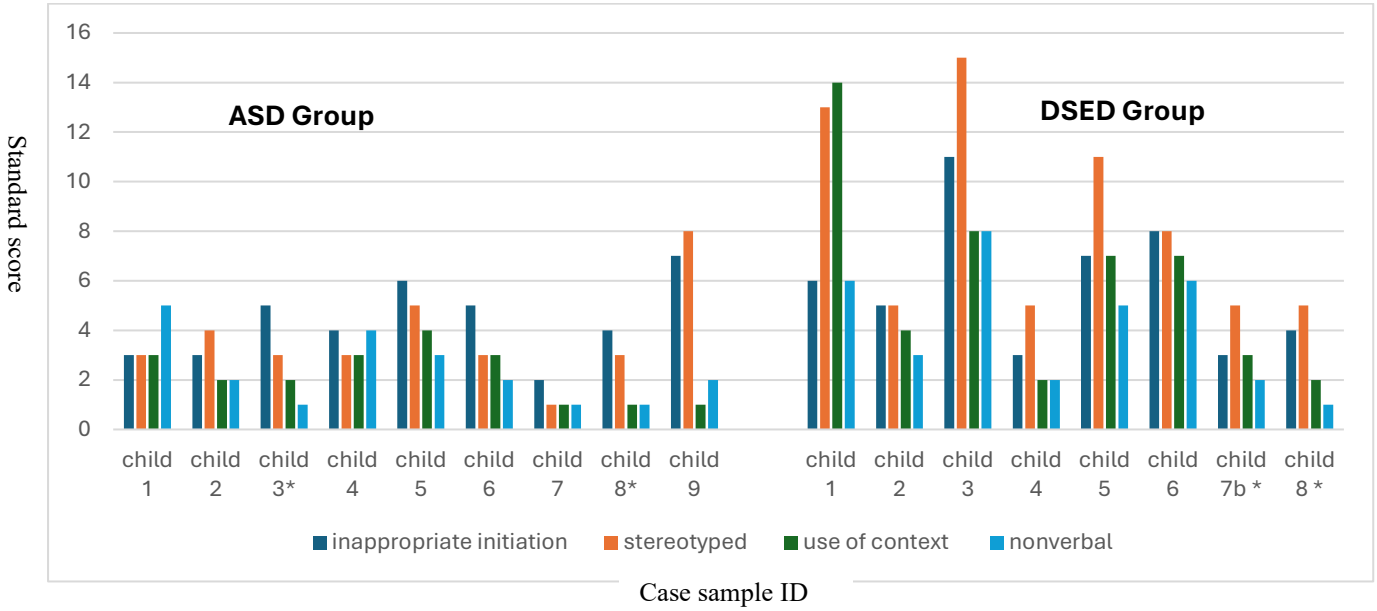
DSED Group: 50% of the children demonstrated a similar number of errors to the children with Autism (<7) and the other 50% a greater number of errors (>7). Most of the errors appeared at clause level, but morphological errors were still apparent and within the error pattern there was greater individual variation. At clause level, most children demonstrated omission errors (one or more of, pronouns, prepositions, articles, auxiliary verb etc) and verb agreement errors but others, such as child 2, 5,7 and 8 also demonstrated word order errors and child 8 also had unusual verb selection errors. For example, *"A hot dog gun, means like this [imitating shooting]."* Through discussion, we decided that 'means like this' was possibly replacing the clause 'it does this.' There was also a wider range of morphological errors across the group compared to the children with Autism: wrong pronoun, omission of s' plural or s' possessive markers, preposition errors, wrong tense marked, as well as -ed overgeneralisation. Child 8 also used s-plural instead of irregular past tense on 2 occasions. These findings seem reflective of the lower expressive language scores on the CELF subtest and in some cases a lower developmental level of language than typically expected for their age.

Of the 4 children with DSED with the greatest difficulties, 2 of these cases are children with DSED+ADHD, and 1 child (child 2) is the individual who was internationally adopted. The relative impact of co-existing conditions and environmental factors, in addition to maltreatment appears an area for future research.

3.2.5. Pragmatic Language (CCC-2 and clinical analysis)

We-used the CCC-2 to broadly explore pragmatic language skills. All the children, except for one, within the TD group scored within the average range for each of the pragmatic language domains. The mean TD scores per domains were as follows: inappropriate initiation (mean, 10.1, range, 5-15), stereotyped language (mean, 9.7, range, 5-13), use of context, (mean, 10.1, range, 6-14) and non-verbal, (mean, 11, range, 6-12). The outcomes of the Autism and DSED groups are presented below in chart 3.

Chart 3: CCC-2 pragmatic language domains, per group (Autism and DSED)



Autism: Consistent with our understanding of pragmatic language being almost universally impaired in children with Autism, all the children scored poorly, in almost every domain.

DSED: 50% of the children scored similarly poorly to children with Autism across all domains of the pragmatic language scale (child 2, 4, 7 & 8) and of the remaining 50%, all bar child 3 scored poorly on at least 2 of the 4 domains. Nonverbal communication was the most reported area of difficulty (87.5%) followed by inappropriate initiation and use of context (75% respectively). Stereotyped language was the least reported but was still present in 50% of cases.

Next, we explored pragmatic skills in relation to reported language difficulties. The CCC-2 provides a General Communication Composite (GCC) which is an indicator of overall communication competence (language skills + pragmatic language) and a Social Interaction Deviance Composite (SIDC) which is calculated by subtracting the language scores (speech sounds, syntax, semantics and coherence) from the pragmatic language scores (scales as shown above). Where children score below the clinical cut off of 55 on the GCC and show a negative score on the SIDC, this is considered indicative of Autism and a negative SIDC score with normal language, indicative of Aspergers (designed pre-DSM-5). In contrast, a low GCC score and a positive SIDC score is indicative of language and communication problems rather than Autism. The

threshold for Developmental Language Disorder (DLD) is a GCC below 55 and a SIDC of 7 or more.

Autism group: Seven children (78%) presented with the CCC-2 profile indicative of Autism. Child 2 and 7 are exceptions: both were classified according to CCC-2 as "Language and Communication Other," as their language skills were reported to be poorer than their social communication skills. For Child 2, this was an unexpected finding as his parent reported 'frequent problems' in response to every language related question yet his CELF scores were within average range and he produced the fewest language errors of the group during conversational speech (n= 2 errors).

DSED Group: 50% of children with DSED, (n=4) scored within the range suggestive of Autism (n=3 Autism and n= 1 Aspergers) which match the reported difficulties on the pragmatic language scale. However, it is also possible that expressive language difficulties are exacerbating the pragmatic problems for some children. Child 2 and child 8 scored particularly poorly on the language domains on the CCC-2 and while child 8's profile was suggestive of language and communication difficulties, child 2 met criteria for DLD. Similarly, child 6 scored within 1 point only of the normative range for language. These findings also mirror the outcomes of the CELF-5 expressive task and the structural analysis.

N.B. The CCC-2 scores and related profiles, per child, are supplied within the supplementary table.

3.2.6. *SLT analysis of pragmatic languages skills during conversation*

We compared the conversational samples of the children with Autism and DSED with a focus on narrative production, inference, the maxims of communication (quantity, quality, relevancy and ambiguity), speech acts and understanding/use of metaphor.

<u>Pragmatic problems which overlapped between groups</u>

Narrative Production (Autism group): For all the children with Autism there were instances where the narratives lacked context or minimal information was provided to cue in the listener, for example, "*There's this thing, it's so funny, because Freddie goes to the shower and then Jason comes to the shower and he's so fat and he's not got his top on and he's just like this (demonstrates the action).*" During the assessment, it took some time to work out that the child was referring to a spoof on YouTube of horror

movie characters, because there was little referent context given even although the subject and their characteristics were described. In some cases, volume of information (too much) information was also flouted, which tended to occur when talking about their specific interests. Some children continued to return to their agenda and use of vague terms or overuse of filler words were apparent.

Narrative production (DSED group): The narratives of most of the children with DSED also lacked context and too little information was provided to cue in the listener. Furthermore, child 5, 7* and 8* often jumped topics, were focused on/returned to their agenda, and use of vague terms added to the confusion. For example, child 7*, in the previous utterance, had spontaneously referred to the brain and labelled different emotions, albeit in a confused manner, but his subsequent response is even harder to follow and suggests language difficulties may be further adding to his pragmatic problems -"*And they're the ones who will control it in your head and then they do that and they shows to where you need to know and let's that, but the thing, the thing that keeps you going, you actually need food*." Child 7* and 8* also tended to speak at a fast pace and run words together, which further added to lack of clarity.

<u>Pragmatic Skills which differentiated DSED from Autism</u>

- Children with DSED were able to infer intent/relevant information regarding the assessors' experiences more often than the children with Autism, or with less cues. In some of the Autism cases, missing the implication then led to reduction in use of -wh questions. For example, when the examiner shared that she hoped her goldfish was still alive when she got home (implication it might be ill, opportunity to ask why, or what was wrong), the child instead responded, *do fish eat peas?*
- The children with DSED used interrogatives (questions), including -wh questions, for a range of purposes. Examples include, enquiring about another's experience, which naturally expanded the conversation, *what games do you play?* Or, including another in their experience, *what one d'you wanna be? (referring to toys)* or showing interest i.e., the examiner shared a story about her co-examiner getting married and the child immediately responded, *when will that be? (referring to the wedding).*
- In the Autism group, -wh questions were also reduced because many conversations tended to take on a question-answer flow, where the examiners were asking to keep the conversation going and the child was only responding. The exceptions to this were

often when something related to the child's particular interest arose. For example, the examiner shared a story about her son's activity and the child responded, *what building is it in?*

- Children with DSED used greater variety of speech acts: at least two of the children used declarative sentences as an offer to share, *I can do the splits* (look, I want to show you what I can do), they were used in protest, and 2 children used commissive statements (showing future intent), *I don't have it with me, but I'll bring it next time.* The children with Autism tended to rely on one form of speech act, mainly declarative sentences (making statements), or in the case of child 5, maintained interaction only by asking questions using inverted forms repetitively.
- Some children with DSED used metaphors, *she's lost her mind,* and most showed understanding of these. In the Autism group, understanding of metaphors was mixed, but metaphors were never used, and there was more of a tendency for literal interpretation.
- Odd phrasing/idiomatic language was only present in 2 children with DSED (both of whom may have language difficulties) but was more common in the Autism group.
- Both groups of children showed lots of false starts and repetitions in sentences, which may reflect speech processing, but true echolalia was only present in the Autism group. Of note, we did not explore the nonverbal elements of pragmatic language in this sample of children as these have already been explored elsewhere, (see Davidson et al, 2023 for further information).

4. Discussion

In this exploratory study, we investigated the receptive, expressive and pragmatic language skills of children with Autism, children with DSED and typically developing children. No significant group differences were found regarding receptive vocabulary when verbal IQ was accounted for. However, in keeping with the literature, the children with Autism had greater problems than the DSED and TD groups when semantic skills were required (Tager-Flusberg et al., 2005). In contrast, the children with DSED scored more poorly on the CELF-5 expressive language sub-test compared to the Autism and TD groups. In conversational speech, the error patterns of the children with Autism tended to be developmental errors associated with early language acquisition and wrong pronoun, the latter of which is commonly associated with

Autism (Ren et al., 2023; Finnegan et al., 2021). Whereas, in the DSED group, a greater number of non-developmental morphological and syntactical errors presented compared to the children with Autism. Regarding pragmatic language skills, both children with Autism and children with DSED presented with narrative discourse difficulties which lacked context and flouted maxims of quality, quantity and relevancy, which for clinicians concerned with differential diagnosis may add uncertainty. However, in this sample, specific differences in pragmatic skills were also identified. The communication literature regarding samples of maltreated children suggests these children are at higher risk of language difficulties, and in particular expressive language and pragmatic language difficulties (Carr et al., 2018; Hyter, 2021, Mcool and Stevens, 2011). Our study supports these findings and is the first to suggest that maltreated children with DSED may also present with the communication difficulties reflected in the wider literature.

This is also the first study to investigate *both* language and social communication profiles of children with DSED in comparison to children with Autism (average verbal IQ) and tentatively suggests some patterns which may be supportive of differential diagnosis. Firstly, within the structural analysis of expressive language, we identified that most of the children with Autism rarely used -wh questions; the children with DSED used -wh questions appropriately. Recent research found reduced use of -wh questions, via naturalistic sampling, in a large group of children with Autism compared to children with language delay and typically developing children (Bacon et al., 2018). Lack of -wh question production/-wh question errors have also been found within cross-cultural samples of children with Autism (Sukenik et al., 2021). Sukenik et al found that some -wh question errors were related to problems with syntax and pragmatics, but the errors related to pragmatics, including perseveration of use of alternative question forms, differentiated the children with Autism from children with DLD. The latter finding resonates with our sample as we tended not to see syntactical errors in -wh questions, but pragmatically lack of use and/or mostly lack of range i.e., 'what' used to ask for clarification, but very little -wh questions to ask about experiences of others, to enquire or expand conversation flow. We also noted perseveration, as described above, in one child with Autism, who's communication was largely made up of inverted forms of questions, but zero -wh questions, and very few other speech acts. Thomas et al, (2021) argue that spontaneous language assessment

demonstrates greater expressive language problems in children with Autism compared to standardised assessment and, like Bacon et al (2018), argue for analysis of the speech of children with Autism in naturalistic settings. Our findings support and add to the argument, as the -wh questions pattern was only found because of our analysis of the speech from the unstructured behavioural observation (Live assessment).

Related to the argument for more naturalistic assessment, is preliminary evidence which suggests that *unstructured* clinical observation might be a more useful tool for supporting differential diagnosis between Autism and DSED. Davidson et al, (2015) found that children with Autism often met core diagnostic criteria for DSED on standardised caregiver tools, but observation during unstructured conversation helped to discriminate Autism from DSED in most cases. Davidson et al, (2023) found that children with DSED met core criteria for Autism on the DISCO (caregiver report) and half the DSED sample met core criteria for Autism on the structured ADOS-2 but dynamic unstructured assessment better helped identify key differences in the social interaction and communication of children with DSED. The current study adds to this body of evidence because like Sadiq et al, (2012), we found that on caregiver report (CCC-2) children with DSED present with pragmatic difficulties to a similar degree as children with Autism, yet SLT analysis of conversation more clearly demonstrated the areas of overlap and important areas of differentiation. Poor narrative production was the main area of overlap between the DSED and Autism groups, which is consistent with findings in the wider maltreatment literature (Snow et al., 2020; Hyter et al, 2021; Ciolino, 2021). However, in this modest sample of children with DSED, the children generally showed more skills in areas of pragmatic language that when impaired are considered synonymous with Autism. These were, drawing inference from conversational contexts, using a range of speech acts, less idiomatic speech, except for the children with language difficulties, and metaphorical speech was better understood and used. In addition, echolalia was present *only* in the Autism group. Nevertheless, the fact that children with DSED showed improved pragmatic skills, in some areas, compared to children with Autism, does not necessarily mean that their social communication is not impaired relative to typical development, and this requires further investigation.

4.1.Clinical considerations and areas of future research

While some overlap in social communication problems between Autism and DSED are undoubtably present, the two studies by Davidson et al and the current study taken altogether perhaps raise an argument for a move towards *unstructured* clinical observation for cases causing diagnostic dilemma and this is certainly worthy of future investigation. It may be that standardised measures like the DISCO and CCC-2 are too broad in scope to pick up key differences, or that SLT expertise of finely tuning into the *quality* of the observed pragmatic/communication difficulty is vital.

Secondly, our investigation raises questions regarding the overlap of neurodevelopmental conditions in children who have experienced maltreatment and the relative impact of these, especially if unrecognised, on language and communication. The two children with DSED+ADHD appeared to have the greatest difficulties of the group in the domains that we were able to assess (expressive and pragmatic language). This is of great clinical relevance as we know that there is overlap between language and communication difficulties and neurodevelopmental conditions in maltreated children (Clegg et al., 2021) and robust empirical data demonstrates that maltreated children are at higher risk of having one or more neurodevelopmental conditions, maltreatment-associated disorders such as DSED or *both* (Dinkler et al., 2017; Minnis 2013). Given that a recent systematic review suggested that executive functioning problems, among others, in maltreated children may impact pragmatic language skills, our findings that the children with DSED+ADHD were doing worst is noteworthy. We know that non maltreated children with ADHD are more likely to have pragmatic language difficulties, (Cordier, 2021; Carruthers, 2021) which can impact social relationships (Bagwell et al., 2001) and delays in speech and language acquisition and difficulties related to working memory and phonological manipulation have also been found (Brites, 2020; Tetnowski,2004). The question which we ponder, but cannot yet answer, is whether children with DSED *and* ADHD (or other neurodevelopmental conditions) are at even greater risk of language and social communication problems than environmental controls without additional neurodevelopmental complexity? Gajwani et al, (2023) propose that the interplay of maltreatment & neurodevelopmental conditions may be 'double jeopardy' for individuals, placing them at higher risk of later severe mental health problems if needs are unmet. Drawing on this line of thinking, we emphasise the need for future research to firstly identify DSED within maltreated samples when exploring language and communication, secondly for

clinicians to expect neurodevelopmental overlap in clinic when working with maltreated children and identify it accordingly - not all communication difficulties are the result of maltreatment. Finally, future research must begin to address the interplay between DSED and ADHD, and in relation to communication difficulties as there is a body of evidence starting to suggest that ADHD, or ADHD symptoms such as poor inhibitory control, are often associated with DSED (Bruce et al., 2009; Pears et al., 2010; Kennedy et al., 2017; Seim et al., 2022). All these considerations may help to better identify the individuals at *most* need of support earlier and add further to our knowledge of differential diagnosis between Autism and DSED, and other additional neurodevelopmental complexities where they arise.

4.2.Strengths and Limitations

Our study design was a relative strength, as we addressed concerns regarding appropriateness of standardised assessments for maltreated children (McCool, 2021) through additional independent blinded SLT structural analysis. Our iterative design also enabled in depth case investigation which highlighted possible areas of future investigation for differentiation between Autism and DSED, shed light on use of caregiver screening tools for pragmatic language versus SLT clinical observation and highlighted the benefit of the latter. Nevertheless, there were also limitations. We were unable to stratify the DSED sample by length of time prior to removal due to lack of robust information; while this is reflective of some of the complexities of working with a 'real-life' clinical sample, it remains worthy of future consideration, as it is possible that length of time in the maltreatment environment may have been a confounding variable in some cases. The inclusion of the children with co-occurring ADHD could be argued as a confounding variable but given this was an exploratory study which aimed to identify factors of clinical relevance rather than determine causality, we feel that the inclusion is justified. However, the related limitation is perhaps that the two children in the DSED group had only been recently diagnosed with ADHD and, unlike the two children with ADHD in the Autism group, were not receiving ADHD treatment (in any form). A possible effect of this was greater disengagement with tasks that demanded listening and attention. Without assessment of the children pre and post treatment however, it is not possible to know relative impact on pragmatic language with regards to impulse control and coherence. A second limitation is the small case study sample. This was a balance between conducting thorough multi-modal

investigation and time constraints of SLT expertise and we acknowledge that the findings require replication with additional samples before generalisations can be made. Finally, some selection bias may be present as most of the children were caregiver referred. However, findings suggest that in some DSED cases, the caregivers were right to be concerned, especially given the limited SLT input in the group.

4.3.*Conclusions*

In our sample of children with DSED we found receptive language skills were similar to typically developing children but difficulties with expressive and pragmatic language skills were present. All professionals working with maltreated children should be aware of the potential of DSED *and* the high risk of language and communication problems, and involve SLTs jointly in assessments, when required. Related to this, SLTs may need to advocate for their role in these complex cases as communication problems may be thought of as secondary by caregivers or professionals when presented with difficult behaviours and/or mental health problems (Cummings, 2021). Regarding differential diagnosis of Autism from DSED, we found that receptive *language* skills may be more impaired in Autism than expressive skills and, in this sample, specific *differences* in pragmatic language associated with Autism were less apparent in DSED when assessed by experienced SLTs. Again, there is a key role for SLTs, as analysis of conversational speech may best support identification of these pragmatic differences.

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(supplementary table)

Table 3: CCC-2 General language composite, social interaction deviance composite and suggested profile

Child group+ID	GLC score	SIDC Score	Suggested Profile
ASD Group			
Child 1	44	-9	Autism
Child 2	20	2	Language and Comm (other)
Child 3*	44	-23	Autism
Child 4	48	-21	Autism
Child 5	36	-4	Autism
Child 6	49	-24	Autism
Child 7	2	4	Language and Comm (other)

Child 8	23	-6	Autism
Child 9	56	-25	Aspergers
DSED Group			
Child 1	70	-8	Aspergers
Child 2	23	7	DLD
Child 3	80	1	Within norms
Child 4	27	-7	Autism
Child 5	54	-1	Autism
Child 6	56	5	Within norms
Child 7	32	-11	Autism
Child 8	20	0	Language and Comm (other)

Reflections on Paper 3

Paper 3 highlighted the need to examine the language and communication skills of children with DSED and in-depth SLT analysis of the case study sample suggested specific areas worthy of future investigation regarding differentiation from Autism. The following points are of salience:

- This paper adds to the knowledge base by investigating the language skills of children with DSED and findings are generally consistent with those of samples of maltreated children within the wider communication literature.

- Differences between Autism and DSED, in a larger sample, were not found at the level of receptive vocabulary when verbal IQ was accounted for. However, in the case study sample, some children with Autism scored poorly on the standardised child measure when semantic reasoning skills were required.

- In contrast most children with DSED, in the case study sample, scored more poorly than children with Autism on the standardised child measure of expressive language. In free flowing conversation, half the DSED group made the same number of errors as the children with Autism and half more errors than children with Autism. A further key difference was, children with DSED demonstrated a wider variation of morphological and syntactic errors than the Autistic children.

- We also found that on the CCC-2 caregiver report, children with Autism and children with DSED appeared to present with a similar range of pragmatic language difficulties, although the severity was perhaps greater in Autism.

- Significantly, we found that analysis of pragmatic skills during free-flowing conversation, by Autism experienced SLTs, demonstrated that the overlap in difficulties related to narrative production. Yet, specific areas of pragmatic impairment which are typically associated with Autism were more apparent in the Autism group and seemed to differ from DSED. As opposed to the children with Autism, children with DSED demonstrated appropriate use of -wh questions, including to ask about others experience, they demonstrated use of a range of speech acts and less literal thinking and idiomatic speech was less apparent.

- The findings of this paper confer with the findings of paper 2 and together suggest that specific differences in the social communication domain are worthy of future investigation with larger samples, as these might be areas which best discriminate DSED from Autism.

- The pragmatic differences between Autism and DSED were better identified via unstructured and free flowing communication, and when assessed by SLTs rather than caregiver report.

- Like paper 2, our findings also raised the question about the relative impact of co-occurring neurodevelopmental disorders, as the children with DSED and ADHD appeared to have the greatest difficulties across all language domains.

Chapter 6: An exploratory comparison of sensory problems in children with Autism and children with Disinhibited Social Engagement Disorder

Papers 1, 2 and 3 have explored in detail the symptoms and skills of children with DSED regarding part A of the Autism diagnostic criteria: deficits in social communication and interaction. In the final paper, attention is turned to part B of the diagnostic criteria: repetitive and restricted behaviours. The focus is on sensory processing as this is the one related area within part B which has not been explored within the literature in relation to DSED.

Paper 4 (Submitted to International Journal of Developmental Disorders)

Davidson, C., Gillberg, C., Lowit, A & Minnis.H. (2023). An exploratory comparison of sensory problems in children with Autism and children with Disinhibited Social Engagement Disorder

Abstract

Background: Clinicians are concerned about the overlap in symptoms between children with Autism and children with the maltreatment-associated disorder, Disinhibited Social Engagement Disorder (DSED). But the lack of research regarding sensory processing in children with DSED, compared to Autism, limits understanding for assessment and differential diagnosis. **Method:** The sensory processing of nineteen children: 10 with ASD and no maltreatment history and 9 with DSED were explored using standardised measures and against the Autism DSM-5 diagnostic criteria. **Results:** In this exploratory case study, most children with DSED presented with sensory problems, greater than typically expected, and similarly to children with Autism, although the degree and severity was generally greater in Autism. Of note, hyper-responsiveness and sensory seeking behaviour in children with DSED presented to a similar degree as children with Autism. Nevertheless, particular sensory behaviours appeared more consistently in the Autism group than the DSED group, and if replicated, with larger samples may help clinicians differentiate Autism from DSED. **Conclusion:** Sensory processing difficulties should be considered in children with DSED, as well as children with Autism, and clinicians involved in assessment of children with DSED should be aware that some sensory behaviours, and particularly sensory seeking and hyper-responsivity, can overlap between Autism and DSED.

Keywords: Disinhibited Social Engagement Disorder, sensory processing, sensory seeking, hyper-responsivity, Autism Spectrum Disorder, differential diagnosis

1. Introduction

Sensory processing involves the nervous system and brain rapidly interpreting and making coherent sense of information from the sensory organs to make appropriate motor or behavioural responses. Information is obtained from the environment via visual and auditory senses, taste, touch and smell, regarding physical movement, internal body states such as hunger, pain and temperature changes and the sense of the physical body in space. Everyone has varying sensory preferences, but when sensory signals are either not detected, not organized into appropriate responses or an individual has so many sensory preferences that it negatively impacts their behaviour, then impaired sensory processing may be indicated (Wilmot, 2020). In the general population, difficulties with

sensory processing are estimated to present in about 5%-16% of individuals, (Ahn et al., 2004; Ben-Sasson et al., 2009). One group of children for whom atypical sensory experiences are widely recognised are children with Autism, with an estimated prevalence of up to 90% (DuBois et al., 2007; Tomchek et al., 2007; Tavassoli et al., 2014). Historically, repetitive sensory behaviours such as rubbing surfaces; finger flicking; body rocking; repetitive jumping and using objects ritualistically discriminated Autism from typically developing children and from children with intellectual disabilities (Dahlgren and Gillberg, 1989; Rapin 1996). Many of these sensory behaviours are still considered indicative of Autism and are included in the repetitive and restricted behaviours section of the diagnostic criteria (DSM-5, ICD-11).

Children with Autism also present with a broader range of sensory processing problems spanning *all* sensory domains (Green et al., 2016). For example, Tomchek and Dunn (2007) reported that 95% of children with ASD (n=281) presented with sensory dysfunction and performed significantly differently to typically developing children on 92% of items on the Sensory Profile (SP). Auditory problems are frequently reported (Greenspan and Weider, 1993; Gillberg and Coleman, 1996; Rogers et al., 2003; Tomchek et al., 2007; Schoen et al., 2009) and recent genetic research suggests a strong link between Autism and hyper-responsivity (Taylor et al., 2018). Furthermore, significantly greater sensory seeking behaviour, greater sensory responsivity, greater sensory avoidance and increased registration problems (missing sensory cues), compared to typically developing peers, have been found (Brown et al., 2008; Joosten & Bundy, 2010; Kern et al., 2006; Reynolds et al., 2011). One larger study with children with Autism (n=400) reported patterns of hyper-responsivity in the domains of touch, movement, taste, smell, hearing and vision, as well as sensory seeking, distractibility and under-responsivity (Tomchek et al., 2014).

However, other groups of children present with sensory processing difficulties and we are particularly interested in children with Disinhibited Social Engagement Disorder (DSED) because of the ongoing clinical concern regarding assessment and discrimination of Autism symptoms from overlapping behaviours associated with DSED, (Davidson et al., 2015; Davidson, Moran and Minnis, 2022). DSED is one of two maltreatment-associated disorders of childhood, (the other being Reactive Attachment Disorder). It is characterised by over-friendliness with strangers and poor social

boundaries (American Psychiatric Association, 2013) and has an estimated prevalence of around 1% (Minnis et al, 2013). In DSM-5, DSED, (previously known as the disinhibited sub-type of Reactive Attachment Disorder, (RAD)) is regarded as a separate disorder from RAD because evidence demonstrates that DSED, unlike RAD, is not associated with attachment (Rutter et al., 2010; Zeanah and Gleason, 2015; Turner et al., 2022). DSED is now thought of as a disorder of social relatedness, (Zeanah et al., 2016) which is where the clinical conundrum with Autism arises. Children with Autism and children with DSED have been found to present with superficially similar social interaction and communication difficulties, (Rutter et al.,1999; Moran, 2010; Sadiq et al., 2012; Davidson et al., 2015; Mayes et al., 2017; Davidson et al., 2023) but DSED is associated with maltreatment (abuse/neglect) and Autism is *not* (Turner et al., 2019).

Preliminary research suggests that repetitive behaviours may be less prevalent in DSED than Autism (Sadiq et al., 2012; Mayes et al., 2017) but it is not clear whether this includes sensory behaviours. The wider 'maltreatment literature' tends not to investigate symptoms of DSED within samples; this limitation exacerbates the scarcity of knowledge regarding DSED and how clinicians can best differentiate DSED from Autism.

To the best of our knowledge, sensory processing in children with DSED has not been previously explored but based on evidence from the wider maltreatment literature, we have reason to suspect that children with DSED may present with sensory processing difficulties.

1.1. Overview of maltreatment literature with respect to sensory processing

Most maltreatment related studies investigating sensory processing have been conducted with post-institutionalised children following international adoption. Findings suggest patterns of elevated sensory seeking; especially in the domains of touch, movement, auditory and visual processing (Cermak & Daunhauer, 1997; Cermak and Groza, 1998; Lin et al., 2005; Wilbarger et al., 2010). Severe sensory seeking behaviours such as falling-down, self-spinning and rocking, which were previously associated with Autism, have been reported (Cermak and Groza, 1998; Rutter et al., 1999). Also reported are patterns of sensory avoidance, hyper-responsivity to touch, sights and sounds (Cermak &

Daunhauer, 1997; Cermak and Groza, 1998; Wilbarger et al., 2010) and under responsivity to pain (Cermak and Groza, 1998).

While the generalisation of these findings is somewhat limited due to the unique nature of the rearing environment, similar finding have been reported in community samples. A retrospective examination of 900 clinical cases of complex trauma, referred to a community assessment and treatment centre, highlighted sensory difficulties across *all* domains for up to 23% of the sample on the SP. Sensory seeking was the most prevalent pattern with 51.7% meeting criteria for a 'definite difference.' In 53.1% of cases a 'definite difference' in auditory filtering was also reported (Atchison et al., 2017). In another community sample, Armstrong-Heimsoth et al, (2021) found that 88% (n=12) of children (3-10 years) living in congregate foster placement presented with an atypical sensory processing pattern on the SP-2, but in contrast to the above, hyper-responsivity and avoiding were most prevalent. Furthermore, Cummins et al, (2021) found that adolescents in the care system tended to have hypo-responsivity to pain stimuli compared to typically developing controls.

There is also preliminary research focusing on the impact of 'type' of maltreatment on sensory processing. It is thought that threat impacts the developing nervous system via rapid response fight/flight reactions (Mclaughlin et al., 2014), whereas neglect impacts the nervous system through lack of experience of adequate stimulation (Perry, 2001). While it is very difficult to distinguish between 'types' of maltreatment, (Lacey & Minnis, 2020) some evidence appears to suggest that DSED presents where social and emotional- as well as physical neglect- occur, but the role of abuse is less understood, (Zeanah and Gleason, 2015). Hiles Howard et al, (2020) reported that greater 'definite problems' in tactile hyper-responsivity, visual and auditory hyper-responsivity and auditory filtering were found in children who had experienced abuse and greater 'definite problems' in under-responsivity and sensory seeking in children who had experienced neglect. DSED was not investigated within this sample, but given the current understanding, it would be interesting to know if children with DSED were more likely to demonstrate the sensory patterns associated with neglect.

1.1. Objectives

We aimed to explore the sensory processing profiles of children with DSED and to explore these in the context of differential diagnosis of Autism from DSED.

2. Methods

2.1. Participants

Participants were a case study sample of twenty-one children: 10 children with established diagnosis of Autism and no maltreatment history and 11 children who met DSM-5 diagnostic criteria for DSED, who were participating in a larger study regarding differentiation of Autism from DSED. However, two children from the DSED group had to be excluded from this specific investigation because they were found to also have co-occurring Autism (see Davidson et al., 2023 for further information). For this sensory processing investigation, the case study sample were: 10 children with Autism (7 males: 3 females) and 9 children (6 males: 3 females) with DSED. Nine of the ten children with Autism were referred by caregiver self-referral, following recruitment calls placed on the websites of National Autism charities and one via clinician referral. Lack of child protection concerns were established via health board electronic records. Six of the nine children in the DSED group were caregiver referred, following recruitment calls placed on the charity websites of national adoption/foster carer charities and three via clinician referral. For the DSED group, history of childhood maltreatment was established either via referring clinicians or foster carer/adoptive parent report. All participants were primary school age (age range 5-11 years) and were group matched by age.

Co-existing neurodevelopmental conditions were found in both clinical groups. Of the 10 children with Autism, two had an established additional diagnosis of Attention-Deficit/ Hyperactivity Disorder (ADHD) and were being treated with medication and in the DSED group two children had a recent diagnosis of ADHD, (not receiving treatment). Symptoms of distractibility, fidgetiness, hyperactivity and impulsiveness were reported by caregivers of the remaining children in the DSED group but were not considered to be of a severity to meet criteria for ADHD diagnosis.

2.2. Measures

Reactive Attachment Disorder and Disinhibited Social Engagement Disorder Assessment Interview (RADA) (Lehmann et al., 2020) is the DSM-5-compliant version of the Child and Adolescent Psychiatric Assessment for Attachment Disorders. It is a semi-structured interview for the diagnosis of DSED and is completed by parents/caregivers.

The Teacher Relationship Problems Questionnaire (Teacher RPQ) (Minnis et al., 2007) is a 10-item screening questionnaire for symptoms of DSED completed by teachers.

Waiting Room Observation (WRO) (McLaughlin, Espie, & Minnis, 2010): is a structured 19-item observation of child behaviour with parents/carers and a 'stranger,' carried out in a clinic waiting room. The role of the stranger, a trained rater in the WRO, is to record the interactions of the child with their caregiver, with the clinician and any interaction or social approach by the child to the stranger. In this study, the stranger was a research colleague who was not involved with the study and was unknown to the participants.

The Child Sensory Profile-2 (CSP-2) (Dunn, 1999) is a standardised caregiver report of sensory processing for children aged 3:0 to 14:11 years. There are a total of 86 items which are grouped by six sensory domains (auditory, visual, touch, movement, body position and oral) and three behavioural sections, (conduct, social emotional and attentional). The caregiver rates the frequency of occurrence on a Likert scale, and a total score is calculated per domain. The scores are converted to classifications based on normative distribution. Individual items contribute to total quadrant scores, which reflect patterns of sensory behaviour i.e., sensory seeking; avoiding; sensitivity (CSP-terminology) and registration/bystander (missing cues).

Diagnostic Interview for Social and Communication Disorders (DISCO) (Wing et al., 2002) is a standardised in-depth interview for the diagnosis of Autism which can be used across the age span. The DISCO covers a broad range of interaction and communication difficulties but three sections of part 4 relate specifically to sensory processing; 'responses to proximal sensory stimuli,' (section ii) 'responses to auditory stimuli' (section iii) and 'responses to visual stimuli' (section iv.). Each section is individually scored and can be descriptively examined separately from the overall diagnostic algorithm.

2.3: Ethics and Procedure

Research ethics approval was obtained from the West of Scotland Research Ethics Committee and the lead author achieved funding from the Castang Foundation (UK), [LAY1-WS_LEGAL.FID1730709, the Gillberg Neuropsychiatry Centre, and NHS Greater Glasgow and Clyde.

To confirm symptoms of DSED met DSM-5 diagnostic criteria, parents/ caregivers first completed the RADA, via telephone. Then the Teacher RPQ was sent out and the WRO was completed in clinic. Children were enrolled into the DSED group if their symptoms met diagnostic criteria on this standardised multi-informant package.

Participants in both groups completed the DISCO in the first instance, which was administered by a Speech and Language Therapist. Parents/caregivers completed the CSP-2 at the next session.

2.4. Analysis

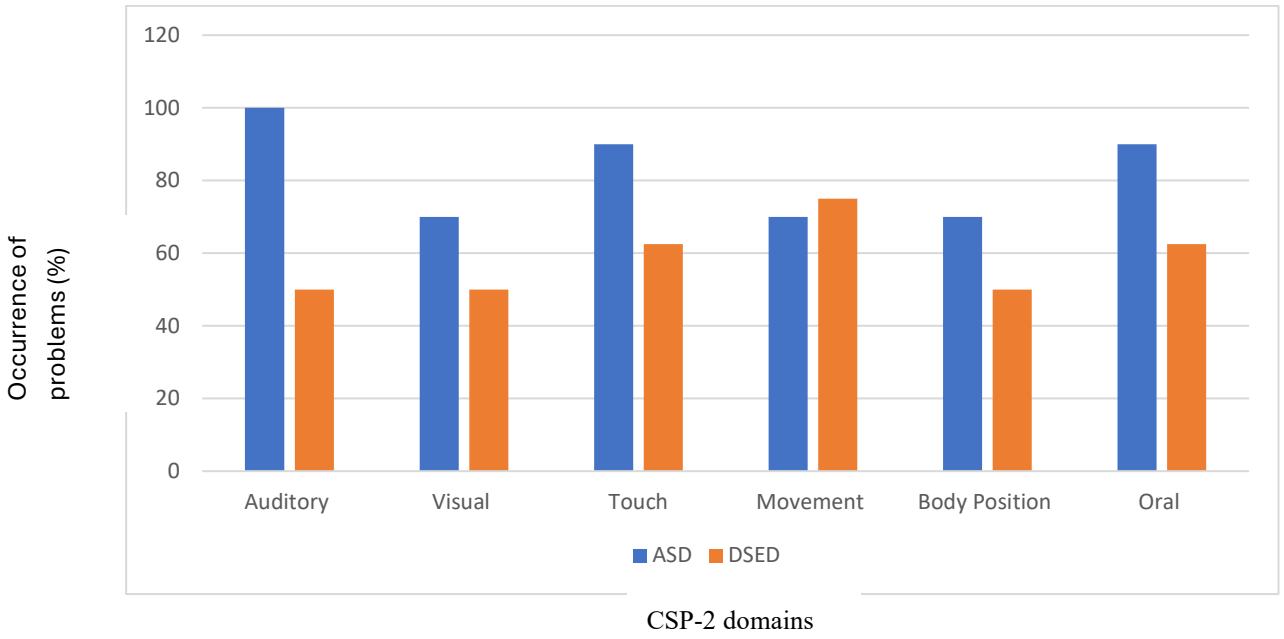
We have not completed statistical group analysis due to the sample size but instead explored the group outcomes descriptively. The proportions per group who scored out-with the normative distribution on the standardised measures are described. Given the sample size, this descriptive analysis also provides a more accurate representation of the distribution of sensory processing differences than means-testing.

3. Results

3.1 Outcomes per diagnostic group: Child Sensory Profile-2 (CSP-2)

All ten parents of children with Autism and eight of the nine caregivers of children with DSED completed the CSP-2. Figure 1 demonstrates the proportion of children in each group whose total score, per domain, fell out-with the normative range (less than others/more than others/much more than others).

Figure 1: Percentage scoring out-with normative range, per group, in each sensory domain.



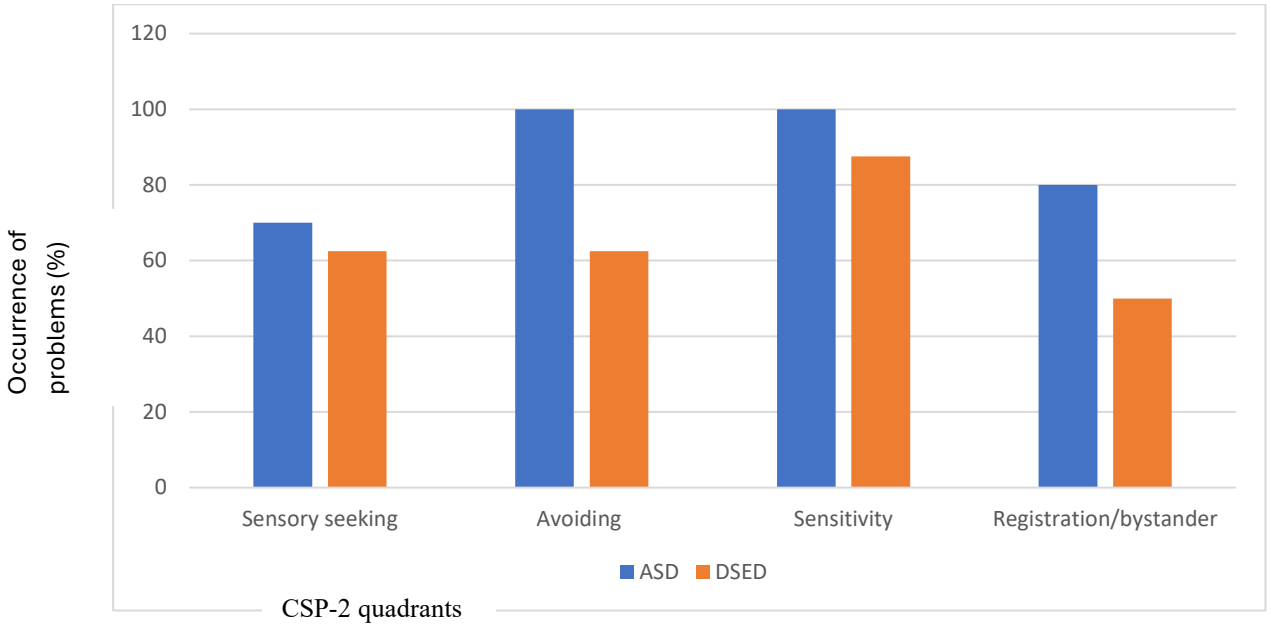
Autism group: the children with Autism experienced sensory differences in *all* domains and the proportion of children was generally greater than the DSED group. A minimum of 70% of the total scores, in the Autism group, fell out-with the normative range in each domain. Auditory processing difficulties were the most prevalent area of difficulty (present in 100%), but both touch and oral processing difficulties were also very apparent (90%).

DSED group: There is a wider distribution of scores within the DSED group but differences in the domain of movement were consistently found (75%). Movement questions tend to regard over-activity or seeking movement, and, in this sample, problems are marginally higher than in Autism. Difficulties with touch and oral processing were also reported in more than half the group (62.5 % respectively) and a slightly smaller proportion presented with auditory, visual and body positioning differences (50%). Bearing in mind the small sample, it appears that some children with DSED may present with similar problems to children with Autism, but the prevalence in Autism is generally greater.

The CSP-2 also provides quadrant scores which demonstrate patterns of behaviour (seeking, avoidant, sensitivity (terminology used within the CSP-2) and registration/bystander). Briefly, children with 'more than/much more than' scores in the

sensory seeking quadrant actively engage in or seek out stimuli. 'More than/much more than' scores in the avoiding quadrant will move away or react negatively to the stimuli. 'More than/much more than' scores in the sensitivity quadrant are children who are more aware of or notice stimuli more and 'more than/much more than' scores in the registration/bystander quadrant means the child is less tuned into or may miss stimuli. 'Less than others' suggest the opposite behaviour in each quadrant. (Dunn, 1999).

Figure 2: Percentage scoring out-with normative range in each group, per quadrant



Autism group: children with Autism, as a group, demonstrate *all* patterns of sensory behaviour and a greater proportion of children with Autism showed differences compared to children with DSED. Findings are also consistent within the group, with avoiding and sensitivity patterns present in *all* children with Autism. In fact, *100%* of the children with Autism scored within the more severe classification range 'much more than others' for the sensitivity quadrant suggesting that the children with Autism have greatly heightened awareness/impacted by sensory stimuli around them.

DSED group: More than half the children with DSED presented with patterns of sensory seeking, avoiding and sensitivity. Similar to children with Autism, sensitivity was the most consistent pattern of sensory behaviour within the group (87.5%), but in contrast to Autism, who *all* scored as having heightened sensitivity, in DSED, one child scored as being under-sensitive to stimuli while the remaining were reported to have heightened

sensitivity. Of interest is also the prevalence of sensory seeking behaviour (62.5%), which in this sample, is similar to children with Autism (70%).

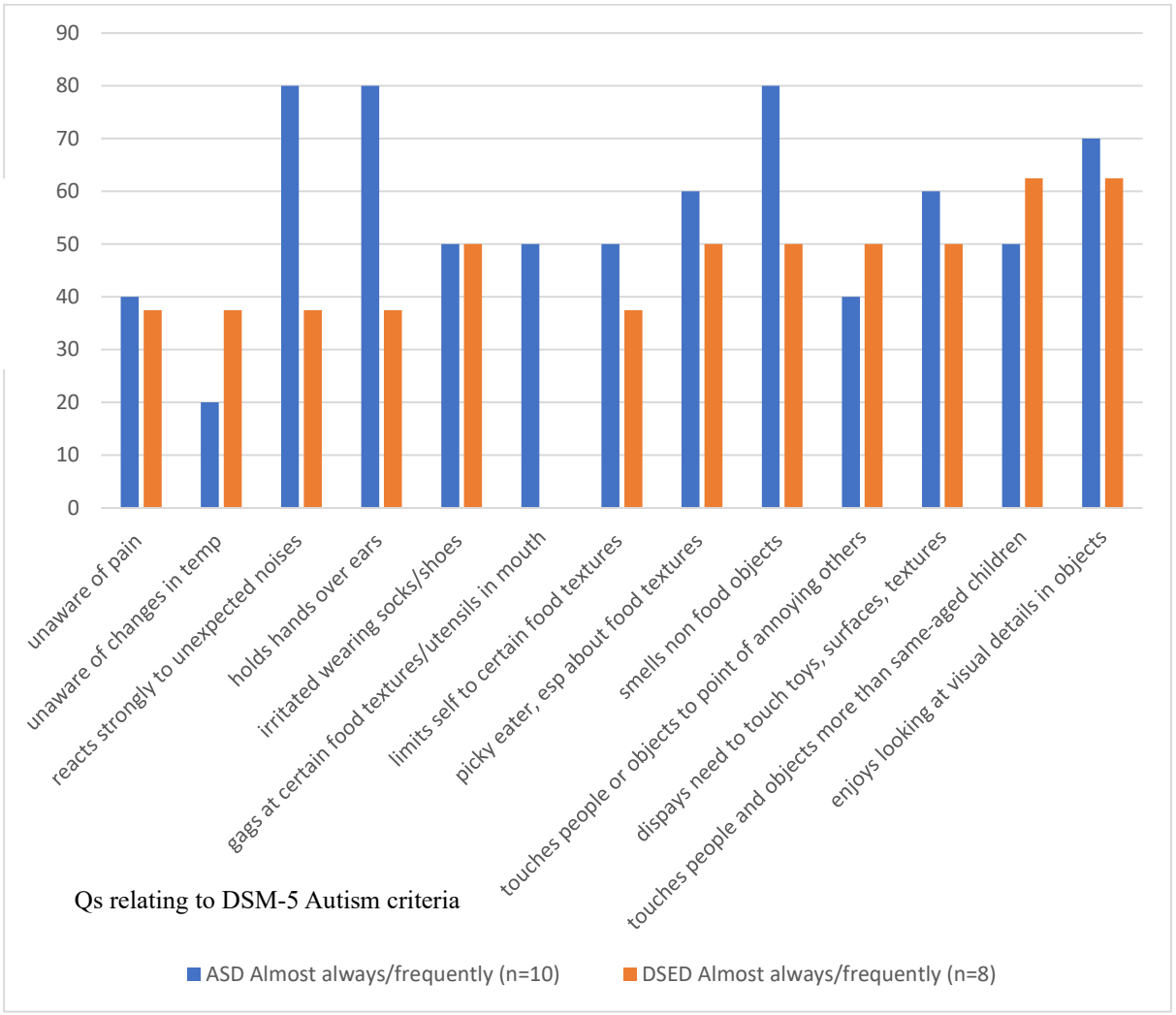
3.2. *Outcomes according to DSM-5 Autism diagnostic criteria for sensory processing*

To further investigate whether children with DSED share similar or differing sensory processing characteristics to children with Autism, we identified individual items from the CSP-2 and the DISCO which specifically relate to the DSM-5 Autism sensory criteria. The DSM-5 states that children with Autism may demonstrate '*hyper- or hypo-reactivity to sensory input or show unusual interests in sensory aspects of the environment*.' The specific examples cited are: apparent indifference to pain/temperature; adverse response to specific sounds; adverse response to specific textures, excessive smelling of objects; excessive touching of objects; visual fascination with lights and visual fascination with movement.

We explored the CSP-2 first, identifying thirteen questions which ask about any of the above DSM-5 items. Figure 3 demonstrates the proportion of children in each group who were rated as 'almost always' or 'frequently' displaying each behaviour.

Figure 3

Questions from the Child Sensory Profile-2 relating to DSM-5 Autism sensory criteria



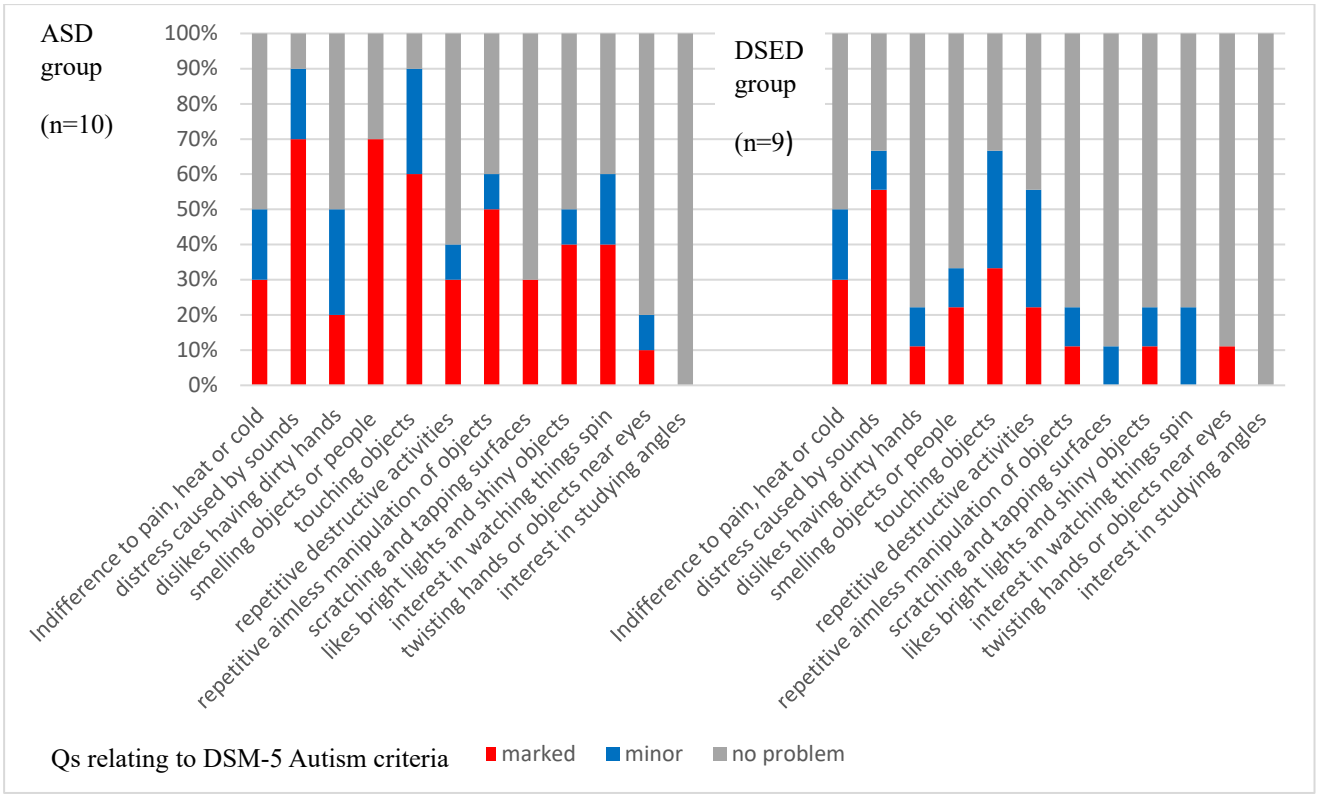
Autism group: A few items stand out. 'Reacts strongly to unexpected noises,' 'holds hands over ears' and 'smells non-food objects' were present in both groups, yet these issues were clearly more prevalent in children with Autism (80% for each item). Of note, 'gags easily at textures or utensils in their mouth' was found *only* in the Autism group.

DSED group: For some items, the proportion of children in the DSED group and the Autism group experiencing the problems are *similar*. Furthermore, problems with 'unaware of changes in temperature', 'touches people/objects to the point of annoying others,' and 'touches people/objects more than same-aged children' are slightly higher in the DSED group.

Next, we explored items on the sensory sections of DISCO which relate to the DSM-5 ASD sensory criteria. Several questions on the DISCO overlap with the CSP-2, but the DISCO measures severity of problem, rather than frequency of occurrence, thus offering an interesting comparison. The perceived severity of the problem is rated by the caregiver as either 'marked', 'minor' or 'no problem.' Of note, we have DISCO outcomes for *all* participants in the clinical groups, therefore the DSED group has increased from eight to nine children for this measure.

Figure 4

Questions from the DISCO directly relating to DSM-5 ASD criteria



The DISCO generally supports the outcomes of the CSP-2 and together both suggest that children with DSED do demonstrate sensory processing difficulties in areas considered to be indicative of Autism. Furthermore, 'indifference to heat or pain' and 'repetitive destructive activities,' were rated similarly across both groups. As in the CSP-2, 'distress caused by sounds' was more prevalent in children with Autism and was rated as a marked problem in 70% of Autism cases (7/10), but it was also rated as marked in 55% (5/9) of the children with DSED; caregivers in *both* groups actually described their

children covering their ears with their hands and in both groups the distressing sounds were almost identical: hand dryers, fireworks and the school bell.

One difference between the DISCO and CSP-2 is the inclusion of additional visual sensory seeking items, 'likes bright lights and shiny things,' 'interest in watching things spin' and 'twisting hands or objects in front of eyes' which are items that are a bit more 'unusual' and some of which fit with the early repetitive behaviours described in Autism. These items were not only rated at a greater severity in children with Autism, but generally were more prevalent in the Autism group compared to the DSED group. This may account for why children with Autism scored lower on the visual processing domain, compared to the other domains, on the CSP-2 as these results suggest they may tend towards seeking visual stimuli rather than missing or avoiding it.

The other repetitive items, 'tapping or scratching surfaces/textures' and 'repetitive/aimless manipulation of objects' were also more prevalent and severe in the Autism group compared to the DSED group. The specific nature of the former touch related items contrasts with the more general item on the CSP-2, 'touching people/objects to the point of annoying others' which was slightly more prevalent in DSED than Autism.

4. Discussion

We conducted an exploratory investigation with a case study sample using standardised measures to investigate the sensory profiles of children with DSED compared to children with Autism without maltreatment histories. Our study is limited by the small sample size, which does not lend itself to statistical comparisons, yet a relative strength is the consistency of findings across some of the domains, suggesting that statistical differences may have been found with a larger sample. Our sensory processing data is also based on caregiver report only, although the comparison of two different standardised measures adds weight to the findings.

Despite these limitations, this is an important first step because, to the best of our knowledge, the sensory processing patterns of children with DSED have not been previously investigated. This is despite evidence that both post-institutionalised adoptees (Cermak and Groza, 1998; Lin et al., 2005; Wilbarger et al, 2010) and community based fostered/adopted children (Atchison, 2007; Armstrong-Heimsoth, 2021) present with

sensory processing problems across multiple domains. This is also the first study to explore the sensory processing problems of children with DSED, in comparison to children with Autism, which is extremely relevant given the ongoing clinical concerns regarding discrimination of Autism from DSED during assessment (Moran 2010; Davidson, Moran and Minnis, 2022).

Our first objective was to explore the sensory processing profiles of a case study sample of children with DSED. While there was individual variation among the DSED group, more than half the group reflected the patterns previously described in post-institutionalised children and in fostered/adopted community samples. These were, increased sensory differences in the domains of touch and movement (Cermak & Daunhauer, 1997; Cermak and Groza, 1998; Lin et al., 2005; Wilbarger et al., 2010), greater sensory seeking behaviour (Cermak & Daunhauer, 1997; Cermak and Groza, 1998; Lin et al., 2005; Atchison, 2017), under-responsivity to sensing pain (Cermak and Groza, 1998; Cummins et al., 2020) and/or changes in temperature. Fifty percent of the sample also presented with auditory and visual processing difficulties, previously found by Cermak & Daunhauer. Findings also reflected reports of increased avoidance and hyper-responsivity compared to typically developing children (Armstrong-Heimsoth et al., 2012; Cermak & Daunhauer, 1997; Cermak and Groza, 1998) and additionally we found some increased problems in the oral processing domain. The findings somewhat reflect the preliminary research regarding 'types' of maltreatment as DSED is thought to be associated with neglect, at least, (Zeanah and Gleason, 2015) and the DSED group most consistently presented with the associated neglect- associated pattern of greater sensory seeking behaviours (Hiles Howard, 2020) but the other prevalent difficulty was hyper-responsivity and this been found to be more associated with abuse. This is an area, clearly warranting further research, and it must be considered that presence of one 'type' of maltreatment does not negate presence of another and are often inter-related (Lacey and Minnis, 2023). In addition, further studies with a larger sample are required to test the reliability of all presented findings.

Our second objective was to explore the sensory profiles of children with DSED in comparison to children with Autism without maltreatment histories. Some overlap in the sensory problems were demonstrated in both groups and areas of difference were nuanced. For example, similar problems were demonstrated on the CSP-2 in both groups but the difference was less consistency of the problem in the DSED group compared to

the Autism group. Similarly, problems on the DISCO in DSED were described to be less severe than in the Autism group. That said, the children with DSED, as a group, did consistently present with sensory hyper-responsivity (87.5%) which is a recognised difficulty in Autism (Taylor et al., 2018; Tomcheck et al., 2014) and children with DSED also presented with sensory seeking behaviour (62.5%) to a similar degree as children with Autism (70%). The latter finding has direct clinical relevance as some standardised Autism diagnostic tools, like the Autism Diagnostic Observational Assessment-2 (ADOS-2) (Lord, et al., 2012), briefly assess sensory seeking behaviour by including the broad item, 'unusual sensory interest in play materials/person,' in the restricted and repetitive behaviours section of the diagnostic algorithm. While this is appropriate for assessment of possible Autism, some of the children within our sample with DSED may also score positively on this item. It is imperative that clinicians are aware of this possible overlap, especially when using instruments like the ADOS-2, to prevent misdiagnosis.

Our third aim was to identify any areas of sensory processing which differed between children with Autism and DSED. In our sample, the proportion of children with auditory processing difficulties, hyper-responsivity and avoidance of stimuli in Autism differed from DSED. *All* the children in the Autism group had problems within these areas, compared to just some of the children with DSED. These findings add to the already wide literature that suggests that children with Autism are at higher risk of experiencing auditory processing problems and hyper-responsivity (Greenspan and Weider, 1993; Gillberg and Coleman, 1996; Rogers et al., 2003; Tomchek et al., 2007; Schoen et al., 2009). Furthermore, the children with Autism presented with the full range of auditory difficulties, as measured by the CSP-2 but in DSED problems tended to overlap with one specific item, 'distress caused by sounds.' This is of note though, as caregivers in *both* groups reported the child covering their ears; a behaviour clinically associated with Autism. Other specific areas of overlap from the DSM-5 Autism diagnostic criteria were indifference to pain, previously described by Cummins et al and Cermak and Groza, indifference to temperature changes and repetitive destructive activities.

Specific behaviours, assessed via DISCO, which stood out as more common in the Autism group and are worthy of investigation with larger samples were visual and olfactory sensory seeking items (fascination with bright objects or lights, interest in watching things spin and smells objects/people). Furthermore, 'gags at textures/utensils

in mouth (item on CSP-2 and DISCO), was present *only* in the Autism group. Other behaviours on the DISCO, such as repetitive/aimless manipulation of objects and repetitive tapping or touching textures' also supported findings from the literature that repetitive behaviours tend to be more indicative of Autism (Adrien, 1987; Dahlgren and Gillberg, 1989; Rapin 1996; Sadiq et al., 2012; Mayes et al., 2017).

The findings related to touch processing above are interesting because, the CSP-2 suggested children with DSED were inclined to touch people or objects, to the point of annoying others, perhaps even more than children with Autism but, the findings from both measures put together, seem to suggest that the *quality* of the seeking touch may be of most relevance for differential diagnosis; it was the repetitive nature and the need to touch *textures* which was the difference between Autism and DSED. Further research focusing on the quality of specific sensory differences between children with Autism and DSED would be extremely beneficial, especially in relation to the sensory seeking element of the ADOS algorithm.

4.1. Implications for clinical practice and future research

As most of the children with DSED presented with more sensory differences than typically developing children, it seems vital for clinicians working with children with a maltreatment history to consider assessment for DSED, and in the context of sensory processing, and to consider joint working with colleagues such as Occupational Therapists who specialise in assessing and supporting impact of any sensory difficulties.

Related to clinical management, and to our findings, is the role of other co-existing neurodevelopmental conditions. In both children with Autism and children with maltreatment associated problems like DSED, co-morbidity is thought to be the norm rather than the exception (Gillberg, 2010; Minnis, 2013), yet few studies from the maltreatment literature consider possible occurrence of neurodevelopmental conditions in their sample. There is a growing body of studies suggesting that ADHD is one of the most common co-existing conditions with DSED (Kennedy et al., 2017; Seim et al., 2022) and was apparent in two DSED cases in our sample. Given the core symptoms of ADHD include distractibility, fidgeting, restlessness and hyper-activity which behaviourally may be observed as frequent movement, inability to sit still and touching things, it appears there could be an overlap between ADHD 'traits' and many of the sensory problems reported in the children with DSED. Further research is required to

better understand the sensory seeking behaviours of children with DSED and in the context of possible ADHD, or not, to ensure the most effective management and treatment.

Finally, regarding differential diagnosis from Autism, it appears to be beneficial for clinicians to identify both a broad understanding of sensory processing patterns *and* be very specific; clinicians may choose to include sections of measures, like the DISCO, which ask in more detail about items that are perhaps more 'Autism-specific' compared to the CSP-2.

Davidson et al, (2023) report that clinical observation using a format of interaction between the child and assessor (s) which is both unstructured and offers greater social challenge is a helpful approach to differentiating Autism from DSED, especially in cases where Autism symptoms tend to be more subtle. Perhaps there is potential for clinicians to actively include tasks and direct discussion which elicit/assess sensory patterns and/or creatively 'test' out some of the items identified in the DISCO, which appear as more 'Autism-specific.' This may help bring clarity for clinicians trying to determine if any reported sensory differences are in keeping with Autism diagnostic criteria.

5. Conclusion

Children with DSED may present with greater than expected sensory difficulties and regarding touch, movement and oral processing these problems were apparent in more than half the sample. In our case study sample, the degree and severity of most sensory difficulties were greater in Autism, but hyper responsivity, sensory seeking, indifference to pain and temperature, distress at sounds and repetitive destructive behaviours presented to a similar degree in both groups. The items which did present more in the Autism group, compared to the DSED group were individual items regarding repetitive behaviours, visual sensory seeking and smelling objects/people and 'gags at textures/utensils in mouth' was found *only* in the Autism group. We encourage clinicians to consider assessment for DSED in maltreated children and consider relative impact of sensory processing, and possible additional neurodevelopmental conditions. Regarding differential diagnosis of Autism from DSED, assessing sensory processing both broadly and specifically, perhaps including sections of Autism-diagnostic measures, like the DISCO, which detail 'Autism-specific' sensory behaviours may be helpful for clinicians. Investigation of the behaviours, in the case study sample, which differed

between Autism and DSED with larger samples would be a vital next step in determining whether such differences could be useful discriminators.

6. Credit authorship statement

Claire Davidson: Conceptualisation, methodology, investigation, analysis, writing, writing- review and funding acquisition. Christopher Gillberg: Conceptualisation, methodology, supervision, review of writing. Anja Lowit: Conceptualisation, methodology, supervision, review of writing. Helen Minnis: Conceptualisation, methodology, supervision, review of writing.

7. Declarations of Competing Interest

None.

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Reflections on Paper 4

We explored whether the children with DSED, in the case study series, had similar sensory processing problems to children with Autism as this had not been previously investigated. Again, useful insight was gained regarding the needs of children with DSED. Some sensory behaviours overlapped with Autism, highlighting the need for caution and further investigation. Other behaviours were also more common in the Autism group than the DSED group and may be areas for future study with regards to differentiation. The most relevant points are as follows:

- The findings demonstrate consistency with the wider maltreatment literature. Children with DSED present with greater sensory processing problems across all domains.

- Greater than 'average' sensory seeking behaviour and sensory sensitivity were found in this sample of children with DSED. Sensory seeking behaviour and sensory sensitivity may unlikely be helpful in discriminating between DSED and Autism.

- The children with DSED also consistently presented with some of the difficulties specifically identified in the Autism DSM-5 criteria. In this sample, the severity of these difficulties were greater in the Autism group but this is a subtle difference which again highlights the need for caution.

- Some of the sensory processing questions from the DISCO, which is designed more specifically for Autism assessment, identified items which presented more commonly in this group of children with Autism and compared to the children with DSED. These were related to visual sensory seeking, touching objects for texture (as opposed to simply touching things/people), repetition of behaviours and gagging at textures in mouth; the latter was only found in the Autism group. Nevertheless, without replication with larger samples it is not known whether these differences are inherent to this specific sample or whether they could be indicators of differentiation between the groups.

Like paper 3, these findings also support areas of unmet need in children with DSED and add further weight to the argument for a holistic ESSENCE approach to assessment, ensuring that like language and communication skills, sensory processing problems are not being missed by relevant professionals in children with DSED.

Chapter 7: Discussion and conclusion summary

7.1 Overview of section

This thesis aimed to explore the profiles of a case study sample of children with Autism and DSED in the context of differential diagnosis of Autism from DSED. The DSM-5 Autism diagnostic criteria consists of a dyad of impairment – impaired social interaction and communication and presence of repetitive and restricted behaviours- which are considered to be the core symptoms of Autism. This dyad was used as a springboard to identify where the gaps in our understanding of DSED compared to Autism were, and how better understanding of these may help discriminate Autism from DSED in clinical and research practice.

Within the social interaction and communication domain of the dyad, it was apparent that a scarcity of research regarding the broader social functioning of children with DSED existed. Furthermore, if children with DSED, like other maltreated children, were at higher risk of social relationship and interaction problems, (Cicchetti et al., 2016), the role of the current 'gold standard' Autism diagnostic tools in discriminating these problems from the social interaction difficulties of children with Autism was unknown. The previous literature seemed to suggest that unstructured observation which included conversation and play, (Moran, 2010; Davidson et al., 2015) may be more beneficial in discriminating symptoms of Autism from DSED but these findings required replication. The receptive and expressive language skills of children with DSED had also not been examined previously, nor in comparison to children with Autism. It was unknown if children with DSED presented with similar language difficulties, as suggested in the wider maltreatment literature, (Carr et al., 2020), and how such problems might impact pragmatic language/broader social communication skills. The latter, is of course, pertinent for our understanding of differential diagnosis, as pragmatic language is considered to be universally impaired in Autism, (Rhea, 2007).

Regarding the 2nd part of the dyad of impairment in Autism, repetitive and restricted behaviours, the previous literature seemed to suggest that repetitive and stereotyped behaviours may be more apparent, and pervasive, in children with Autism compared to children with DSED^{RAD}, (Rutter et al.,1999; Sadiq et al., 2012; Mayes et al., 2017) but there was scant information with regards to sensory processing difficulties in children

with DSED, or in comparison to sensory problems found in Autism. This was despite evidence that maltreated children post-institutionalisation *and* in community samples, generally present with a broad range of sensory processing difficulties, (Cermak and Groza, 1998; Atkinson, 2017).

The overall aim of the thesis was to explore the profiles of children with Autism and DSED in the context of differential diagnosis of Autism from DSED. Four research question were identified which both addressed these gaps in knowledge and supported the overall aim of the thesis. There were as follows:

1. What are the broad social functioning difficulties, if any, of children with DSED? (Paper 1).

2. Do current multi-informant 'gold standard' Autism diagnostic tools support differential diagnosis of Autism from DSED, and how does the structured ADOS-2 assessment compare to an unstructured behavioural observation, in this context? (paper 2)

3. What is the profile of children with DSED regarding receptive, expressive and pragmatic language, and can any differences be identified compared to children with Autism which may support discrimination? (paper 3)

4. What are the sensory processing profiles of children with DSED, compared to children with Autism?

Each of the research questions above was addressed by one of the papers presented in this thesis. The next section will provide a brief summary of the findings relating to each research question. This is followed by a discussion of the key messages relevant within a clinical and research context, and suggestions for future research. The chapter comes to a close with a discussion of the ethics procedure and opportunities to overcome governance barriers and concludes with overall strengths and limitations of this thesis.

7.2 Summary of Findings

What are the broad social functioning difficulties, if any, of children with DSED?(Research question 1, paper 1): A systematic review of the literature demonstrated that children with DSED may be high risk for additional social problems. Findings included poorer social competency across multiple domains, greater peer problems and conflicts in relationships and poorer self esteem/self-concept related to social functioning in children with DSED compared to typically developing peers. Findings regarding social communication were mixed; there was some suggestion of pragmatic language difficulties, but specific interaction skills such as humour, non-verbal communication, non-literal understanding and back and forth social exchanges were found to be better in children with DSED compared to children with Autism.

Do current multi-informant 'gold standard' Autism diagnostic tools support differential diagnosis of Autism from DSED, and how does the structured ADOS-2 assessment compare to an unstructured behavioural observation, in this context? (Research Question 2, paper 2): Outcomes suggested that the Autism diagnostic tool, the DISCO assessment, (caregiver interview) did not discriminate between children with Autism and children with DSED but the standardised observation tool, ADOS-2, discriminated to some degree. In 'straightforward' cases of DSED (no additional co-morbidity), the ADOS-2 discriminated between Autism and DSED, but false positive outcomes were found for cases of DSED with moderate to severe ADHD symptoms (more complex cases). There is a danger that maltreated children could be misdiagnosed with Autism instead of DSED if clinicians are not aware of the lack of discriminatory ability of these tools, especially for complex cases. Equally, Autism could be missed if clinicians hold on too tightly to the notion that the tools tend to 'over diagnose' Autism in maltreated children and are not open to the possibility of both maltreatment/DSED and Autism and/or other neurodevelopmental conditions. This may be helped by a more flexible approach to assessment. For example, the unstructured, and more socially challenging, Live assessment more easily supported discrimination between DSED and Autism in *all* cases because the unstructured nature increased unpredictability and pushed the children to use their social skills, where present. Therefore, the better communication/interaction skills of the children with DSED stood out, and the difficulties of the children with Autism were exacerbated. Furthermore, the free-flowing dynamic enabled additional ADHD related behaviours to come to the fore, with less disruption to social skills than the seated and structured ADOS-2.

What is the profile of children with DSED regarding receptive, expressive and pragmatic language, and can any differences be identified compared to children with Autism which may support discrimination? (Research Question 3, Paper 3): Findings demonstrated that receptive vocabulary skills were not especially impaired in a larger sample of either children with Autism or DSED when verbal IQ was accounted for, but when receptive *language* was investigated with the case study sample, which included semantic reasoning, some of the children with Autism (average verbal IQ), had much greater difficulty. In contrast, the children with DSED tended to perform more poorly than children with Autism on standardised assessments of expressive language and when error patterns in conversational speech, from the Live assessment, were analysed by SLTs, 50% of the children showed a similar number of errors to children with Autism, (7 or less) and 50% showed greater, (8-17 errors). Furthermore, the children with DSED showed a much wider range of non-developmental errors in their speech than the children with Autism. Regarding pragmatic language, on caregiver report, the children with DSED had a similar range of problems as children with Autism. SLT analysis of their conversations, however, demonstrated that both groups showed overlapping problems regarding narrative discourse but the children with Autism demonstrated additional impairments that were not present in the DSED group (lack of use of /range of -wh questions, limited speech acts, reduced ability to draw inference from context, literal thinking, idiomatic speech and echolalia). These difficulties are patterns that are already associated with Autism, (Baird and Norbury, 2016) and most Autism experienced SLTs would be able to identify these. Papers 2 and 3 together suggest that specific areas of social interaction and social communication differ in children with Autism compared to children with DSED, but these are better identified during observation/analysis of unstructured conversation and play. Samples were necessarily small due to the intensity of the assessments, but this is a potentially fruitful area for future larger-scale research.

What are the sensory processing profiles of children with DSED, compared to children with Autism? (Research question 4, Paper 4): Sensory processing problems broadly overlapped between children with Autism and children with DSED, but the sample size was particularly small and the differences were subtle. For example, the severity tended to be greater in Autism compared to DSED. For these reasons, caution is warranted in interpretation. In the case series sample, ome specific items from the DISCO assessment, which is more Autism specific, were more prevalent in Autism compared to DSED. Such items related to behaviours which to be more repetitive in nature and regarded specific processes i.e., visual fascination with objects, touch of textures and gagging at foods/utensils in mouth -the latter was found *only* in the Autism group. It

is not known if these differences are due to characteristics inherent to this specific sample or whether the same differences would be found in another sample. Yet, the consistency of the findings within the group suggests that investigation of these differences with larger samples could be worthwhile – the CSP-2 item, 'gags at utensils/textures in mouth' found only in the Autism group, as mentioned above, is just one example of this consistency.

Overall, the outcomes from this thesis have contributed to the gaps in knowledge, identified areas of overlap between Autism and DSED and, importantly, added further to the knowledge base regarding possible areas and methods of differentiation, and directs future research focus to these areas.

Additional questions have also been raised which offer interesting areas for future study; one example being the inter-play of additional neurodevelopmental conditions with DSED and what this means for both child outcomes and supportive interventions. In the remainder of this chapter, the findings of this thesis are brought together through a discussion of key messages and implications.

7.3 Overall message 1:

Social relationship difficulties overlap between Autism and DSED, but areas of differentiation can be found in specific areas of social communication, and regarding some repetitive/ sensory behaviours.

Two key findings evidence this overall message:

Key finding 1a: Problems with social relatedness do not differentiate Autism from DSED; **paper 1** demonstrated that children with DSED, like children with Autism, may have poorer social competencies across multiple domains, including difficulties with friendships.

Key finding 1b: Children with Autism had greater difficulties than children with DSED regarding broad social communication skills, **(paper 2)** and areas of receptive language and pragmatic language **(paper 3);** while a few sensory processing behaviours appeared more Autism-specific **(paper 4).**

7.3.1. Key finding 1a: Problems with social relatedness do not differentiate Autism from DSED.

Indiscriminate behaviours and poor social boundaries are core feature of both Autism and DSED (DSM-5). Paper 1 now adds to the knowledge base by demonstrating that children with DSED, like children with Autism, have additional difficulties with social functioning, across multiple domains (Millward et al., 2006; Pritchett et al., 2013; Guyon-Harris et al., 2019). For example, children with DSED have difficulties forming friendships and experience greater peer conflicts (Guyon-Harris et al., 2019; Raaska et al., 2012; Bennet et al., 2009), they may have poorer self-esteem related to social relationships (Seim et al., 2021; Vacaru et al., 2022) and like children with Autism, these social difficulties may persist over time (Guyon-Harris et al., 2019).

In Autism, social relationship difficulties are thought to be largely attributed to impaired theory of mind (ToM), (Baron-Cohen; Tager-Flusberg, Ozonoff and McEvoy, 1994) weak central coherence and/or impaired social imagination (Wing, Gould and Gillberg, 2021). Studies from the wider maltreatment literature have found that some children who have experienced maltreatment perform more poorly on false belief tasks than peers without experiences of maltreatment (Cicchetti et al., 2003; Tarullo et al., 2007; O'Reilly & Peterson, 2015; Pears et al., 2015) but, in DSED, ToM has largely been investigated with regards to its role in the underpinning of core DSED symptoms, rather than its social implications. A small number of studies suggest that theory of mind is not associated with *core symptoms* of DSED, i.e., indiscriminate behaviours (Colvert et al., 2008; Elovainio et al., 2015), although Fallon et al, (2018) found a negative association between theory of mind and DSED behaviours, when utilising a novel first person perspective via a computer-based role play. Only one study investigated ToM in DSED with regards to social functioning. Kay and Green (2016) found that theory of mind may play a part in other social difficulties of children with DSED, such as attributions regarding friendships (Kay and Green, 2016). Within the wider maltreatment literature, one recent systematic review reported mixed findings regarding social attributions, with some studies reporting increased hostile attributions, (aggression towards peers etc.), others reporting gender difference and others finding no significant difference between maltreated and non-maltreated children. Several methodological limitations were identified, however, and included lack of investigation of co-occurring psychiatric conditions (Benarous et al. 2015). Lack of investigation of co-occurring Autism or investigation of DSED could be added as a further limitation. The implications of ToM on core DSED symptoms and on social functioning is clearly an area worthy of future

research. If core DSED symptoms are not associated with theory of mind, it may suggest that the indiscriminate behaviours of children with DSED and children with Autism appear superficially similar but occur for different reasons. For example, Davidson et al, (2015) found that children with Autism were likely to make social approaches to speak about something that *they* were interested in - demonstrating lack of theory of mind in accounting for the interests of the other person, whereas an attempt to 'get to know you,' albeit in an inappropriate manner, has been described in children with DSED, (Minnis et al., 2009). Papers 2 and 3 also showed that the social interactions of children with DSED, compared to children with Autism, were more socially reciprocal, even when lack of empathy and controlling behaviour, were at other times apparent.

Kay and Green, (2016) hypothesise that poor inhibitory control, which has been associated with DSED in pre-school and school age children (Bruce et al., 2009; Pears et al., 2010) may underpin core DSED symptoms (indiscriminate behaviours), but that difficulties with theory of mind impact more broadly on poorer social skills. It would be interesting to test whether the association between greater DSED behaviours and poorer theory of mind found by Fallon et al, (2018) had any bearing on broader social assertions made by the children within the computer paradigm or whether they simply related to indiscriminate behaviours. Furthermore, testing of Kay and Green's hypothesis would help with answering the question about the underlying 'cause (s)' of social problems in DSED compared to Autism; although, co-existence of neurodevelopmental problems, including Autism and ADHD with DSED make these questions much more complex to answer.

Key message for clinicians and researchers*:*

- It is imperative that clinicians are aware that several of the relational difficulties within the DSM-5 Autism diagnostic criteria can be indicative of both Autism and DSED. For example, like children with Autism, children with DSED present with poorer social competencies including difficulties with peers.

Key message for researchers

- Children with DSED present with poorer social competencies than non-maltreated peers, and some studies suggest they may have greater social

problems than children with RAD or maltreated children without DSED or RAD. In order to better support these children, we need to identify DSED within samples of maltreated children when investigating social functioning.

- Strengthening our knowledge of the role of cognitive abilities such as theory of mind in the social functioning of children with DSED, and the relative impact of impaired inhibitory control, is an important area of future research for both understanding possible underpinnings of DSED and how they differ from Autism.

7.3.2. Key finding 1b: Children with Autism had greater difficulties than children with DSED regarding specific areas of social communication and language and some sensory processing behaviours may be more Autism-specific.

Sadiq et al, (2012) expanded the communication literature by demonstrating that children with DSED presented with pragmatic language impairments similar to children with Autism, when assessed via the CCC. However, the findings in this thesis, together, present a clearer picture of the communication skills of children with Autism compared to children with DSED. While language and social communication difficulties are undoubtably present in DSED, specific skill differences within the social communication domain also exist. These are best identified via observation, which includes free flowing conversation. In most situations, any clinician skilled in Autism assessment can do this because the differences tend to relate to difficulties already synonymous with social communication difficulties in Autism but, in tricky cases, the expertise of an SLT would be particularly helpful in discrimination (Baird and Norbury, 2016).

Children with DSED, compared to children with Autism, better used nonverbal communication skills (paper 2), engaged in back and forth communication, using a range of speech acts, including inquiring about others experiences (paper 2 and 3), better understood humour and showed better imagination skills (paper 2), understood and used metaphorical language (paper 2 &3) and were able to better draw inferences in conversation (paper 3). In contrast, literal interpretation and idiomatic speech were more present in the Autism group than the children with DSED, who did not have expressive language difficulties, (paper 3) and echolalia was present *only* in the Autism group

(paper 3). The latter finding replicates the experiences reported in the Coventry grid (Moran, 2010) and findings build on the early work of Rutter et al, (1999) who reported that spontaneous social communication and flexibility in social communication discriminated the children with DSED (then called Quasi-Autism) from children with Autism.

There is also tentative evidence to suggest that, in some cases, receptive *language* skills, (not receptive vocabulary), maybe poorer in children with Autism, (without ID), compared to children with DSED, particularly when semantic reasoning is required (paper 3). This finding does fit with previous literature regarding Autism ((Tager-Flusberg et al., 2005), but further research is required as some studies have reported receptive language difficulties in maltreated children (Sylvestre et al., 2016). Also, the sample in paper 3 was small so generalisation might be impaired by biases within that sample.

Regarding differences in presentation between Autism and DSED within the restricted and repetitive behaviours domain of the dyad of impairment, the findings suggest that need for routines and predictability and broad sensory processing difficulties overlapped, (papers 2&4) and this is important for all individuals working with maltreated children to be aware of. However, the preliminary difference that was apparent between the children in the case study series was that visual fascination with item movement, touching textures and behaviours that were more repetitive in nature were more common in the children with Autism than the children with DSED. While it is yet unknown if these differences are replicable in other samples, there are pockets of evidence in the literature which also report greater prevalence of repetitive behaviours in Autism compared to DSED^{RAD} in modest community samples of clinic referred children (Moran 2010; Sadiq et al., 2012; Mayes et al., 2017). In addition, Mayes et al, (2017) found in a small sample of clinically referred children with DSED^{RAD} (n=7) and DSED^{RAD} + Autism (n=13) compared to a larger sample of clinically referred children with Autism (n= 486) that sensory difficulties related to eating were apparent *only* in the Autism groups (children with Autism and no maltreatment and children with DSED^{RAD} + Autism). Interestingly, within the case study sample of this thesis, the sensory difficulty of 'gagging at textures/utensils in mouth' (more than peers of a similar age) was also found to be present only in the Autism group and *not* in the DSED group (including children within the DSED group who also had ADHD). This finding is also in line with

the literature regarding food selectivity and eating problems in Autism, with a previous systematic review reporting that eating difficulties tend to be related to sensory sensitivity to textures (Cermak et al, 2010). Furthermore, it also fits with recent findings from a large-scale prospective study regarding Avoidant/Restrictive Food Intake Disorder (ARFID) in a non-clinical sample (n=3728) (Dinkler et al., 2022). Neurodevelopmental problems, including Autism, were assessed over time and symptoms of ARFID were assessed between 4-7 years old. Dinkler et al, found that the odds of having suspected ARFID were 3 times higher for children with neurodevelopmental problems.

Key message for clinicians and researchers:

- Children with DSED may present with language difficulties, particularly expressive and pragmatic language difficulties.

- However, *specific* pragmatic language and social communication skills might help differentiate children with DSED from children with Autism and is a key area for future research.

- Need for routines are apparent for both groups of children, but some sensory behaviours including visual fascination with movement, behaviours that are repetitive in nature, interest in touching textures or in avoidance of textures in mouth have been found, in small samples, to be more common in children with Autism compared to children with DSED.

- Further research regarding both the language and sensory processing difficulties of children with DSED is required to replicate and test reliability of the findings.

Key messages for clinicians

- SLTs have a key role to play in both supporting children with DSED and in differential diagnosis between Autism and DSED.

- For children with Autism or children with DSED who have sensory processing problems impacting daily participation, referral to Occupational Therapy for a sensory assessment/supportive intervention should be considered.

7.4 Overall message 2:

We have the tools and clinical expertise available to make accurate differential diagnosis between Autism and DSED, but unstructured observation may better support discrimination in complex cases.

Two key findings evidence this overall message:

Key finding 2a: Current 'gold standard' observational assessment (ADOS-2) discriminates Autism from DSED, but only in fairly straightforward cases. **Paper 2** demonstrated that the unstructured Live assessment better supported differential diagnosis in complex cases and it is the latter that are more likely to present difficulties for clinicians.

Key finding 2b: SLTs have a key role to play in both supporting children with DSED *and* in differential diagnosis of Autism from DSED: **paper 2** demonstrated that within the unstructured observation, differences in nonverbal communication and reciprocal interaction were identified. **Paper 3** identified areas of language difficulties for children with DSED and expanded on the findings of **paper 2**, demonstrating that specific pragmatic language differences were found only via SLT analysis of the conversational elements of the Live assessment.

7.4.1. Key finding 2a: Current 'gold standard' observational assessment (ADOS-2) discriminates Autism from DSED, but only in straightforward cases.

Although papers 2 and 3 both demonstrated that caregiver report does not support differential diagnosis of Autism from DSED, paper 2 showed that in straightforward cases of DSED, the ADOS-2 can differentiate Autism from DSED. To a degree, this is reassuring, because in local health boards, clinicians typically assess for Autism via informal interview with caregivers, school questionnaire and administration of the ADOS-2. For clear-cut cases of Autism, clinicians are already on track and for clear-cut cases of 'not Autism but possible DSED,' adding the RADA, Teacher RPQ and WRO to the assessment following an ADOS-2, could be a relatively simple but supportive procedure (as paper 2). The difficulty is that expertise in assessing for

DSED, including use of tools like the RADA, RPQ and WRO, still tends to sit with small groups of clinicians and researchers who have specialist interest in the field; knowledge and skills are not yet widely embodied across child services &/or those researching maltreatment associated difficulties in mental health and communication research. Dissemination of findings from this thesis will be another positive step to raising awareness and promoting the needs of these children, but further research with larger samples will help to provide the much-needed evidence to contribute to service design and training that could make real world impact for accurate differential diagnoses of Autism and DSED.

Additional difficulties with current Autism assessment approaches (caregiver report, ADOS-2, school questionnaire) arise with additional complexity- subtle Autism symptoms, overlapping neurodevelopmental conditions (as paper 2), or incomplete/unclear case history- (Davidson et al., 2015; Davidson et al., 2023). Paper 2 demonstrated false positive outcomes on the ADOS-2 when children with DSED presented with moderate to severe co-occurring ADHD or co-occurring Autism. While the ADOS is recognised as a valid tool for identification of Autism from no Autism diagnosis, there is paucity of research regarding its ability to discriminate between Autism and other neurodevelopmental conditions (De Giacomo, et al., 2021). One study found that neither the Autism -G (original version) or the ADOS-2, discriminated ADHD and other neurodevelopmental conditions from Autism, (De Giacomo, et al., 2021), but in that sample the Autism group were children with sub-threshold Autistic symptoms therefore Autistic symptoms were likely to be less prominent. Additional data is required regarding the use of ADOS-2 to discriminate from other neurodevelopmental conditions. Given that CAMHS is a service for supporting children with severe mental health problems, and that unrecognised neurodevelopmental conditions, like Autism or ADHD, may be more likely to referred to CAMHS for internalising and externalising behaviours, rather than core problems of the disorder itself (Byrne, 2003; Kantzer et al., 2013), it is likely that clinicians will be left scratching their heads, wondering about Autism or DSED (or both) for these kinds of cases unless a different approach to assessment is undertaken. Neurodevelopmental overlap is not uncommon in Autism (Gillberg, 2010; Gillberg and Fernell, 2014) or DSED (Minnis, 2013; Dinkler et al., 2017) and is even more common if any child has experienced maltreatment (Dinkler, et al., 2017). This means that every maltreated child presenting in clinic with mental health and/or behavioural difficulties requires a holistic assessment, rather than an assessment which focuses solely on trauma symptoms or maltreatment/trauma related disorders (Hiller et al., 2023). However, clinicians can take heart from the knowledge that it may be easier to discriminate between Autism and DSED

in complex cases, via *unstructured* behavioural observation, like the Live assessment, where skilled assessors are manipulating aspects of the social interaction to provide opportunities, prompts and probes (papers 2 and 3).

Wing, Gould and Gillberg, (2011) argued that social imagination, the ability to intuitively know how to react or respond in a social setting, predict one's role and predict what may happen next is a core feature of Autism and the finding that the unstructured Live better supports identification of Autism, especially in complex cases, perhaps speaks to Wing et al's argument. The unstructured nature of the Live and the back-and-forth- dynamic of the 2 assessors increased the unpredictability in the social aspect of the assessment. Furthermore, the 2 assessors interacting individually and together with the child, replicates a 'group' dynamic, and this in addition to use of social presses/social stressors, created greater social challenge. Essentially, the children were pushed to use their social skills, where present, to manage the dynamic and therefore these skills stood out and, in contrast, core Autistic symptoms were exacerbated. Furthermore, core disinhibited behaviours were elicited in the DSED children during the social stress elements, and the free-flowing unstructured nature enabled easier observation of ADHD symptoms. By observing the behavioural patterns, as suggested by Moran (2010), in an unstructured setting it was possible to identify the different quality of the interaction and communication between the groups.

For clinicians who are used to using a guided tool like the ADOS-2, moving to an unstructured observation, particularly for their most difficult cases, may feel uncertain. Yet, the Live assessment is already a clinical tool, not a research tool, which has been used by the Scottish Centre for Autism, (SCA) in the NHS for over 30 years. It can be administered in a similar, or shorter, time frame to the ADSOS- 2 (approx, 30-45 mins) and with a similar number of clinicians as is currently used clinically to administer ADOS (2 clinicians). This means that training for clinicians who wish to enhance their skills would be an option. It is also important to remember that experienced clinicians in Autism already have the skills and knowledge to utilise the Live assessment or develop their own locally relevant version. Within the Live, clinicians are applying their Autism knowledge and observing child responses from the same perspective as an ADOS-2 assessment. The change is simply moving from a structured piece of equipment with set tasks to a free-flowing dynamic, where the clinicians have in their 'mental toolkit' tasks they may consider using, but will chose how and when to use them, or to try something different, depending on the response of the child.

Message for researchers and clinicians:

- Unstructured observation that increases unpredictability and social challenge may make it easier to unpick the nature of the behaviours that are different between Autism and DSED, especially in complex cases.

- Future research may focus on feasibility trials of assessments, like the Live, with larger samples, in comparison to 'care as usual' for the assessment of complex cases to establish whether there is an appropriate evidence base to inform future change in service delivery.

Message for clinicians:

- The Live assessment is readily applicable to clinical settings and while training may be required, many experienced Autism clinicians may already have the skills to utilise such an assessment.

7.4.2. Key finding 2b: SLTs have a key role to play in differential diagnosis of Autism from DSED

The combined findings of papers 2 and 3 demonstrate that SLTs have a role both in supporting children with DSED and in differential diagnosis of Autism from DSED. For example, Sadiq et al, (2012) found significant overlap between the broad pragmatic language difficulties of children with Autism and children with DSED via caregiver report and these findings were replicated in paper 3. Yet specific differences in pragmatic language were still identified, in this case study sample, when independent SLTs, blinded to the diagnosis, analysed conversational elements from the Live assessment. In addition, when SLTs structurally analysed the morphology and syntax of the conversations, it became clear that children with Autism rarely or never used -wh questions, which was in stark contrast to children with DSED who used them appropriately (paper 3). These findings were not only in keeping with the previous literature (Bacon et al., 2018; Sukenik et al., 2021) but were only identified because the skills of SLTs enabled detailed investigation. While all professionals with experience of assessing Autism can be trained to administer diagnostic tools such as the DISCO, or screening tools like the CCC-2, papers 2 and 3 suggest that in cases of complexity such measures are perhaps too broad in scope to unpick the specific differences, and/or the quality of the differences are better identified by professionals such as SLTs, who are trained to investigate within the complex layers of communication. It may not be the

case that every child being investigated for Autism, or DSED, will require SLT input, but papers 2 and 3 suggest that an SLT lead in the observational assessment of cases which are difficult to unpick and/or where there are concerns about unclear communication, delayed language skills or where detailed analysis of pragmatic language skills is required, may be beneficial.

Nevertheless, recent reports suggest that some groups of children, like those who have experienced early maltreatment, are still underrepresented by SLT, which in part, may be because communication problems might be thought of as secondary by caregivers or professionals when presented with difficult behaviours and/or mental health problems (Cummings, 2021; McCool, 2024). This is despite substantial evidence which suggests that maltreated children are at higher risk of communication difficulties (Carr et al., 2020; Sylvestre et al., 2016). Paper 3 suggests that children with DSED can be considered within this group of underrepresented children and SLTs may need to advocate for their role in supporting such groups of children.

The other possibility is that SLTs themselves may not be aware of the important role which they can play in differential diagnosis of complex cases, like Autism from DSED. In my clinical and SLT training experience, dilemmas that are diagnostic in nature, and/or associated with parent-child relationship problems tend to be considered the role of psychiatry and perhaps psychology, sometimes with more emphasis being placed on trauma and attachment than the relative role of communication needs (McCool, 2024).

Taking together the key findings from message 2, one recommendation is development of a training package to further the awareness, knowledge and skills of clinicians who may be faced with assessment of complex cases. This training could be undertaken by CD-J in collaboration with the SCA, and other relevant experienced multi-disciplinary clinicians. Training may cover the following:

- clinician confidence in following their clinical intuition, as opposed to a manual.
- Clinical practice of using a 2-assessor dynamic to bounce off each other and push the child to see how far they can go, while maintaining sensitivity.
- Awareness regarding DSED generally, and regarding differential diagnosis from Autism.
- The role of SLT as a possible lead professional in a Live assessment, and how they can use their skills to home in on pragmatic language differences, as well as assessment of other areas of language.

Message for researchers and clinicians:

- While caregiver report has its place in assessment, especially regarding developmental histories, broad standardised caregiver reports can be too broad in scope to identify skill differences in social communication/pragmatic language.
- SLTS have the skills to analyse conversational samples in detail, relative to the language domain being assessed (expressive language, pragmatic language etc).
- All professionals working with maltreated children/children with DSED should be aware of the high risk of communication difficulties and include SLTs in joint assessments.
- Training and awareness raising among SLTs may be required regarding their role in supporting children with DSED and differential diagnosis between Autism and DSED – and in the identification of co-occurring DSED/Autism and other disorders.
- Additionally, the skills of SLTs would be beneficial in research, and researchers could consider how they can utilise collaborations in future communication research in this field.

7.5. Overall message 3: We need to further raise awareness that maltreated children may be high risk for DSED and support clinicians to think about identification of DSED in clinic. Additionally, we need more research to better support their needs.

Key finding 3a: Children with DSED have a specific set of difficulties compared to other maltreated children, but DSED is still not widely identified. **Papers 1-4** demonstrate that across multiple domains of child development, DSED is not widely recognised within the wider maltreatment literature; **papers 1, 3 and 4** were the first consider DSED within their respective areas.

Key finding 3b: Children with DSED may also present with additional neurodevelopmental conditions, yet little is known about the outcomes for these children. Across **all 4 papers** the question of relative impact of additional neurodevelopmental problems was raised, but these questions cannot yet be answered.

7.5.1. Key finding 3a+3b: Children with DSED have a specific set of difficulties compared to other maltreated children, and may present with additional neurodevelopmental conditions, but DSED is still not widely identified or understood.

It is clear from the findings across all 4 papers that DSED is still not widely considered by the majority of professionals working with children who have been maltreated, despite consistent evidence of greater social functioning difficulties, (Guyon-Harris et al., 2019; Guyon-Harris et al., 2019, Bennet et al., 2009; Kay and Green, 2012), possible overlap of symptoms with Autism, (Moran 2010; Sadiq et al., 2012 & Davidson et al., 2015), associations with ADHD (Pears et al., 2009; Bruce et al., 2010; Kennedy et al., 2017; Seim et al., 2021; Davidson et al., 2023) and/or co-occurring Autism (Mayes et al., 2017; Davidson et al., 2023). Papers 3 and 4 now add to the knowledgebase the risk of expressive language difficulties, some areas of pragmatic language and sensory processing problems. As many of these difficulties have been described in maltreated children (without DSED/DSED not investigated) (Cichetti et al., 2016) clinicians, and researchers, may wonder why it is important to assess for DSED, as opposed to treating presenting behaviours within a trauma/maltreatment framework, and the answer is multi-faceted. Firstly, it is crucial to remember that children with DSED, as opposed to maltreated children without DSED, have specific social relatedness difficulties - indiscriminate behaviours and poor social boundaries – that are core to the disorder (DSM-5) and these are in addition to more general social functioning and communication difficulties. Secondly, the lack of identification of DSED within the research means that, in comparison to other groups of children, like children with Autism, we still know very little about the impact of the core symptoms on child outcomes, or relative impact of additional relational, communication or sensory problems. Papers 2, 3 and 4 investigated interaction and communication skills and sensory processing in children with DSED compared to children with Autism, but better skills (in some domains) relative to Autism should not be taken to assume impairments are not present; the children with DSED were, in general, doing more poorly than the typically developing groups (as in papers 3 &4). Furthermore, in paper 1, some studies identified that children with DSED had greater peer problems than children with RAD, or the environmental controls (Seim et al., 2021), which raises the question of why, if maltreatment is the main precursor to these problems? Many questions of this type

cannot yet be answered but bringing together strands of neurobiological and genetic research with research specifically on DSED, and RAD, may be of future importance.

It is not well understood why some maltreated children develop DSED, some develop RAD and some develop neither, however some evidence suggests that pathways may differ depending on type of maltreatment (Ellis et al., 2022; Sheridan and McLaughlin, 2014). For example, severe neglect (deprivation) is associated with reduced cognitive functioning, including poorer executive functioning, language and learning skills (McLaughlin, 2017), and findings from the Bucharest Early Intervention Project, (BEIP) suggested that less white matter connectivity in areas such as the frontal lobes and the amygdala impacted higher cognitive functioning skills, as well as emotional development (Eluvathingal et al., 2006). Neither DSED nor RAD were specifically investigated within these studies but as mentioned previously, there is evidence to suggest that poor inhibitory control, an executive functioning skill more typically associated with ADHD, was associated with DSED symptoms in non-institutionalised samples of children with DSED (Pears et al., 2010) and children several years post-adoption from the Romanian institutions (Bruce et al., 2009). Additionally, a genetic study from the English-Romanian Adoptees Study (ERAS) suggested a further association with ADHD symptoms in some of the severely deprived children, (Stevens et al., 2009). A set of DNA variants along one chromosome, a dopamine transporter, which has been associated with ADHD in non-maltreated children, was found to moderate the relationship between symptoms of ADHD and severe deprivation in later childhood and adolescence (Stevens et al., 2009). This raises an interesting question - could the interplay of maltreatment with poor inhibitory control be one reason why some maltreated children are so severely socially disinhibited they warrant diagnosis of DSED? It seems plausible given symptoms of ADHD may be frequently co-occurring with DSED in both the post-institutionalised Romanian adoptees (Zeanah et al., 2009; Kennedy et al., 2017) and non-institutionalised samples (Kocovska et al., 2012; Seim et al., 2022; Davidson et al., 2023) and that, in the BEIP, ADHD symptoms were found to persist despite placement in nurturing foster care (Zeanah et al., 2009). Future research would benefit from replication of the ERAS investigation but specifically with samples of children with DSED. One study has investigated the possible role of genetics in DSED specifically. Minnis et al, (2007) reported in a general population twin study (n= 6771 twin pairs), high correlations between Monozygotic twins, (MZ) (genetically

identical/shared environment), and lower correlations for Dizygotic twins, (DZ) (share 50% of genes, like other siblings/shared environment) on the total score of the Relationships Problems Questionnaire (RPQ), (screening for RAD and DSED), and on the sub-scale for DSED symptoms. The difference between MZ and DZ twins on the total RPQ score was highly significant for males only, but significant differences for both males and females were found regarding the DSED subscale. Some caution in interpretation is warranted however as the sample were children who had experienced harsh or negative parenting, which is likely to be less severe than the maltreatment experiences associated with DSED. The findings tentatively suggest that shared genes had a stronger influence on DSED symptoms than shared environment.

The ERAS and BEIP reports tend to suggest the environment, (severe deprivation) as the 'cause' of the modification of the genes impacting the range of difficulties, including DSED and ADHD symptoms (Bos et al., 2011). However, this view could be challenged. It is possible that some of these children were already genetically susceptible to development of neurodevelopmental conditions (Minnis et al., 2007; Dinkler et al., 2017; Minnis et al., 2020), especially as ADHD, like Autism, has also been shown to be highly heritable (Larsson et al., 2014). A recent rigorous review of systematic reviews and meta-analysis, which formed the update to the International Consensus Statement on ADHD, reported that "*ADHD is rarely caused by a single genetic or environmental risk factor, but most cases of ADHD are caused by the combined effects of many genetic and environmental risks each having a very small effect* (Farone et al., 2021).

Taking together the findings from research into DSED, research into types of maltreatment and associated outcome for neurodevelopment and relevant genetic research, it seems that epigenetic research, the study of how one's environment can lead to changes in the way genes are expressed (Centre for Disease Control and Prevention, 2022) could be a crucial next step in the understanding DSED, especially as there is already some research investigating epigenetics in the context of maltreatment, (Lang et al., 2020). A formative understanding of the mechanisms of the disorder could lead to targeted intervention research and improved case management.

Another area for future research regards the interplay of co-occurring neurodevelopmental conditions with DSED and relative impact on social functioning. For example, the possibility of overlapping Autism in community clinically referred

samples of children with DSED/DSED^{RAD} has been demonstrated. Kocovska et al., (2012) found that 70% of their sample of children had possible/likely Autism (n=17) and Mayes et al., (2017) reported co-occurring Autism in 65% of their sample (n = 13). These findings were replicated in two children within this case study sample, as described in paper 2. The DSM-5 DSED diagnostic criteria recommends that Autism be excluded before diagnosing DSED, but this may create arbitrary classifications, especially within research studies, as there appears to be no aetiological reason why DSED+Autism cannot co-exist (Minnis et al., 2020). Furthermore, it may create barriers to appropriate treatment since overlap in neurodevelopmental conditions is the norm, not the exception (Gillberg, 2010; Minnis, 2013; Minnis et al., 2020), and treatment approaches to DSED and Autism may differ. For example, two small scale community studies have suggested that children with DSED^{RAD} may better respond to interventions which are psycho-social and dyadic in nature compared to children with Autism, (Mukkades et al., 2004; Becker-Weidman et al., 2006). We have already discussed the possible co-occurrence of ADHD with DSED, but as evidence suggests that it may be the interplay of ADHD with DSED which has negative functional outcomes, rather than DSED by itself, (Kennedy et al., 2017), future understanding, identification and support regarding ADHD is also imperative.

McCrory et al, (2022) proposed that for maltreated children both *stress generation* (susceptibility to cumulative stress due to maltreatment-associated problems) and *social thinning (*diminished social networks due to poorer social competence and missed opportunities) could have a negative effect on later mental health, and Gajwani and Minnis, (2023) argued that children with DSED, or RAD, may experience 'double jeopardy' regarding mental health outcomes due to interplay of co-occurring neurodevelopmental conditions. Across all 3 clinically related papers of this thesis, the children with DSED+ADHD were those who were hardest to assess (paper 2+3), their difficulties were hardest to identify (paper 2) and they had the greatest language and communication difficulties (paper 3). Furthermore, in paper 4, *all* the children with DSED showed increased sensory seeking behaviour on the CSP-2 but some of these behaviours included inability to sit still, always running or moving, and fidgetiness, which could be considered symptoms of ADHD (DSM-5). Thus, in cases of childhood maltreatment, *both* DSED *and* possible overlapping neurodevelopmental conditions

must be considered alongside impaired social function and communication to provide a fuller picture for health and social care management (Hiller et al., 2023).

This leads to perhaps the most important reason for raising awareness of DSED and supporting clinicians to consider assessment for DSED and additional neurodevelopmental conditions. It is through our knowledge of Autism that clinicians are able to provide environmental and communication strategies to support families, which can help reduce stress and improve quality of life (Davis & Carter, 2014; SIGN, 2016). There have been 599 times more studies of Autism compared to DSED since the advent of DSM-5, (cf. figure 1, introduction, p27) which demonstrates how little we know about DSED, in comparison. Qualitative studies of caregivers of children with DSED/DSED^{RAD} have voiced concerns about lack of support, a need to be the child's advocate and not being listened to (Turner et al., 2023; Schlein et al., 2023; Vasquez et al., 2015). However, there is a distinct lack of research giving a voice to the young person. To date, there has been only one qualitative research study conducted with children with DSED (community sample, n= 8) which used Interpretative Phenomenological Analysis to understand peer relationship difficulties from the child's perspective (Bennet et al., 2009). Children with DSED are expected to form new relationships in foster placements, with adoptive families and with peers, which in some cases may have involved change of locality or school, and this is often while trying to maintain some contact with their biological families. It is possible that unmet relational or communication needs could negatively impact ability to engage, in addition to any trauma related factors (McCool, 2024), but a future goal must be to use rigorous qualitative research methods to understand the social relationship needs, strengths and challenges faced by children with DSED by asking the children themselves.

The Live assessment, in paper 2, is somewhat related to the above as one of the benefits of utilising an unstructured conversational approach was the flexibility to ask the child about their experiences, as required, and when in different emotional states. The children with DSED, for example, often talked about activities with peers when calm during back-and-forth conversation, but when aroused or stressed their descriptions became vague and ambiguous. The children with Autism tended to have similar levels of description regarding their friendships, regardless of their arousal. The other benefit was ability to probe for challenges within peer relationships. For example, conflicts in

relationships were not uncommon in both children with Autism and DSED, and especially for those with neurodevelopmental overlap. Other socially related experiences such as empathy and basic theory of mind were targeted during conversation in the Live assessment, with the former being impaired in both groups but difficulties with the latter more common in the Autism group. Sensory differences were also asked about directly in the Live assessment and the reports by the children often supported those of their caregivers. For example, some children with DSED and some children with Autism both talked about dislike of specific noises and were able to give examples of times when they felt distressed by these (school bell), whereas other children with Autism talked about specific food tastes being unbearable- a finding discussed earlier that often arises in Autism (Mayes et al., 2017; Dinkler et al., 2022). Caregiver report and expert clinical observation are core elements of good neurodevelopmental assessment, but alternative approaches such as the Live assessment demonstrates that beneficial information can be garnered from asking the young person about their experience (or lack of) directly and arguably adds to a more complete holistic understanding during clinical assessment.

There is a paucity of treatment research regarding DSED and RAD but most treatment recommendations and strategies tend to focus on attachment patterns (Zeanah et al., 2016), parenting and psycho-educational approaches (Ziberstein, 2023; Mukkades et al., 2004; Becker-Weiderman, 2006) and/or trauma focused attachment approaches such as Dyadic Developmental Psychotherapy (DDP) (Turner-Halliday, 2014; Hughes, 2017; Wingfield et al., 2018) rarely taking into account co-morbid difficulties (Ziberstein, 2023). Yet, all of the evidence discussed thus far appears to point to the need for future research to take a holistic approach to the treatment of DSED. This will be better achieved with increased understanding of the likely-intertwined underlying mechanisms of genetics, neurobiology and neurodevelopment and the role of environmental factors in how these develop, but perhaps there is enough evidence to advocate that in addition to attachment and trauma, case management could consider the need for, and offer if required, supportive behavioural strategies regarding impulse control and hyperactivity and if severity warranted, treatment for ADHD. Communication passports, which are designed to provide a brief but informative snapshot of an individual's preferences, needs, how they communicate and how to best communicate with them may be useful (McCool, 2024) for every maltreated child in care; especially where children are moving across multiple placements and may find it challenging to communicate their needs.

Following assessment, decisions regarding further language and communication intervention would be considered. In Autism, social skills support or social skills training has been one widely discussed and systematically researched management approach (Radley et al., 2020; Reichow et al., 2013) and another is use of social stories, (Karkhaneh et al., 2010), however future research may determine whether a form of social skills support may be beneficial for children with DSED in navigating the social difficulties as per paper 1. Finally, embracing an ESSENCE framework (Gillberg, 2013) in assessment may support a holistic approach to treatment. Essentially, this means moving away from a diagnosis specific pathway of assessment and approaching the assessment jointly with relevant professionals involved together at the same time. ESSENCE is a move towards a broad perspective, identifying *all* key symptoms as they arise and expecting that they may eventually form multiple diagnoses. The free-flowing dynamic of the Live assessment lends itself to an ESSENCE framework, and as shown in paper 2, may better support different underlying behaviours to come to the fore to be observed as they emerge. The purpose is not to give the child lots of labels but to identify core difficulties within severity ranges so that each can be treated accordingly and as equally as possible. For example, if DSED and Autism and ADHD are identified along with expressive language difficulties, there may not be clear evidenced treatments for DSED yet, but treating the ADHD, which has one of the most well established evidence bases for treatment (Faraone et al., 2021) and imputing Autism friendly environmental and behavioural supports as well support for communication needs may go a long way to supporting the child's needs, as opposed to taking only a trauma related approach.

<u>Message for researchers and clinicians:</u>

- Be open to learning about DSED and how to assess and support young people with DSED and consider assessment/reaching out to local expertise, if required, to consider DSED when working with children with a maltreatment history. In addition, always consider co-morbidity.
- Differentiating between Autism and DSED does not mean that DSED+Autism cannot co-occur, and further research is required to better understand this presentation and to provide management support.

- The purpose of differentiation between Autism and DSED is to ensure appropriate treatment options, and that Autism is not wrongly mistaken for a maltreatment-associated disorder.
- An ESSENCE approach may be a supportive framework for assessment of complex cases, with the purpose of identifying each disorder to provide appropriate and timely treatment.

7.6. Strengths and Limitations of the thesis as a whole

The thesis contributes to the knowledge by a thorough exploration of various symptoms which may overlap between Autism and DSED and findings together suggest that there may be specific skills and behaviours which differ in Autism, compared to DSED and that these now warrant investigation with larger more rigorously tested samples. The Autism diagnostic criteria and clinical assessment guidelines were used as springboard from which to conduct investigation using an in-depth case study design, which was a relative strength as there was a scarcity of research into DSED generally. There were also specific gaps regarding areas impaired in Autism, (social functioning (paper 1) language ability (paper 3) sensory processing (paper 4)) and to the best of our knowledge, papers 3 and 4 were the first studies to investigate receptive and expressive language skills and sensory processing in DSED, and the first to examine any overlaps and differences to Autism. There was also scarcity of research which investigated differential diagnosis of Autism from DSED using multi-informant measures, and paper 2, which is published, was the first to use both gold standard Autism assessment tools and an unstructured observation. The second strength of this thesis is the clinical relevancy of the findings. Clinicians have been striving for a better understanding of DSED, itself, and how to discriminate from Autism and because of this thesis, additional behaviours and assessment methods can now be identified for future investigation which could have direct impact on clinical practice, supporting clinicians in assessing tricky cases. Furthermore, in each of the case sample studies, (papers 2-4), we used assessment tools which are already clinically available (DISCO, CCC-2, CSP-2), and in some cases, already widely used (ADOS-2) or practices which can be undertaken with minimal training (LIVE assessment) or by already skilled clinicians (SLT analysis of language skills from conversation samples). This means that findings can be easily translated into clinical knowledge. It also means that there are already skilled clinicians within health services capable of conducting the assessments utilised, albeit with some additional training (RADA, WRO, LIVE, pragmatic analysis), if required. Finally, this thesis adds

to the knowledge of DSED which is important because this group of children remain grossly under-researched in comparison to other groups of children with neurodevelopmental conditions. Enhanced knowledge regarding the language, social communication, social functioning and sensory processing of children with DSED goes some way to identifying the professionals best skilled to support their needs.

As with any research project, there are also imitations to this thesis. The findings are based on a case study series with small sample of children, but the smaller numbers did enable in depth analysis and determined which tools and/or behavioural differences may be best tested again with a larger sample to assess replication and generalisability. In addition, some samples bias may be present in both the Autism and DSED groups as the majority of children were self-referred by caregivers. It is possible that these are families who are most motivated and therefore less representative of a wider population, or they were families actively seeking help and therefore represented a group more likely to have difficulties. Alternatively, it could be argued that not all cases were reflective of the complexities seen in CAMHS. Despite possible sample bias, it was still possible to detect differences between the groups. The inclusion of children with co-occurring ADHD in both the Autism and DSED groups was perhaps a confounding variable, as ADHD itself is associated with poorer social functioning and some difficulties with social communication. However, the sample also reflects the complexities that clinicians are likely to face and the inclusion actually benefitted the findings as it demonstrated the differing outcomes for 'straightforward' versus 'complex' cases. Further questions about the inter-play of conditions like ADHD with DESD were then raised, which are important for further research. A final limitation was the reliance on foster carer/adoptive parent report in the DSED group regarding length of time in the maltreatment environment/age at removal. Gaps in knowledge meant that we were unable stratify the DSED group by length of time in the maltreatment environment. While ethics permission had been sought to access social work records to obtain objective and measurable information, unavoidable circumstances prevented collection of this data. Again, this reflects the difficulties faced in clinical practice, but does require further investigation in future studies.

While reflecting on the limitations of the thesis, it seems pertinent to also reflect on the challenges raised by governance barriers during the process of obtaining ethical

approval. One of the toughest challenges in running a multi-faceted research study, of any type, is management of time as each phase, (ethics, recruitment, data collection, analysis and write-up/dissemination) has to be conducted efficiently and time sensitively in order to meet the expected outcomes within the period of allocated funds. Yet significant delays, of at least 6 months, occurred for this doctoral work at the outset due to governance factors that are not yet avoidable, but could be improved upon. There were several small challenges but the one that created the significant time barrier was the lack of understanding of the ethics committee about the complex nature of DSED diagnosis. As substantiated maltreatment is inherent within the diagnoses, it may not be in every family's best interest to label it due to the possible association of blame. In the case of *some* birth families, who perhaps have worked hard to rehabilitate and have had their child returned, describing the associated behaviours of DSED as a behavioural formulation rather than a diagnostic label may be more beneficial to *that* family whereas for other families, including often foster/adoptive families, the diagnosis may be very helpful in understanding the child's presentation. The other reason for being cautious with DSED diagnosis is because elements of maltreatment cannot always be clearly known. It may be strongly suspected and there may be good analogous evidence but unless substantiated evidence can be gathered it is generally better to describe the symptoms of DSED in light of the environmental circumstances known than give the diagnosis of DSED. All the cases within the included case study sample had known histories of maltreatment but, particularly with clinically referred samples, at the planning stage we could not be sure that this would be the case. The proposal to ethics to treat the labelling of DSED on a case by case basis in tandem with the referring clinician was a clinically reasoned argument with the best interests of the family/young person in mind. However, the ethics committee rejected this proposition more than once, regarding it as withholding information. Labelling on a case by case basis is clinically justified with other diagnoses that have inherent causal factors such as Foetal Alcohol Spectrum Disorder for the same reasons; blame may be attributed and sometimes it is not possible to be certain, yet the lack of knowledge/research regarding DSED seemed to negatively bias against this clinically justified argument. The six-month delay, during which several iterations of explanations to the ethics committee were required to resolve the issue, led to a late start and therefore reduction in recruitment time. This was significant because recruitment from clinical services is already challenging due to clinician time and capacity.

Despite the challenges faced, the back-and-forth discussions had with the ethics committee were insightful and highlighted the need for a broader range of child mental health minded individuals to be involved in these panels and even more so highlighted the need for insight from those involved in the research, i.e. caregivers and families of fostered and adopted children (and with DSED) to be brought to the fore. This could mean involving experts by experience in the ethics panels as they would bring an alternative experience, with real-world application, to the discussion and may better support a more rounded approach to governance. It is vital that governance does not create more barriers than opportunities to research with complex groups. Arguably, groups like children with DSED are some of the most vulnerable yet under-researched populations and therefore most in need of the outcomes that good clinical research can provide.

7.7. Conclusions

Bearing in mind that small samples were included which may involve some sampling bias, and that replication of results with larger samples are required before findings can be generalised, the in-depth examination of the case sample within this thesis suggests that in some areas there are substantial overlap between Autism and DSED, but it is also possible to discriminate between the two diagnostically distinct disorders. Furthermore, we have both assessment tools and clinical expertise available. Social relationship problems may broadly overlap between Autism and DSED *but* specific differences may lie within the social communication domain. However, to better identify these important differences, a change of approach to use of unstructured observational and conversational assessment is recommended; by enhancing the social challenge, the pragmatic language difficulties and broader social communication difficulties of children with Autism are exacerbated, the skills of children with DSED, compared to Autism, are highlighted *and* impact of any overlapping neurodevelopmental conditions may be better identified. The latter appears to play a crucial role in adding complexity to cases.

The implications of this doctorate research for clinicians are hopefully reassuring and help build confidence in what is not a straightforward area. Training for clinicians may be required, and in advocating the key role of SLTs in the assessment. Many questions were also raised which cannot yet be answered and further research regarding DSED, as a whole, and the interplay of co-occurring disorders with DSED is needed, in addition to replication of the findings with larger samples, so that we can better understand the

needs of individuals and provide appropriate supports and treatments to those who need it most.

7.8. References

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