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Reactive Attachment Disorder in Infants in Foster Care and associated Mental Health and Cognitive Functioning

AND CLINICAL RESEARCH PORTFOLIO

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Submitted in partial fulfilment of the requirements for the
degree of Doctorate in Clinical Psychology (DClinPsy)

September 2016



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(c) Molly Bruce [Sept 2016]

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Reactive Attachment Disorder: A Systematic Review

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Submitted in part fulfillment for the Doctorate of Clinical Psychology
(DClinPsy)

Supervised by: Prof. Helen Minnis and Dr Susan Turnbull



Prepared in accordance with guidelines for submission to Infant Mental Health Journal
(see appendix 1.1)

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Abstract

Background: Reactive attachment disorder (RAD) is found in maltreated children and is characterised by a failure to demonstrate important attachment behaviours such as signalling to a caregiver or accepting comfort, when experiencing distress. According to the DSM-V criteria, children with RAD are likely to be socially and emotionally withdrawn, and show limited positive affect. The aim of this review was to explore factors associated with RAD, giving consideration to the clinical profile and prevalence of the disorder. **Method:** A comprehensive search consisting of electronic database and citation searches identified 14 articles, published between 2004 and 2015. The articles were appraised using an adaption of the NICE (2012) Quality appraisal checklist for quantitative studies reporting correlations and associations. **Results:** Overall the quality of studies was moderate to good. The review showed that RAD is associated with various situational, psychological, and behavioural factors. RAD was found to be a somewhat rare but nonetheless established disorder. Limitations of included a lack of observational measures to inform a diagnosis of Reactive Attachment Disorder. **Conclusions:** Various factors associated with RAD have been highlighted however further research is needed. RAD seems responsive to intervention in the form of improved care giving environments. Implications of the results are discussed and suggestions for future research are made.

Keywords: *Reactive Attachment Disorder, Inhibited Attachment, Child Maltreatment, Institutionalised*

Attachment is a fundamental instinct that infants are born with, enabling them to form a close relationship with their carer and therefore survive in their environment (Bowlby, 1969). Infants with reactive attachment disorder (RAD) do not appear to demonstrate key attachment behaviours such as seeking or signalling to their caregiver when experiencing distress: There appears to be a deactivation of the attachment system (Prior & Glaser, 2006). Children with RAD may be socially and emotionally withdrawn in a wide range of situations. This is likely to limit children's ability to make use of love or care from others and reduce opportunities for learning. Therefore, it is probable that RAD has a considerable impact on child development (Prior & Glaser, 2006). It is widely accepted that this disorder exclusively occurs in the context of maltreatment, where the infant's attachment needs have been consistently neglected by their primary carer(s) from a young age. It has been argued that RAD is most prevalent in infants raised in institutional settings (Corval, Baptista, Fachada, Beiramar & Soares, 2014).

Reactive attachment disorder has been described as one of the least researched and most poorly understood disorders listed in the DSM (Chaffin et al., 2006). In the last decade there has been much debate around the disorder. There were previously two forms of the disorder as defined by the DSM-IV and DSM-IV-TR (APA, 1994; 2000), these being 'inhibited reactive attachment disorder (I-RAD)' and 'disinhibited reactive attachment disorder (D-RAD)'. Some researchers have argued that these two forms of RAD involve similar symptoms and can be co-occurring (e.g. Giltaij, Sterkenburg & Schuengel, 2013), whereas other research has demonstrated that the two forms have discrete symptomology, phenotypic characteristics and response to intervention (see literature review by Zeanah and Gleason, 2015). The ICD-10 divided the subtypes into two distinct disorders and recently the DSM-5 (APA, 2013) followed suit and updated the classification of I-RAD to simply 'Reactive Attachment Disorder' and changed the classification of disinhibited reactive attachment disorder to 'Disinhibited Social Engagement Disorder' (DSED). This change reflects that DSED is no longer considered to be a disorder of attachment (Zeanah & Gleason, 2015).

On the whole, researchers are fairly confident about the prevalence of DSED (e.g. Gleason et al., 2011). Much less is known about the prevalence of RAD although it appears to be a rarer disorder (Corval, Baptista, Fachada, Beiramar & Soares, 2014; Gleason et al., 2011). The DSM-V (APA, 2013) reports that less than 10% of children who have been severely neglected develop RAD.

Much of the research involving Attachment Disorders has explored the two disorders collectively rather than individually. The new diagnostic criteria emphasises the very different clinical presentations of the two disorders and the change in terms supports the notion that individual exploration of the two disorders would be more meaningful. Given that the previously termed RAD Disinhibited type is considerably more prevalent than the Inhibited form, failing to explore the two forms independently has often meant

that findings are more applicable to the disinhibited form of the disorder. This has left little being known about the inhibited form.

In summary, there is little research investigating RAD independently and factors reported to be associated with the disorder seem to vary widely. There are no known existing systematic reviews focussing on RAD. In order to develop effective interventions and prevent the development of the disorder, it is crucial that we learn more about the aetiology and course of RAD. We can begin to inform a fuller understanding of RAD by considering its correlates, as reported in the research literature to date. As well as collating findings on factors associated with RAD, this paper aimed to review the quality of such research and highlight areas that would benefit from further exploration.

Aims of Review

This systematic review aimed to provide an updated investigation into the prevalence of and factors associated with RAD. Findings pertaining to the stability of symptoms of the disorder are reported. The quality of the research was investigated including consideration of any potential bias.

Review Questions

- What factors are associated with RAD?
- What is the prevalence and stability of symptoms of RAD?

Search Methodology

An electronic search of the following databases was conducted: MEDLINE, Psychinfo (and PsychARTICLES), Web of Science, and British Library Ethos. Searches were limited to articles published in English and after 2004. Search terms were developed in consultation with the NHS Highland librarian. The search terms “reactive attachment disorder”, “attachment disorder” and “inhibited attachment disorder” were used to allow for a thorough review given the apparent rarity of the disorder.

A search on Google Scholar using the term “reactive attachment disorder” yielded one further paper for inclusion. Reference lists of the included studies were hand-searched and an additional paper was selected for inclusion. A research conference on Attachment Disorders in Europe, held in Glasgow (April 2016) highlighted a further unpublished paper for inclusion. Figure 1 illustrates the search process; see appendices 1.2, 1.3, and 1.4 for tables showing more detailed descriptions of the search process. When

it was unclear if inclusion criteria were met, papers were discussed with the academic supervisors of the project.

Inclusion Criteria

- Articles that were written in or have been translated in to English.
- Articles that were published between 2004 and 2016.
- Unpublished studies.
- Articles that directly explored factors associated with RAD, for instance factors relating to the development, presentation or comorbidities of RAD.

Exclusion Criteria

- Previous reviews, book chapters, and case studies.
- Qualitative research.

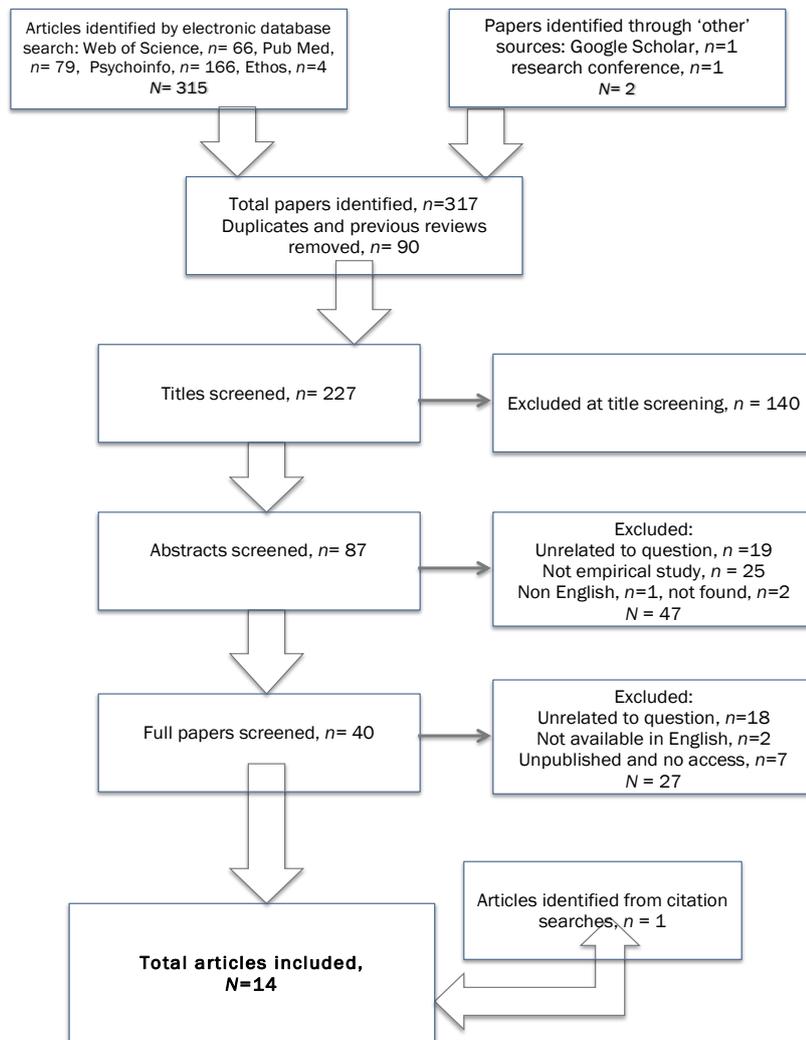


Fig. 1. Flow Diagram of Search Process

Data Extraction

Relevant data regarding RAD diagnosis, prevalence and associated factors were extracted. A meta-analysis of findings was not possible due to a lack of consistency in designs, measures and samples across studies; therefore a narrative synthesis approach was taken.

Quality Rating Criteria

The quality appraisal tool selected to establish level of bias was a checklist developed by NICE (2012) for assessing the internal and external validity of studies reporting correlations. The checklist is based on the appraisal step of the 'Graphical appraisal tool for epidemiological studies' (Jackson et al., 2006). It enables an appraisal of a study's internal and external validity by addressing the following key aspects of study design: characteristics of study participants; definition of independent variables; outcomes assessed and methods of analyses. The checklist can also be used to make comparisons across differing research designs, including randomised control trials and observational studies. The checklist was adapted for the current review (see appendices 1.5 and 1.6) to allow for numeric scoring that informs an overall quality presented as a percentage. Papers were considered poor quality if they scored below 50%, moderate quality if scored between 50-80% and high quality if scored above 80%.

To ascertain interrater reliability, an independent reviewer (Trainee Clinical Psychologist) rated a random sample of 50% of the articles. Agreement was high and any discrepancies between reviewers were discussed and resolved.

Results

Table 1 presents information on the quality ratings, design, sample characteristics, outcome measures, results, effect sizes and reported limitations of included studies. Findings in relation to prevalence and stability of RAD symptoms, factors investigated and factors found to be associated with RAD are reported. Fourteen articles were included in the final review with six being rated as high quality and eight as moderate quality (for scoring results see appendix 1.7). When reporting results, studies are referred to by their number in the table.

Design of Studies

A variety of study designs were utilised by the included studies: randomised control trials (4); a cross-sectional survey (n=1); a cross-sectional case control (n=1); retrospective cohort studies (n=3); cross sectional cohort studies (n=3); a retrospective twin cohort study (n=1); a longitudinal cohort study (n=1). The majority of papers (n=12) explored factors associated with RAD through a correlational design, the remainder (n=2) explored between group differences using t-tests.

Participants

Studies included in the review were carried out in various countries around the world including: USA (2), UK (1), Scotland (1), Japan (1), Netherlands (2), Romania (4), Finland (1), Norway (1), and Germany (1). Reported ethnicities of participating children were Romanian, Roma, African American, European American, Biracial, Asian, African, Eastern European, Caucasian, Hispanic, Dutch and Native American. Although not reported, it can be inferred that some further participants were Scottish, German, and Japanese.

A total of 15 555 participants were included across all 14 studies. It should be noted however that this number is increased considerably due to Minnis et al (2007) including 13 472 participants. Furthermore, four of the studies reported findings from the same pool of data (The Bucharest Early Intervention Project; BEIP) and therefore there will be some overlap in participating children. The mean number of participants reported in the studies using data from the BEIP was 156.

Overall, the gender split of participants across 14 studies was 49.7% female and 50.3% male. Minnis et al (2007) did not report exact figures in relation to the gender of their participants.

Summary of Settings

Four studies involved participants in foster care (one of which included kinship foster care and treatment foster care), three studies involved participants in institutions only, two included group institutional settings as well as foster care. One study recruited internationally adopted participants, two studies used samples from mixed care settings and two studies did not describe the care setting of participants. Four studies used BEIP data, three of which included longitudinal data; one non- BEIP study used longitudinal data. No studies utilised observational measures investigating symptoms of RAD specifically. Eleven studies were published in peer-reviewed journals, three studies were unpublished.

Table 1. Summary of studies

Study	Quality rating	Design	Sample characteristics	Measures of RAD	Results		Strengths and limitations
					Prevalence	Reported findings in relation to associated factors	
1. Shimada et al. (2015)	81.8% High	Cross-sectional, case control between groups design	<i>N=43</i> living in Japan <i>RAD group:</i> <i>n=21</i> living in care facility; Mean age 12.76 (10-17) years; 62%F / 38%M <i>n=22</i> typically developing matched on age and gender; mean age =12.95 years, 55% F / 45% M	Clinical judgement by Psychiatrists using DSM-V	n/a	RAD group found to have reduced visual cortex Grey Matter Volume (GMV) RAD and GMV associated with SDQ internalising problems ($R^2 = .55$) RAD group had sig lower FSIQ ($p < .01$), higher levels of psychiatric symptom scores ($p < .01$)	<i>Strengths</i> Control group Fairly large number exhibiting RAD <i>Limitations</i> Fairly small sample size IQ differences between groups No observation data in relation to RAD
2. Jonkman et al. (2014)	75.0% Moderate	Case control cohort study	<i>N=126</i> children living in the Netherlands in kinship and non-kinship foster families from two data pools gathered for two previous studies Mean age=60.28 (22-89) months; 50% F / 50% M <i>n=65</i> in treatment foster care, <i>n=61</i> in regular foster care	Disturbances of Attachment Interview (DAI; Smyke and Zeanah, unpublished instrument)	3% in regular foster care group had RAD and 14% in treatment foster care group	Children with RAD symptoms older entry in to care ($d = -0.90$; $p = .006$), in foster care for shorter time ($d = 0.82$; $p = .013$), and shorter time with current foster family ($d = 0.69$; $p = .035$) RAD more likely than DSED to improve following improved care conditions RAD not associated with types of abuse (physical abuse, sexual abuse or neglect), parents/teachers report of internalising or externalising symptoms, number of placements; gender	<i>Strengths</i> Control group <i>Limitations</i> Limited measures used No observational data Selection not clearly described Small numbers exhibiting RAD

3. Giltaij et al. (2015)	78.8% Moderate	Cross sectional cohort study	<p><i>n</i>=102 children with borderline or mild LD referred for psychiatric consultation in the Netherlands.</p> <p>Mean age 8.8 (5-11) years; 29% F / 71% M</p>	DAI	15.7% (<i>n</i> =16) of children had clear symptoms of RAD; 11 showed symptoms of RAD and DSED	No associations between RAD and gender, ethnic background, IQ, age, or PDD	<p><i>Strengths</i></p> <p>Fairly large number exhibiting RAD</p> <p><i>Limitations</i></p> <p>No control group</p> <p>No observational measure</p>
4. Zeanah, et al. (2005)	83.3% High	RCT – cross-sectional baseline data	<p><i>N</i>=145 children in Romania (Bucharest Early Intervention Project; BEIP)</p> <p><i>n</i>= 95 institutionalised; Mean age 23.8 (12-31) months</p> <p>45% F/ 55% M</p> <p><i>n</i>=50 never institutionalized, living with parents; Mean age 22.25 (12-31) months</p> <p>50% F/ 50% M</p>	DAI	Institutionalised children had higher levels of RAD compared to never institutionalised children (<i>M</i> = .26, <i>S.D.</i> =0.57), <i>t</i> (135)= 5.3, <i>p</i> <.001, <i>d</i> =.91.	<p>A continuous measure of attachment moderately correlated with caregiver ratings of RAD (<i>r</i>= -.44, <i>p</i><.01).</p> <p>Quality of institutionalised caregiving was related to RAD (<i>r</i>= -.32, <i>p</i>= .001)</p> <p>Institutionalized children had sig higher levels of RAD (<i>p</i><.001)</p> <p>No association between length of institutionalization and RAD</p> <p>Regardless of rearing environment, categorical organised attachment was not significantly related to caregiver ratings of RAD</p>	<p><i>Strengths</i></p> <p>RCT design</p> <p>Observational data (but not specific to RAD)</p> <p><i>Limitations</i></p> <p>Coders not completely blind to group status</p> <p>No observational measure of RAD</p> <p>No detailed historical information</p>

5. Gleason et al. (2011)	81.3% High	RCT-with longitudinal follow-up	<p><i>N</i>=136 children in Romania (BEIP)</p> <p><i>n</i>=68 receiving care as usual (institution), <i>n</i>=68 placed in foster care following institution</p> <p>Mean age at baseline 22 (6-30) months; 50%M / 50% F</p> <p>54% Romanian, 29% Roma, 15% other</p>	DAI The Preschool Age Psychiatric Assessment (PAPA)	<p>RAD at baseline (6/130; 4.6%); 30 months (4/123; 3.3%); 42 months (2/125; 1.6%) and 54 months (5/122; 4.1%)</p> <p>Non significant pattern of decline</p>	<p>RAD and DSED correlated at baseline, 30m, 42m, and 54m ($r = .40, .34, .41, \text{ and } .43, p \leq 0.001$). No association between categorical diagnoses</p> <p>No associations between exposure to institutional care and RAD at baseline</p> <p>RAD associated with caregiving quality at baseline ($r = .33$), 30m ($r = .38$), 42m ($r = .29$) and attachment security at 42 m ($r = -.51$). RAD associated with depressive symptoms at all time points ($r = .44, r = .35, r = .72$), social emotional difficulties, and functional impairment at all time points ($r = -.64, r = -.25, r = -.60$) and contributed to variance on ITSEA competence at baseline ($R^2 = .43$), 30m ($R^2 = .2$), and 42m ($R^2 = .46$) but not at 54m. RAD predicted functional impairment at 54m in children in institutions ($r = .41$). RAD associated with impairment at baseline ($R^2 = .43$), 30m ($R^2 = .2$), and 42m ($R^2 = .46$).</p>	<p><i>Strengths</i></p> <p>RCT design</p> <p>Various follow up's</p> <p>Observational measures (but not specific to RAD)</p> <p><i>Limitations</i></p> <p>As above and low rates of RAD limited statistical analyses</p>
6. Smyke et al. (2012)	85.4% High	RCT -with longitudinal follow-up	<p><i>N</i>=208 children in Romania (BEIP)</p> <p>Age at baseline 6-30 months, no mean reported. Follow up at ages 30, 42, 54m & 8 yrs</p> <p><i>n</i>=68 care as usual (institution): 51%F / 49% M, 50% Romanian, 50% Roma or other. <i>n</i>=68 placed in foster care following institution: 50% F/ 50% M, 62% Romanian, 38 % Roma or other. <i>n</i>=72 never institutionalized: 57% F/ 43% M, 92% Romanian, 8% Roma or</p>	DAI	<p>Categorical prevalence not specified. RAD decreased differentially for usual care and foster care groups ($p < .01$)</p> <p>Differences between foster care group and never-institutionalised group found at baseline, 30m and 8 years ($p < .01$). RAD in children placed in foster care <24m did not differ from children placed > 24m of age</p>	<p>In the institution group only, more signs of RAD found in those with lower baseline cognitive abilities across intervention period ($t = 4.24, df = 127, p < .0001$)</p> <p>Effect sizes were not reported</p>	<p><i>Strengths</i></p> <p>RCT design</p> <p>Fairly large sample size</p> <p>Follow ups</p> <p><i>Limitations</i></p> <p>As above</p>

			other				
7. McGoron et al. (2012)	80.0% High	RCT with longitudinal follow-up	<p>Total n=136 children in a Romanian institution (BEIP)</p> <p>Mean age 22.0 (6-30) months. Follow up at ages 30, 42 and 54 months</p> <p>“Slightly more than half” of children were Romanian and female.</p>	DAI	Not reported	<p>Associations between symptoms of RAD at 54 m and 30m caregiving quality ($R^2 = -.32$), 42-month attachment security ($R^2 = -.44$), 54 month DSED ($R^2 = .44$), 54m stereotypies ($R^2 = .45$), 54m internalising disorders ($R^2 = .28$) and 54m functional impairment ($R^2 = .40$) were found.</p> <p>Association between 30m caregiving quality and symptoms of RAD at 54m ($R^2 = .10$)</p> <p>RAD not associated with ethnicity or 54m externalising disorders</p>	<p><i>Strengths</i></p> <p>RCT design</p> <p>Follow ups</p> <p><i>Limitations</i></p> <p>As above and magnitude of associations fairly small</p>
8. Zeanah et al. (2004)	77.7% Moderate	Retrospective cohort study	<p>Clinicians treating toddlers in foster care in the USA, n=94. Mean age 27.7 (10-47) months</p> <p>56%F/ 44%M</p> <p>60% African American, 28% European American, 8% biracial, and 4% other</p>	DAI “interview probes” around DSM/ICD-10 criteria	RAD found in 35%	<p>RAD associated with mothers’ psychiatric history ($R^2 = .120$, $p < .001$)</p> <p>Mothers’ education, teenage parent, partner violence, criminal history, depressed mood, maltreatment as a child, and history of substance abuse found not to be associated with RAD. No relationships between RAD and gender, ethnicity, or length of time in care were found.</p>	<p><i>Strengths</i></p> <p>High levels of RAD may increase statistical validity</p> <p><i>Limitations</i></p> <p>No control group</p> <p>No observational data</p> <p>Researchers aware of maltreatment history when conducting interviews</p> <p>Low level of variance predicted</p>

9. Elovainio et al. (2015)	73.8% Moderate	Cross sectional cohort study	<p><i>n</i>= 853 internationally adopted children in Finland</p> <p>Mean age 8.5 (6-15) years; 54% F/ 46% M</p> <p>Asian (51%), African (11%), American (16%), Eastern European (22%)</p>	The attachment-related symptoms measure	<p>RAD occurred in 18% (<i>n</i>= 137)</p> <p>Symptoms of both RAD and DSED co-occurred in 25% (<i>n</i>= 214)</p>	<p>RAD associated with emotional problems, behavioural problems, and ADHD (hyperactive symptoms, not attention problems) ($R^2=-0.22$). RAD children also found to have higher internalising, externalising, and total CBCL problem scores ($R^2=.33$)</p> <p>Some effect sizes not reported</p>	<p><i>Strengths</i></p> <p>Large sample size</p> <p><i>Limitations</i></p> <p>No control group or follow up</p> <p>No observational data</p>
10. Sheaffer (unpublished, 2010)	75.8% Moderate	cross-sectional survey	<p><i>n</i>= 34 children receiving treatment in USA for RAD, in mixed care settings.</p> <p>Mean age 10.6 (6-19) years 44%F/ 56% M 22 (64.7%) Caucasian, 3 (8.8%) African American, 2 (5.9%) Bi-racial, 2 (5.9%) Hispanic, 2 (5.9%) Native American, and 3 (8.8%) Romanian</p>	Relationship Problems Questionnaire (Minnis et al, 2007)	n/a	<p>RAD symptoms correlated with number of placements ($r =.587$, $p = .003$)</p> <p>no associations between RAD and age, age removed from home, years in care, or years in therapy</p> <p>Association between RAD and attribution bias for recognition of happy in other facial expressions ($r=.39$)</p>	<p><i>Strengths</i></p> <p>Recruitment procedures well described</p> <p>Exploration of RAD in older children</p> <p><i>Limitations</i></p> <p>No control group or follow up</p> <p>Small sample size</p> <p>No observational data</p> <p>Missing historical information</p> <p>Purposeful sampling strategies and small referral base</p>

11. Lehmann et al. (2015)	77.7% Moderate	Cross sectional questionnaire cohort study	<p><i>n</i> = 122 foster children living in foster care in Norway</p> <p>Mean age 8.0 years (ranging from 6-10 years)</p> <p>57% F/ 43% M</p> <p>No ethnicity information reported</p>	The DAWBA RAD	Not reported but authors note mean RAD scores were sig lower than DSED scores ($p < .001$)	<p>Male gender ($r = -.18$), parental mental disorders ($r = .21$), conduct problems ($r = .79$), hyperactivity ($r = .77$) and DSED ($r = 0.59$) associated with RAD. RAD associated with functional impairment ($r = .44$) and help seeking from services ($r = .011$)</p> <p>RAD did not predict contact with school psychology services, nor associated with parents substance abuse, violence exposure, number of placements, years in current foster home, age placed in current foster home, age at first placement or age</p>	<p><i>Strengths</i></p> <p>Reasonable sample size</p> <p><i>Limitations</i></p> <p>Size of sample reduces the power of some statistical analyses</p> <p>Relatively low scores on the RAD scale</p> <p>No observational data</p>
12. Moran (unpublished, 2014)	81.8% High	Cross-sectional Cohort study	<p><i>n</i> = 29 youth justice participants in Scotland</p> <p>Mean age 16.2 years (ranging from 12-17 years)</p> <p>34% F / 66% M</p> <p>No ethnicity information reported</p>	<p>RPQ</p> <p>Adapted Child and Adolescent Psychiatric Assessment, Reactive Attachment Disorder (CAPA RAD; Minnis et al., 2009)</p> <p>Adapted Observational Schedule for RAD (McLaughlin, Espie & Minnis, 2010)</p>	10% had RAD, 10% had mixed presentation of RAD and DRAD. 10% had borderline RAD (RAD & DSED combined)	<p>RAD associated with SDQ total difficulty scores, hyperactivity ($R^2 = .50$, $p = .006$), prosocial behaviour ($R^2 = -.59$, $p = .001$) and conduct problem items ($R^2 = .44$, $p = .018$)</p> <p>RAD not associated with emotional symptoms or peer problems</p>	<p><i>Strengths</i></p> <p>Power calculation included</p> <p>Observational measure used</p> <p><i>Limitations</i></p> <p>Small sample size; exclusions due to acute mental health problems or chaotic circumstances, may limit validity</p> <p>Measures completed by staff who had known the young people > 1 month only</p> <p>Limited historic information; no control group or follow up</p>

13. Minnis et al. (2007)	75.8% Moderate	Retrospective Twin Cohort study	<p>$n=6,736$ twin pairs Mean age= 7.9 years</p> <p>The study reports that minor differences in ethnicity and maternal educational attainment were found, no further details reported.</p> <p>51.3% F/ 48.7%M</p>	RPQ	Prevalence not reported	<p>Factor analysis suggested RAD is distinguishable from other child psychiatric symptoms.</p> <p>RAD associated with harsh parenting ($p<.001$) parental negativity ($p<.001$), and parental positivity (negatively). Association between RAD symptoms and monozygotic pairs in both males ($r=0.880$) and females ($r=0.846$) and dizygotic pairs: males ($r=0.571$) females ($r=0.713$)</p> <p>For males, majority of variance in RAD due to additive genetic effects. Majority of the variance in RAD for females due to shared environmental effects</p>	<p><i>Strengths</i></p> <p>Large sample size</p> <p><i>Limitations</i></p> <p>Non-clinical and unrepresentative sample</p> <p>Response rate <50%</p> <p>No observational data</p>
14. Zimmerman (Unpublished thesis 2015)	79.5% Moderate	Longitudinal cohort study	<p>$n= 55$ foster children in Germany.</p> <p>Mean age 33.4 (S.D = 18.7) months.</p> <p>49% F/ 51% M</p> <p>No ethnicities reported.</p>	DAI Reactive Attachment Disorder Questionnaire (Minnis et al., 2002)	<p>5.5% ($n=3$) at T1, 1.8%($n=1$) at T2.</p> <p>reduction of RAD during the first year of placement, $p < .001$. Namely in first 6 months, $p = .003$. No significant change over time found when using RAD Questionnaire.</p> <p>Emergency foster parents described significantly more RAD symptoms than long term foster carers ($p= .006$)</p>	<p>RAD associated with parental mental illness (T3 $R^2= .43$)</p> <p>RAD not associated with early adversity at T1 or T2, mental illness of parents at T1, number of placements at T1 or T2, and visitation with parents at T1 or T2</p> <p>Age at placement ($R^2= .07$) and severity of early adverse care ($R^2= .27$) predicted RAD symptoms at baseline. RAD at 12m was predicted by mental health problems of biological parents ($R^2= .08$) and age at placement, pre-placement characteristics no longer predictive.</p>	<p><i>Strengths</i></p> <p>Multiple RAD measures</p> <p>Follow up</p> <p>Use of observational measures (not specific to RAD)</p> <p><i>Limitations</i></p> <p>Singular cases of RAD limits statistical analysis</p> <p>No observational measure of RAD</p> <p>At baseline foster parents might not know the children well enough for reliable rating</p>

Key Findings

Caregiving

The following caregiving factors were found to be associated with RAD: institutionalisation (Study 4); quality of institutionalised caregiving (Study 4; Study 7; Study 5 found this at all time points except 54 months); harsh parenting and parental negativity, parental positivity (negative association) (Study 13); and parental mental health problems (Study 8; Study 11; Study 14, T2 only). Severity of early adverse care did not correlate significantly with RAD in longitudinal research but was found to predict RAD symptoms at baseline (Study 14).

RAD was not found to be associated with the following factors relating to caregiving environment: particular types of abuse (physical abuse, sexual abuse or neglect) (Study 2); exposure to violence (Study 11); length of time in care (Study 8; Study 10; Study 11) or an institution (Study 4; Study 5); mothers' education, teenage pregnancy, partner violence, criminal history, depressed mood, mothers' maltreatment as a child (Study 8); parental substance misuse (Study 8; Study 11); and visitation with birth parents (Study 14).

There were contradictory findings in relation to length of time in care, number of previous placements and attachment patterns. Study 2 found a shorter period of time in foster care to be associated with RAD, whereas other studies found no such association (Study 8; Study 10; Study 11). This difference may be due to varying measures of RAD across studies (Study 2 used the DAI where as Study 8; Study 10; Study 11 used the DAI with interview probes, RPQ, and DAWBA RAD, respectively). Cultural differences or differences in mean age of participants across studies (5 years, 8 years, 11 years, and 2 years) may also have contributed to the difference in findings. Study 2 had the largest sample size (n=126) of the studies exploring RAD and its relationship with time in foster care, which may account for the significant association. Study 10 found a positive correlation between RAD symptoms and number of placements whereas Study 2, Study 11 and Study 14 did not. Study 10 had a smaller sample size (n=34) but all of the children participating had RAD, which may add more validity to the findings. A limitation reported by Study 11 and Study 14 was the relatively low occurrence of RAD symptoms in their samples, a limitation that also applies to Study 2. Using the DAI and Strange Situation Procedure (Ainsworth & Bell, 1970), Study 4 and Study 5 found an association between RAD and attachment patterns. Using a different categorisation of attachment (organised versus non-organised), Study 4 also found that organised attachment was not associated with RAD across care settings. In the institutionalised group in which this study was conducted, however, this finding may lack validity because of the small number of children meeting the criteria for the organised attachment category: the authors state that most institutionalised children had failed to organise an attachment with their preferred caregiver.

Individual characteristics

The following characteristics and demographic factors were found to be associated with RAD: Male gender (Study 11); older age when placed in to care (Study 2; Study 14); reduced grey matter volume (Study 1); lower cognitive ability in institutionalised children (Study 6); attribution bias for recognition of happiness in other facial expressions (Study 10); and genetic factors, particularly for males (Study 13).

Individual and demographic factors found not to be associated with RAD were: gender (Study 2; Study 3); age (Study 3; Study 10; Study 11); age when placed in current foster home (Study 11); ethnic background (Study 3; Study 7; Study 8); age when placed in to care (Study 6; Study 10; Study 11); pervasive developmental disorder or IQ in individuals with a mild learning disability (Study 3); cognitive ability in institutionalised children moved to foster care (Study 6).

There were conflicting findings in relation to RAD and gender, age when placed into care, and cognitive functioning. A number of studies reported contradictory findings in relation to demographic variables with no clear link between the quality or sample size of the studies and the findings that they reported. This suggests that further research exploring RAD in males and females of varying ages is required. It should be noted that Study 6 is rated as a high quality paper reporting no significant differences between RAD and age placed into care however the study solely made comparisons across two groups, participants placed in to care before 24 months and after 24 months of age (a non significant pattern of lower scores for those placed in care <24m was observed) and for such categorical analysis, study power may not have been great enough to demonstrate small effects. Studies that reported a relationship between RAD and age at placement (Study 2 and Study 14) explored age as a continuous variable. In Study 2 however, only 11 children in the sample exhibited symptoms of RAD thus potentially limiting generalizability of findings. Given what is known about early intervention being most effective, it would be expected that being placed into care at an earlier age would be associated with fewer symptoms of RAD. When considering this factor, it is important to note that this is an institutional sample in a country and at a time when children placed into institutional care (e.g. Study 6) may not have experienced the improved care conditions that may be expected when removing a child from maltreatment; therefore limiting the association between improvement in RAD symptoms and age removed from care. Differences in associated cognitive functioning were observed by the same study but only across groups in differing care settings. Study 3 reported no association with cognitive functioning but the study specifically involved children with a learning disability. Study 6 found an association between lower IQ and age but it is difficult to make direct comparisons with Study 3 as Study 6 had a larger sample size including children with a wider range of cognitive ability. Study 3 was rated as moderate quality where as Study 6 was rated as high quality due the larger sample size and RCT design.

Psychological and behavioural factors

The following psychological and behavioural factors were found to be associated with RAD: depressive symptoms (Study 5); social and emotional difficulties (Study 9; Study 5); functional impairment (Study 5; Study 7; Study 11); behavioural and conduct problems (Study 9; Study 11; Study 12); hyperactivity (Study 9; Study 12; Study 13); more CBCL/PAPA/SDQ internalising symptoms (Study 1; Study 7; Study 9), more CBCL externalising symptoms (Study 9); higher total difficulty SDQ/CBCL scores (Study 9; Study 13; Study 12); stereotypies (Study 7); help seeking from services (Study 11); and symptoms of DSED (Study 5; Study 7; Study 11).

Psychological and behavioural factors not associated with RAD were: reported CBCL internalising symptoms (Study 2), CBCL and PAPA externalising symptoms (Study 2; Study 7); emotional symptoms or peer problems (Study 12); attention problems (Study 9); years in therapy (Study 10); incorrect selection of sad or fearful facial expressions (Study 1); categorical diagnoses of DSED (Study 5).

There were differing findings in relation to RAD and its relationship with emotional difficulties, internalising and externalising symptoms, and symptoms of DSED. It is important to consider that a number of these studies (e.g. Study 2, Study 11) reported low levels of RAD symptoms in their samples, which may increase the risk of a type II error (failing to find statistical significance due to small sample size), particularly when exploring RAD as a categorical diagnosis. Furthermore, study designs and sample sizes varied considerably. Study 13 used a non-clinical sample, which may limit generalisability of findings to RAD populations. Study 12 found no association between RAD and emotional or peer problems however the study had a small sample size (n=29) consisting of adolescents in which levels of emotional and peer problems may have been high with little variance; whereas the studies reporting social emotional difficulties (Study 5; Study 9) had considerably larger samples (n=136, n=853 respectively) and participants were younger (means 22 months and 8.6 years respectively).

Prevalence and Stability of RAD

In foster care samples, the prevalence of RAD was reported to vary between 3% and 35%. RAD was found to be present in 3%, 14% (Study 2, regular foster care group and treatment foster care group respectively), 5.5% (dropping to 1.8% following one year in foster care, Zimmerman, unpublished 2015) and 35% (Study 8) of samples. Mixed symptoms of RAD and DSED were found in 3% and 15% (Study 2). Some studies referred to a RAD diagnosis whereas other studies (e.g. Study 8) referred to a presence of RAD symptoms. This and the fact that clinicians rather than carers were providing clinical information may explain why Study 8 reported a higher prevalence.

Study 12 investigated RAD in a small youth justice sample (n=29) that consisted of adolescents living in differing care settings. This study found that 10% of the sample had RAD, 10% had a mixed presentation of inhibited and disinhibited RAD and it was noted that a further 10% had “borderline RAD” (where data for multi-informant diagnoses were missing or for unclear/mixed presentations of inhibited and disinhibited forms). The range of placement moves in the sample varied from 0-12, suggesting that some participants were living with birth families and it is likely that many had spent time in foster care and/or residential units. As the study was conducted in Scotland, where young children are not placed in institutional settings, it is fair to assume that the sample were unlikely to have spent their early years living in institutions.

Study 9 had a large sample size (n=853) and found that RAD occurred in 18% of internationally adopted children and symptoms of both RAD and DSED co-occurred in 25%. In this study, the care setting prior to adoption was not reported. Study 3 found 15.7% of children with a borderline or mild learning disability referred for psychiatric consultation had clear symptoms of RAD and 10.7% showed clear symptoms of DSED as well as RAD. However, the care setting of participants was not reported.

In institutionalised samples, prevalence of RAD was found to be 4.6% (dropping to 3.3% at 30 months, 1.6% at 42 months and 4.1% at 54 months following foster care, Study 5).

In summary, the prevalence of RAD varies widely across studies and it is difficult to make comparisons across samples given the considerable differences in care settings, historical care settings, role of informants, measures used to inform RAD, and use of diagnosis versus symptomology of RAD. This may account for the unusual finding of RAD being apparently no more prevalent in institutionalised samples than in clinical and some foster care samples. With the exception of Study 5 and Study 12 that were rated as high quality, studies reporting prevalence were rated as moderate quality. Study 5, Study 6 and Study 14 followed prevalence over time, which provided helpful insights in to the stability of the disorder: symptoms appear to decrease over time with improved care conditions. Furthermore, using data from the Bucharest Early Intervention Project, Study 5 and Study 6 utilised control groups, which increases the ecological validity of their findings.

Summary of general research limitations in the field

Across all of the studies, there is a heavy reliance on the DAI to inform symptomology of RAD and a lack of observational measures that could introduce bias. Furthermore, the DAI has not been normed and does not provide clinical cut-offs for RAD. Research has found low consistency between carer reports of RAD on the DAI and observed symptoms (Corval, unpublished personal communication) and significant differences between DAI ratings informed by two different foster carers (Study 14). Therefore, to increase reliability, it may be helpful, in addition to carer report, to include an

observational measure to inform a diagnosis of RAD. At present, there is no published validated observational measure of RAD.

Another potential source of bias is the somewhat limited pool of data; data from the Bucharest Early Intervention Project are used in almost one third of the reported studies exploring factors associated with RAD.

Discussion

Although there is limited research exploring reactive attachment disorder, this review has shown that there are a number of moderate to high quality studies from around the world investigating the disorder and its associated factors. RAD was found to be associated with many psychological, situational and behavioural factors. These associations, however, were not consistent across the reviewed studies. This may be due to a number of likely confounding factors in studies of maltreated children as well as small sample sizes, low levels of RAD symptoms and a wide variety of participant samples. The exploration of associated factors was often limited by the lack of historical information pertaining to children in care settings. Further, studies were limited by their lack of consistency in measures used to inform the presence of RAD and no studies utilised observational assessment of RAD.

The review demonstrated that although rare, RAD is an established disorder that occurs consistently in a minority of children who have been maltreated. Prevalence varied widely, ranging from 1.6% to 35% with no observable differences between prevalence in institutional samples and foster care samples. It is difficult to make direct comparisons across studies due to the variance in measures of RAD and samples, for example care setting varied widely and ages of participants ranged from six months to 19 years old. Given that RAD is thought to be most prevalent in institutionalised children, it is logical that studies are often conducted in countries where, unlike the UK, the use of institutional care for maltreated young children is still common practice. However, in order for findings to be generalisable internationally, future research in to RAD and maltreated children in family care settings is important. Furthermore, studies with between group designs, allowing for comparisons between maltreated children without RAD (or with other diagnoses) and maltreated children with RAD, would further inform what factors are associated with RAD in particular.

Four of the studies included utilised data from the Bucharest Early Intervention Project. Although it provides an incredibly useful source of information, the widespread use of this pool of data increases the potential for bias. The BEIP is a high quality study nevertheless some important methodological limitations have been noted by the authors, for instance a reliance on translation of measures to Romanian, which may

cause some inaccuracies. Furthermore the kind of extreme environments found in institutions developed under Ceausescu are thankfully unusual in most countries and this may limit generalisability.

A considerable number of papers were excluded from this review due to their focus on DSED (previously disinhibited reactive attachment disorder) only. Additional studies were excluded as they explored the two disorders collectively; often this was not made clear which could be misleading for the reader. Exploring associated factors without separating the two disorders skews what is known about the inhibited form as DSED is much more prevalent.

Research tends to be heavily reliant on the DAI for informing a diagnosis of RAD. Almost all studies lacked observational measures and no observational measures specific to RAD were utilised. The development of observational scales and multi-informant diagnoses of RAD is likely to improve validity and improve the clinical value of research findings (De Los Reyes et al., 2015).

Given the nature of the systematic review research question, many studies included used a correlational design to investigate factors associated with RAD. A correlational design can be criticised for lacking the ability to investigate causation between variables. "A concern in any study examining correlates of attachment is whether there are variables other than attachment that might account for the findings" (Kerns, Abraham, Schlegelmilch & Morgan 2007, p. 36). In addition, according to Hills' (1965) criteria of causation, if a relationship is causal it is expected to be demonstrated consistently across different studies and among different populations. The inconsistency between reported findings in relation to factors associated with RAD limits the likelihood of an evident causal relationship. Studies that included regression analyses acknowledged that the factors explored accounted for a fairly low amount of variance in RAD, suggesting that further research investigating other potential predictive factors is warranted. Research would benefit from study designs with an ability to draw causal conclusions in relation to RAD rather than just associations. Longitudinal studies where outcomes are measured following exposure and there is potential to investigate dose-response relationships would be more valid, however such studies involving child maltreatment and RAD would need to address important ethical considerations. The BEIP is unique in applying RCT methodology to an investigation of care setting and it has demonstrated that family care is an effective intervention for RAD. It is likely that future research into care placements as interventions will not be possible due to ethical considerations, so trial investigations into other interventions for RAD will be necessary.

Review Limitations and Strengths

The results of this review are limited to papers published in English. A strength is the inclusion of unpublished research as this minimises publication bias; however a number of unpublished papers were excluded due to the author being unable to obtain full

access to the articles. Additionally, a lack of homogeneity across samples and measures limits the ability to make comparisons across studies. A further limitation of the review may be the quality appraisal tool used as, similar to many other tools used in systematic reviews, it did not weight items so it is possible that scoring may not be wholly representative of quality.

Conclusion and Clinical Implications

The understanding of RAD, its prevalence, stability, and clinical profile is still in relatively early stages. Further understanding of the disorder will begin to inform increased clinical recognition and the development and evaluation of interventions. At this stage, little research has explored therapeutic interventions but it is encouraging that intervention in the form of improved care giving environment seems effective in decreasing symptoms of RAD over time.

Research into RAD has been emerging since the DSM-V has separated it from DSED, the more prevalent disorder. Although 14 papers were found, the majority were from studies in the past five years and four papers were from a related study. It is hoped that more research focusing on solely RAD will be conducted. In summary, it has been highlighted that more research informing a valid assessment of RAD is imperative, including the validation of observational tools. Investigating factors associated with, and the prevalence of RAD may help increase our understanding of the disorder's development and stability. In turn, an increased understanding could inform intervention and ultimately prevention of a disorder that is thought to have a considerable social, emotional, and functional impact.

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Reactive Attachment Disorder in Infants in Foster Care and associated Mental Health and Cognitive Functioning

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Plain English Summary

Background

Reactive attachment disorder (RAD) has been described as one of the least researched and most poorly understood psychiatric disorders. According to the DSM-V, RAD is defined as:

- A consistent pattern of inhibited, emotionally withdrawn behaviour toward caregivers.
- Persistent social and emotional disturbance.
- The child has experienced patterns of extremes of insufficient care.

The stability of RAD symptoms over time is relatively unknown. In addition, there is limited research exploring mental health difficulties and intellectual functioning in children with RAD and it would be useful to explore this further.

Until recently, it has been difficult to investigate the presence of RAD due to limited measures for informing a diagnosis. However, this study will explore the validity of a new tool that has been developed (The Rating of Inhibited Attachment Behavior; RInAB).

In summary, we know little about the prevalence and stability of RAD in maltreated infants coming into foster care, virtually nothing about the associations between RAD and other mental health problems. Furthermore, there are no well-validated observational tools to examine RAD.

Aims

- To establish the prevalence of RAD in a maltreated sample shortly after placement in foster care and the stability of RAD one year later.
- To explore the validity of the RInAB Scale.
- To establish the association between symptoms of RAD and mental health difficulties and cognitive functioning.

Methods

Participants

The sample consisted of 55 maltreated children between the ages of 12 and 60 months.

Inclusion and Exclusion Criteria

All parents/carers with a child aged 12-60 months who came into a period of care due to child protection concerns were invited to take part. Children were excluded from the study if:

- They had a profound learning disability and/or
- Their primary caregiver was unavailable to take part in the study

Recruitment Procedures

The sample had been recruited for an on-going trial and video footage is stored on a hard drive. Assessments were administered one month after a child became accommodated into care and again, one year later.

Consent

Informed Consent from the parents and foster carers of potential participants was obtained by a recruitment officer (study social worker).

Design

This is a prospective longitudinal cohort study.

Data Collection

RAD diagnoses were made based on the various assessments and video recorded interaction between the child and their carer.

Main Findings and Conclusions

Prevalence of RAD was found to be 7.3% at T1 and at T2, only 4.3% met a borderline RAD diagnosis. Levels of observed RAD symptoms decreased significantly at T2 in comparison to T1 but carer reported symptoms of RAD did not. Children whose RAD symptoms did not improve were found to be older and showed less prosocial behaviour. Differences between observed symptoms and carer reported symptoms of RAD were noted. RAD was associated with some mental health problems and cognitive difficulties. Lower Verbal IQ and unexpectedly, prosocial behaviour, were found to be predictive of RAD symptoms.

RAD is likely to have profoundly negative effects on the development of children. Findings from the study in hand will hopefully provide a greater insight into RAD however further research is needed. Such findings will be pertinent for professionals working with children, particularly those children who may have been maltreated. Given the inhibited nature of the disorder, children with RAD can be easily missed.

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Abstract

Background: Reactive attachment disorder (RAD) has been described as one of the least researched and most poorly understood psychiatric disorders (Chaffin *et al.*, 2006). Despite this, given what is known about maltreatment and attachment, it is likely that RAD has profound consequences for child development. Very little is known about the prevalence and stability of RAD symptoms over time. Until recently it has been difficult to investigate the presence of RAD due to limited measures for informing a diagnosis. However this study utilised a new observational tool **Method:** A cross sectional study design with a one-year follow-up explored RAD symptoms in maltreated infants in Scotland (n=55, age range= 16-62 months) and associated mental health and cognitive functioning. The study utilised the Rating of Inhibited Attachment Behavior Scale (Corval, et al., unpublished 2014) that has recently been developed by experts in the field along side The Disturbances of Attachment Interview (Smyke & Zeanah, 1999). Children were recruited as part of the *BeST* trial, whereby all infants who came in to the care of the local authority in Glasgow due to child protection concerns were invited to participate. The study sample was representative of the larger pool of data in terms of age, gender, mental health and cognitive functioning. **Results:** The sample was found to be representative of the population of maltreated children from which it was derived. Prevalence of RAD was found to be 7.3% (n=3, 95% CI [0.43 – 14.17]) at T1, when children are first placed in to foster care. At T2, following one year in improved care conditions, 4.3% (n=2, 95% CI [below 0 – 10.16]) met a borderline RAD diagnosis. Levels of observed RAD symptoms decreased significantly at T2 in comparison to T1 but carer reported symptoms of RAD did not. Children whose RAD symptoms did not improve were found to be significantly older and showed less prosocial behaviour. RAD was associated with some mental health and cognitive difficulties. Lower Verbal IQ and unexpectedly, prosocial behaviour were found to predict RAD symptoms. **Conclusions:** The preliminary findings have added to the developing understanding of RAD symptoms and associated difficulties however further exploration of RAD in larger samples would be invaluable.

Keywords: *Reactive attachment disorder, child maltreatment, inhibited attachment, foster care*

Attachment is a fundamental instinct across species, whereby to protect itself from threat and to survive in its environment, an infant instinctively seeks to be close to its caregiver when distressed (Bowlby, 1969). In humans, as well as physical safety, the formation of secure attachment relationships allow for positive social development and emotional regulation, which protects against mental health problems (Prior & Glaser, 2006). Children with an attachment disorder such as Reactive Attachment Disorder (RAD) do not appear to demonstrate important attachment behaviours such as seeking or signalling to their caregiver when experiencing distress. There appears to be a deactivation of the attachment system in that these children do not seek and accept comfort or signal distress when frightened or hurt (Prior & Glaser, 2006). It is widely accepted that RAD exclusively occurs in the context of maltreatment, where the infant's attachment needs have been consistently neglected from a young age. Children with RAD may be socially and emotionally withdrawn in a wide range of situations. This is likely to limit children's ability to make use of love or care from others and reduce opportunities for learning. Therefore, it is likely that RAD has a considerable negative impact on child development (Prior & Glaser, 2006).

RAD was first defined in 1980 and it has been revised several times since (Zeanah & Gleason, 2010). It has been described as "one of the least researched and most poorly understood disorders listed in the DSM" (Chaffin et al., 2006, p.80). There were previously two forms of the disorder as defined by the DSM-IV and DSM-IV-TR (APA, 1994; 2000), these being 'inhibited reactive attachment disorder (I-RAD)' and 'disinhibited reactive attachment disorder (D-RAD)'. The ICD-10 divided the subtypes into two distinct disorders and more recently the DSM-5 (APA, 2013) has similarly updated its classifications. The previously termed inhibited form is now defined as 'Reactive Attachment Disorder' (RAD) and the previously termed disinhibited form is now classified as 'Disinhibited Social Engagement Disorder' and is no longer considered a disorder of attachment (Zeanah & Gleason, 2015). The DSM-5 defines RAD as "a consistent pattern of inhibited, emotionally withdrawn behaviour towards adult caregivers, and persistent social and emotional disturbance, in the context of extreme patterns of insufficient care". The international classification of diseases 10th edition (ICD-10; WHO, 2010) details that children with RAD may exhibit misery, huddling, clinginess, an inappropriate lack of response, or aggression.

Researchers are fairly confident about the prevalence of disinhibited social engagement disorder. The prevalence of RAD, however, is less known but appears to be a rarer disorder (Corval, Baptista, Fachada, Beiramar & Soares, 2014; Gleason et al., 2011). The DSM-5 (APA, 2013) states that less than 10% of children who have been severely neglected develop RAD and it is considered to be most common in children with an experience of institutionalisation (Corval et al., 2014). Only a relatively small number of studies have investigated the prevalence of RAD distinctly and of those that have, the findings vary widely across studies. Furthermore, it is difficult to make comparisons across studies given the considerable differences in care settings, historical care settings, role of informants, measures used to inform RAD, and use of diagnosis versus

symptomology of RAD. In foster care samples, the prevalence of RAD has been found to vary between 3% and 35% (Jonkman et al., 2014 and Zeanah et al., 2004 respectively). Few studies have explored prevalence over time. In conducting the Bucharest Early Intervention Project with previously institutionalised Romanian children, Gleason et al., (2011) explored RAD over time and found that the number of children meeting diagnostic criteria varied at each time point (4.6% at baseline, 3.3% at 30 months, 1.6% at 42 months and 4.1% at 54 months). Zimmerman (unpublished 2015) investigated RAD in foster children over one year and found a prevalence of 5.5% (n=) at T1 and 1.8% (n=1) at T2.

Some researchers argue that further clarity around the definition of RAD is needed. Zeanah and Gleason (2010) propose that symptoms of RAD are signs of current maltreatment rather than a persistent disorder. Zeanah, Mammen and Lieberman (1993) assert that the frozen watchfulness associated with RAD is, in fact, a response when confronted by an abusive caregiver rather than an expressed sign of attachment disorder. In order to be a true disorder, RAD would have to be pervasive across different situations. If RAD were simply a 'state' associated with current maltreatment, it may be expected to disappear once a child is placed in a stable, nurturing foster family. Jonkman et al. (2014) found that if children experienced an improvement in caregiving conditions (being placed in foster care) RAD persevered less than DSED. This study reported negative associations between RAD symptoms and time in foster care and time in current placement. Jonkman et al. (2014) went on to report that, following improved caregiving settings, RAD symptoms disappeared. Other studies however have found that although prevalence of RAD decreases, it persists after one year in a foster care placement (Zimmerman, unpublished 2015). Therefore, it is unclear if RAD is a state associated with current maltreatment or if it is a disorder that is pervasive across time and contexts.

Until recently it has been difficult to investigate the presence of RAD due to limited measures for informing a diagnosis specific to RAD. However, The Rating of Inhibited Attachment Behavior (RInAB) (Corval, Baptista, Fachada, Beiramar & Soares, unpublished 2014), an observational tool for the assessment of RAD has now been developed by a group of experts in the field.

There is limited research exploring the mental health of children with RAD, however behaviours indicative of attachment disorders have been shown to be distinct from conduct problems, emotional problems and hyperactivity (Minnis et al., 2007). Yet, given the link between early childhood psychopathology and difficulties in the parent-child relationship (Stovgarrd et al., 2007), it is likely that children with symptoms of RAD have a higher likelihood of experiencing mental health difficulties. Millward et al. (2006) found a significant association between attachment disorders and other mental health symptoms ($r = 0.84$), however, this study was not specifically exploring RAD. Moran (unpublished 2014) explored RAD independently in a youth justice population (12-17 years) and found a strong association between RAD and other mental health symptoms with a large affect size ($R^2 = .6$).

Further research investigating RAD specifically has shown that it is associated with: depressive symptoms (Gleason et al., 2011); social difficulties (Elovainio, Raaska, Sinkkonen, Makipaa & Lapinleimu, 2015; Gleason et al., 2011); and higher total difficulty scores (Elovainio et al., 2015; Minnis et al., 2007; Moran, unpublished 2014). However, Minnis et al. (2007) explored difficulties in a non-clinical sample, which may limit generalisability of findings to RAD populations. There are conflicting findings in relation to RAD and its relationship with emotional difficulties, internalising difficulties and externalising difficulties. Elovainio et al. (2015) and Gleason et al. (2011) found an association between RAD and emotional problems whereas Moran (unpublished 2014) did not. Studies have found associations between RAD and internalising difficulties (McGoron et al., 2012; Elovainio et al., 2015) whereas Jonkman et al. (2014) found no such association. Elovainio et al. (2015) reported an association between RAD and externalising difficulties, however McGoron et al. (2012) did not and Lehmann, Breivik, Heiervang, Havik and Havik (2015) found no association with either internalising or externalising difficulties. However, it should be noted that studies finding no associations often reported low levels of RAD symptoms in their samples (for example Lehmann et al., 2015 and Jonkman et al., 2014) and had smaller sample sizes, which may increase the risk of a type II error, particularly when exploring RAD as a categorical diagnosis. In addition, differences in findings may be due to the differing age ranges of samples, warranting further longitudinal research.

With regard to cognitive functioning, research has found that RAD is associated with lower cognitive ability in institutionalised children (Smyke et al., 2012). Other studies have found that both forms of RAD collectively (Pritchett, et al., 2013b) and RAD independently (Gleason et al., 2011) are associated with below average cognitive functioning. Furthermore, studies combining both types of RAD have demonstrated associated language difficulties (Minnis et al., 2009; Sadiq et al., 2012). It would be useful to further consider the relationship between cognitive functioning, verbal comprehension and RAD specifically.

In summary, there is insufficient evidence regarding the prevalence and stability of RAD symptoms, particularly in non-institutionalised samples of maltreated children. Furthermore, very little is known about the relationships between RAD, other mental health problems and cognitive functioning. This study is an attempt to address some gaps in the scientific literature and investigate RAD in maltreated infants over a one-year time period.

Aims and Hypotheses

Aims

- To establish the prevalence and stability of RAD symptoms in a maltreated sample, comparing symptoms shortly after placement in foster care (Time 1) to the level of symptoms exhibited after one year in foster care (Time 2).

- To explore the relationships between symptoms of RAD and mental health difficulties and cognitive functioning.

Hypotheses

- It was hypothesised that the level of RAD symptoms would reduce over time but clinical levels of symptoms would remain for some.
- It was hypothesised that symptoms of RAD would be significantly associated with other mental health difficulties and lower cognitive functioning.

Method

Participants

The sample consists of 55 children aged between 16 and 62 months who have been accommodated in to the care of local authority Social Work in the Scottish city of Glasgow. The sample was recruited for the BeST² Services Trial, an on going randomised control trial investigating an infant mental health intervention (Clinical Trials.gov trial registration number NCT01485510:<https://clinicaltrials.gov/ct2/show/NCT01485510?term=New+orleans&rank=3>).

Inclusion and Exclusion Criteria

All parents (or recognised parental guardians) with a child aged between approximately 6 and 60 months who come into a period of care due to child protection concerns are invited to take part in the BeST² Trial (Pritchett et al., 2013a; from which data are being used). Children are excluded from the trial if they have a profound learning disability (as some assessment measures would not be appropriate) or their primary caregiver is unavailable to take part in the intervention (such as long-term imprisonment, death, or being uncontactable by services or the research team for 3 months or more).

Additional exclusion criteria for the current study: Children under 12 months old were excluded as “in typical development, selective attachment behaviours develop up until this age” (Schofield & Beek, 2006), therefore measures may not be appropriate for younger children. One child was excluded because of Autism Spectrum Disorder, the observational measure utilised advises that it should only be used to assess children with no sensory, neurological or genetic disorders.

	Time 1 (n=55)	Time 2 (n=46)
Age	M=39.9 (SD=13.8), months Range= 16-62 months	M=50.1 (SD=13.3) months Range= 22-74 months
Placement	Foster carer no.1 - 94.5%(52) Foster carer no.2 - 1.8%(1) Adoptive family- 3.6%(2)	Foster carer no.1- 63.6%(35) Foster carer no.2- 16.3%(9) Adoptive family- 3.6%(2) Kinship carer- 1.8%(1) Birth parent- 14.5%(8)
Gender	45.5%F (n=25); 54.5%M (n=30)	46.0%F (n=21); 54.0%M (n=25)

Table 1. Demographics of sample

Recruitment Procedures

Recruitment (for the BeST² Trial) took place between December 2011 and April 2013. Each eligible child who entered care due to child protection concerns during this period was considered. Consent from birth parents and foster carers to be approached by the research team to discuss the BeST² Trial was obtained by a social worker recruitment officer who gave potential participants an information leaflet and a video explaining the study. The study's recruitment officer obtained informed consent from those agreeing to be contacted (see Appendix 2.1 for consent form). It was made clear to the carers and birth parents of the eligible participants that participation was entirely voluntary and would not affect any aspect of their care or management. At time of data collection, The BeST² Trial had complete data for approximately N=80 children with a recruitment rate of 58% of eligible families at baseline (T1) and a current retention rate of around 79% of eligible families at 1year follow-up (T2). A researcher in the team attempts to establish reason for drop-out or non-follow up at T2. Most often dropout is due to birth parent(s) being un-contactable at this time point (4.2%) or withdrawing consent (4.1%), or participants being excluded following baseline assessment, for example courts failing to establish grounds for the child being in care (3.6%). For the current study, participants were selected at random from the pool of potential participants using ID numbers. The study sample was found to be representative of the larger pool of data from which it was selected in terms of age, gender, mental health and cognitive functioning [mean age 39.9 and 38.1; gender split 45.5%F 54.5%M and 44.7%F 55.3%M; mean SDQ score 14.0 and 12.93; mean FSIQ 87.5 and 86.7; study sample and larger sample from which it was selected respectively].

The number of participants in the current study was revised due to time constraints however according to the power calculation detailed in the data analysis section, the sample size was adequate. The intent to recruit 100 participants was to increase the chances of detecting rare stable cases of RAD. As this is still considered an important aim, the author and other members of the research team are continuing to collect data for further participants. This is with a view of the author submitting for publication once the target sample is reached (n=100).

The current study is covered by NHS ethical approval (see Appendix 2.2). The researcher was vigilant for any information that highlighted potential risk of harm to a child or others. All data were anonymised and kept confidentially. The time period of data storage is in accordance with University of Glasgow policies and it will only be used for the purposes outlined. Any publications arising from the study will only contain non-identifiable data.

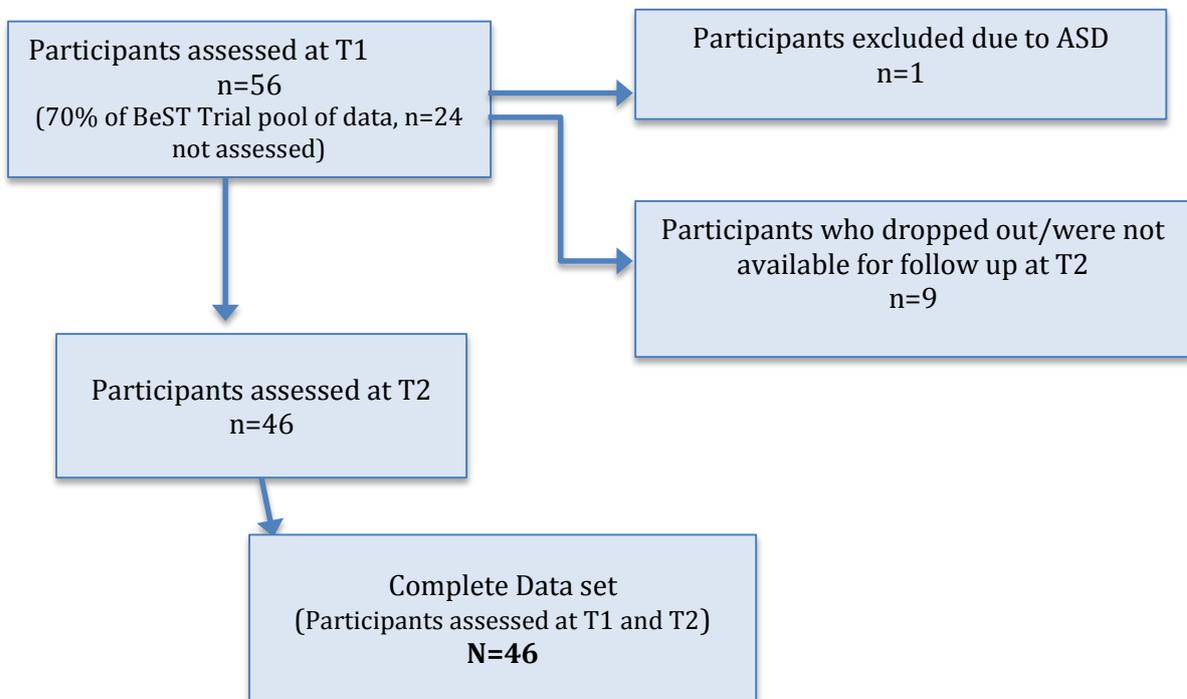


Figure 1. Flow chart of study participants in current study

Baseline assessments were administered approximately one month after the child was placed into care (T1). This time period allowed the child to begin to settle in to the placement and for the child and carer to familiarise with each other. Follow up assessments were completed one year later (T2). At baseline, the assessments were completed with participating children and their foster carers. At follow up, the assessment was completed with the child and their primary caregiver at that time, in most instances this was still a foster carer but in some cases it was a birth parent, adoptive parent, or kinship carer (as indicated in table 1).

Measures

- The Strengths and difficulties questionnaire (SDQ) (Goodman, 1997) was administered at T2. The SDQ is a brief behavioural screening questionnaire for 2-17 year olds. It includes 25 items involving psychological attributes, divided between five scales: emotional symptoms, conduct problems, hyperactivity, peer relationship problems and prosocial behaviour. Subscales can be used individually and the first four subscales are summed to form a Total Difficulties Score. The SDQ has been well validated across a wide age range by various studies (Goodman, 2001).
- A cognitive assessment at T2 was utilised. Children were assessed using the Wechsler Pre-school and Primary Scale of Intelligence (WPPSI IV) (Wechsler, 1967), for use with children between the ages of 30 and 91 months old. The assessment generated Full-scale IQ (FSIQ), Performance IQ and Verbal IQ scores. Scores were not available for one child at T2 due to them being younger than the minimum age for assessment, therefore the test was not administered.
- The Disturbances of Attachment Interview (DAI) (Smyke & Zeanah, 1999) was administered at T1 and T2. The DAI is a semi-structured interview designed to be administered by clinicians to caregivers. This study focused on the emotionally withdrawn/inhibited subscale, these items explore how well the child differentiates among adults, whether the child shows a clear preference for a particular caregiver, seeks comfort from a preferred caregiver, responds to comforting when offered, the degree to which the child responds reciprocally in social interactions and whether the child shows developmentally appropriate levels of emotional regulation. This scale produces scores of 0 to 10, with higher scores reflecting increasing signs of RAD. Smyke, Dumitrescu and Zeanah (2002) stated that the DAI scales demonstrate strong internal validity for RAD (Cronbach's alpha 0.80) and they found inter-rater reliability to be excellent ($\kappa=0.88$).
- The Rating of Inhibited Attachment Behavior Scale (RInAB) (Corval, Baptista, Fachada, Beiramar & Soares, unpublished 2014) (Appendix 2.3) was administered at T1 and T2 using video footage. The RInAB is an observational measure of RAD for preschool aged children developed based on scientific literature, DSM-5 criteria, and repeated observations of interactions of child-caregiver dyads (Corval et al., unpublished 2014). The scale contains 18 items rated between 1 (not at all characteristic) and 5 (very characteristic), grouped into three sub-scales: Attachment behavior, Exploratory behaviour and Socioemotional behaviour. The tool can be used with The Strange Situation Procedure (SSP; Ainsworth & Bell, 1970) and other video material. For the current study some participants (52%) had video recordings of SSP's available, which were used to inform RInAB rating. Formal SSP scoring did not take place, as this was not the focus of this study. The authors of the RInAB are currently investigating its validity (personal communication, May 2016).

Clinical Diagnosis

As in Gleason et al. (2011) the research diagnostic criteria for RAD were applied to create a categorical variable of carer reported RAD whereby at least 3 DAI items must be endorsed. Where observational criteria and carer report criteria were met, cases were discussed with a supervisor of the project (Child Psychiatrist specialising in RAD) and a multi-informant, clinical diagnosis was given. A borderline diagnosis of RAD was given if there was substantial disagreement in observed and carer reported symptoms (i.e. where it was clear that only carer-report criteria or only observational criteria were met) or if from the information available, some elements of RAD remained unclear.

Design

This is a prospective longitudinal cohort study using a within groups design. A correlation and regression approach will be taken to explore associated variables.

Research Procedures

Each video clip was approximately 30-50 minutes in duration and included approximately 25 minutes of the infant playing with their caregiver and around 15 minutes of them having lunch together (provided by the research team). The RInAB scale was administered for each participant using the recordings of child-carer dyads. It was not always possible for the main researcher to be blind to whether the footage being observed was at T1 or T2. A BeST² Trial researcher provided inter-rater reliability by scoring 20% of the sample, this included a range of cases that did and did not meet observational criteria for RAD. A good level of agreement was found (>90%; Kappa=0.8) and any discrepancies were resolved through discussion. Test-retest was carried out with the only child meeting observational criteria at T1 and T1, whereby the RInAB was administered initially and again six months later; consistency was high (97%). Training and supervision of rating was provided by HM and in addition, the main author of the RInAB (Raquel Corval) provided some training and inter-rater reliability where individual scoring was compared and discussed.

Data Analysis

Given that this is the first study in the UK to consider the prevalence and stability of RAD and associated difficulties, it should be considered as an exploratory study that will inform future research. A power calculation was made based on the hypothesis that there would be an association between RAD symptoms and mental health symptoms as indicated by SDQ scores. A previous study (Millward et al., 2006) found a correlation of ($r = 0.84$) between RAD and SDQ scores. Using G*Power (Faul, Erdfelder, Lang & Buchner, 2007) and inputting a large effect size of ($r = 0.5$), setting power at 0.8 and alpha at 0.05, it was calculated that a sample size of 29 was adequate. The distribution of

data was explored through histograms and normal Q-Q plots, on the whole assumptions of normality were not met and non-parametric statistical analyses were utilised. Within group comparisons between symptoms at T1 and T2 were made using Wilcoxon Signed Ranks tests. Mann-Whitney Tests were used to make independent group comparisons between those whose symptoms improved and those who did not. Effect sizes were calculated using the Rosenthal (1994) formula for non-parametric data: $r = Z/\sqrt{N}$ or $r = Z/\sqrt{n_x + n_y}$ when the test was repeated measures; where 0.1, 0.3 and 0.5 indicate small, medium and large effect sizes respectively (Cohen, 1988).

Based on previous research findings, it was anticipated that RAD would be prevalent in a small number of children in the study; thus limiting the validity of statistical comparisons between children with and without RAD. This was founded as at T2, no children met a diagnosis of RAD, meaning categorical comparisons between children with and without RAD would not have been possible. It may have been possible to compare children with some RAD symptoms to children with no RAD symptoms. However, as both scales used to assess RAD do not stipulate descriptive categories (for instance borderline, mild, moderate or severe RAD), this type of comparison was likely to have lacked clinical meaning. In line with the majority of other studies exploring relationships between RAD and other factors (e.g. Elovainio et al., 2015; Gleason et al., 2011; Lehmann et al., 2015; McGoron et al., 2012; Minnis et al., 2007; Moran, unpublished 2014; Zeanah et al., 2004; Zimmerman, unpublished 2015), a correlational approach was taken.

A correlational approach was carried out. A correlation matrix was completed prior to analysis to reduce the number of predictor variables used and enhance power. Factors significantly associated with RAD ($p < .05$) were included in the regression analyses, factors approaching significance were also considered ($p < .09$). Observed and carer reported RAD symptom scores were entered into general linear models as dependent variables with mental health, cognitive functioning and demographic variables as predictors.

A Statistical Package for the Social Sciences (SPSS) version 22.0 was used to investigate the aims and hypotheses. Consultation with the Robertson Centre and NHS GG&C statisticians in regard to statistical analysis and reported findings took place.

Results

	Time 1 (n=55)	Time 2 (n=46)
RInAB scores	Mdn=1.74 (Q1=1.40 Q3=1.89) [Mdn=1.66 (Q1=1.37 Q3=1.84) adjusted for drop out]	Mdn=1.45 (Q1= 1.26 Q3=1.66)
DAI scores	Mdn= 2.0 (Q1= 0.0 Q3=3.0)	Mdn= 1.0 (Q1=0.0 Q3=2.0)
SDQ scores		
Total difficulties		Mdn= 14.0 (Q1= 11.0 Q3=19.0)
Internalising		Mdn= 6.0 (Q1=5.0 Q3=8.0)
Externalising		Mdn= 8.0 (Q1=6.5 Q3=12.0)
Emotional problems	Not collected	Mdn= 1.0 (Q1=0.0 Q3=3.0)
Behavioural problems		Mdn= 3.0 (Q1= 2.0 Q3=5.0)
Hyperactivity		Mdn= 5.00 (Q1= 4.0 Q3=7.0)
Peer problems		Mdn= 4.0 (Q1= 4.0 Q3=6.0)
Prosocial behaviour		Mdn= 2.0 (Q1=0.0 Q3=3.0)
Cognitive functioning		
FSIQ	Not collected	M=87.51 (SD=11.69), 95% CI [84.0-91.0]
Verbal IQ		M=90.86 (SD=11.16), 95% CI [87.5-94.3]
Performance IQ		M=87.49 (SD=12.58), 95% CI [83.6-91.4]

Table 2. Descriptive Statistics

Aim 1: Prevalence and Stability of RAD

It was found that 7.3% (n=4, 95% CI [0.43-14.17]) of participants met observational criteria for RAD and 3.6% (n=2, 95% CI [below 0 - 8.52]) met carer report criteria for RAD at T1. At T2, 2.2% (n=1, 95% CI [below 0- 6.44]) met observational criteria and 2.2% (n=1, 95% CI [below 0 - 6.44]) met carer report criteria for RAD (Figure 2 illustrates these findings). It should be noted that the children meeting observational and carer report diagnostic criteria at T2 were two different participants; however for

the child meeting observational, a carer report measure of RAD was not obtained. For the child meeting carer report diagnostic criteria at T2, the observational score was 1.47 (below the median), they were found to be introverted but marked RAD symptoms were not observed. Only 1.8% of children (n=1, 95% CI [below 0 – 5.31]) met both observational and carer report diagnostic criteria for RAD at T1 and 0% (n=0) at T2. The child who met diagnostic criteria on both measures at T1 no longer met criteria on either measure at T2. Two of the children meeting observational RAD criteria at T1 and one of the children meeting carer report criteria of RAD at T1 dropped out/were not available for follow up at T2.

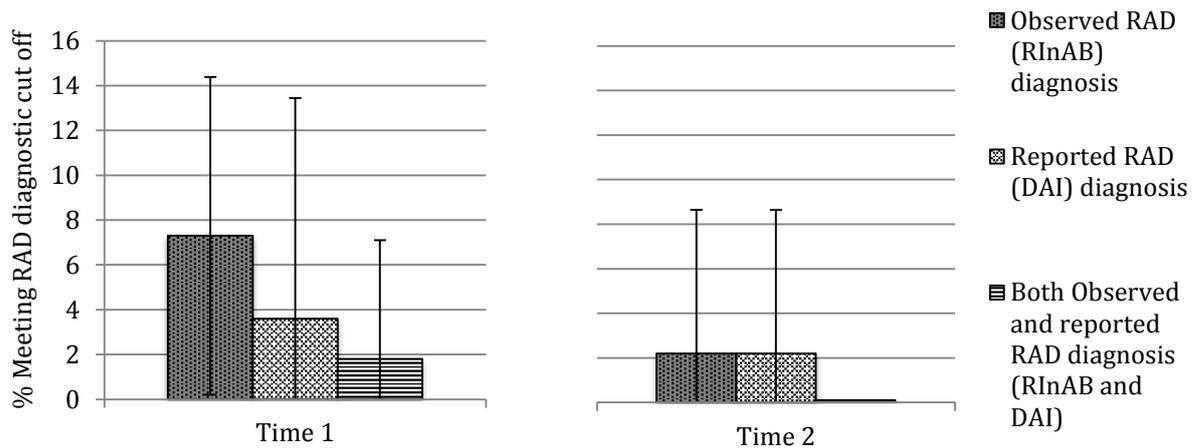


Figure 2. Categorical diagnostic prevalence and stability of RAD

Multimodal, multidisciplinary (Psychiatrist and Trainee Clinical Psychologist) diagnoses were made based on the above information. All children in the study meeting at least one form of diagnostic criteria at either T1 or T2 are featured in Table 3. Based on the clinical diagnoses, RAD was prevalent in 7.3% (n=3, 95% CI [0.43 – 14.17]) at T1 and 4.3% (n=2, 95% CI [below 0 – 10.16]) met a borderline RAD diagnosis at T2.

	Time 1 (n=55)				Time 2 (n=46)			
	RInAB	DAI	Diagnosis	Borderline Diagnosis	RInAB	DAI	Diagnosis	Borderline Diagnosis
Child 1	Y	*	Y	-	<i>Dropped out</i>		-	-
Child 2	Y	N	Y	-	Y	N	-	Y
Child 3	Y	N	Y	-	<i>Dropped out</i>		-	-
Child 4	Y	Y	Y	-	N	N	N	N
Child 5	N	N	N	-	N	Y	-	Y

Table 3. Children meeting multimodal diagnostic criteria at T1 or T2 *denotes missing data

In order to establish the stability of RAD symptoms over one-year, the observed (RInAB) and carer reported (DAI) RAD symptoms were investigated at both time points and compared using Wilcoxon Signed Ranks Test. Data distribution was explored using a histogram and normal QQ plot (included in appendix 2.4), data were observed not to meet parametric assumptions. Observed symptoms of RAD were found to decrease, symptom scores at T2 (Mdn=1.45, Q1=1.27 Q3=1.67, n=46) were significantly lower than symptom scores at T1 (Mdn=1.65, Q1=1.37 Q3=1.83, n=55; $Z(46) = -2.93, p = .003, r = .31$), as illustrated in Figure 3. Carer reported RAD symptoms were not found to be significantly lower at T2 than at T1 (Mdn=2.0, Q1= 0.0 Q3=3.0, n=54 versus Mdn=1.0, Q1=0.0 Q3=2.0, n=45; $p = .194$). This suggests mixed findings in relation to symptoms of RAD significantly improving following one year of being placed in to care, however, there does appear to be a non-significant trend of improving carer reported RAD symptoms (as demonstrated by figure 4). Yet is it also true that the initial level of symptoms was low/not severe (Mdn=2) so there is limited potential for reduction. Furthermore, the test-retest reliability of the observation measure is not known. No significant differences were found in observed and reported RAD symptoms between males and females.

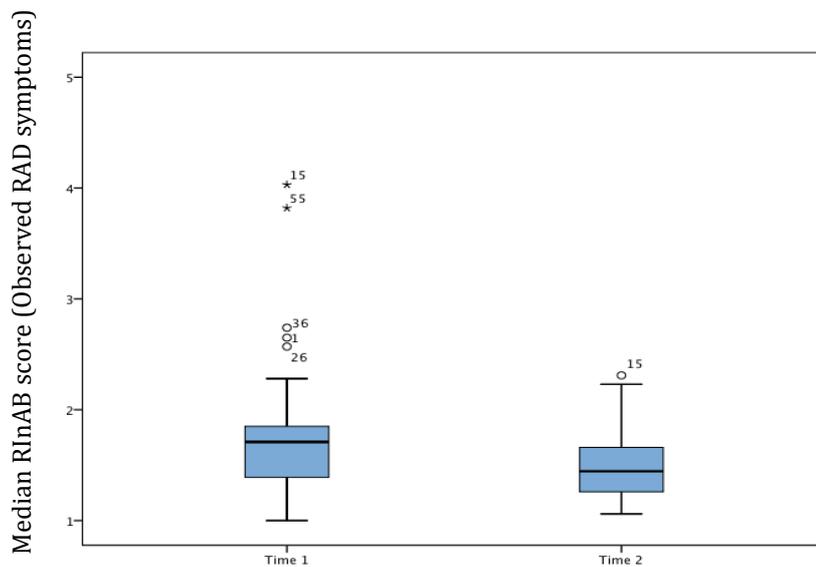


Figure 3. Prevalence and stability of observed RAD symptoms

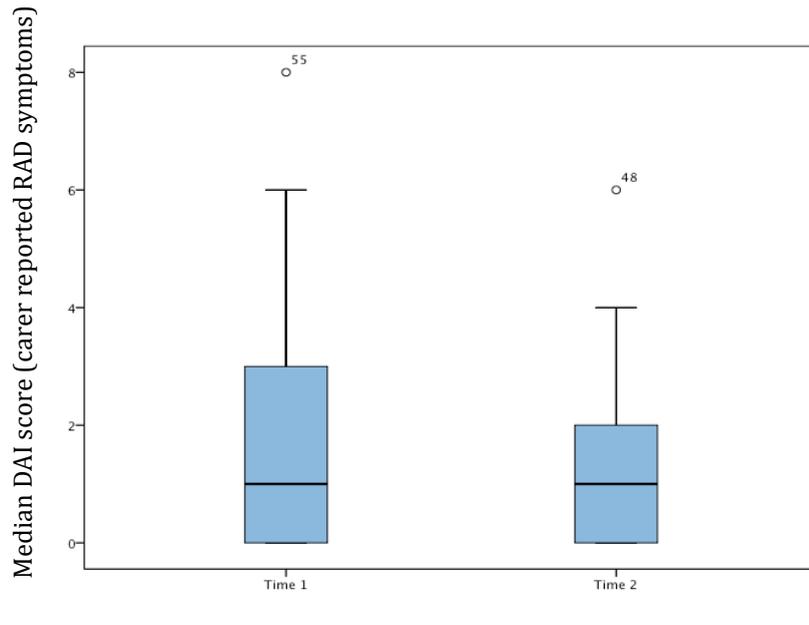


Figure 4. Prevalence and stability of carer reported RAD symptoms

Considering observed symptomatology at T2 individually, although the severity of RAD symptoms was low overall, the results demonstrated that 65.2% (n=30) of children's RAD symptoms had improved and 34.8% (n=16) of participants' symptoms worsened (although not to clinical levels) from T1. In relation to carer reported RAD symptoms at T2, 35.6% (n=16) of children showed an improvement, 33.3% (n=15) remained the same and 31.1% (n=14) of children's symptoms worsened in comparison to T1. As the data did not meet parametric assumptions, Mann-Whitney Tests explored differences between groups. It was indicated that children who improved observationally were significantly younger in age (Mdn=46.0, Q1=37.5, Q3=53.5) than those who did not improve (Mdn=58.0, Q1=48, Q3=67; $z(45)=-2.45$, $p=.014$, $r=.37$). Children who improved according to observational assessment were also found to have significantly higher prosocial behaviour than those who did not improve (Mdn=2.0, Q1=1.0, Q3=4.0 versus Mdn=0.5, Q1=0.0, Q3=2.0; $z(37)=-2.09$, $p=.037$, $r=.34$). No significant differences between observed improvers and non-improvers were found in cognitive functioning ($p=.11$), other mental health difficulties ($p=.49$), placement moves ($p=.21$), category of carer ($p=.52$), or gender ($p=.50$).

According to Spearman's Rho correlations, no significant associations were found between observed RAD symptoms and carer reported RAD symptoms at T1 ($r=.18$, $p=.21$) or T2 ($\rho=.18$, $p=.30$).

An independent samples Mann-Whitney U test demonstrated no notable or statistically significant difference in observed or reported RAD symptoms between children who had returned to birth parents at T2 (n=8) and children who had stayed in placements (n=38) ($p=.75$; $p=.70$), although numbers returning to birth parents were small.

Aim 2: RAD symptoms and associated mental health and cognitive functioning

In order to explore demographic, mental health and cognitive functioning variables that may be associated with symptoms of RAD, correlations were carried out. Data distribution was explored using histograms and normal QQ plots, the data were observed to be nonparametric. Spearman's Rho correlations found significant associations between observed RAD symptoms at T1 and prosocial behaviour ($\rho=.47$, $n=37$, $p=.003$). No significant relationship was found between observed RAD symptoms at T1 and placement moves ($p=.15$), T1 age ($p=.13$), T1 DAI score ($p=.21$), FSIQ ($p=.57$), Verbal IQ ($p=.57$), Performance IQ ($p=.44$), SDQ total difficulties ($p=.92$), internalising problems ($p=.80$), externalising problems ($p=.91$), emotional problems ($p=.47$), behavioural problems ($p=.23$), hyperactivity ($p=.57$), or peer problems ($p=.34$).

Significant associations were found between observed RAD symptoms at T2 and lower FSIQ ($\rho=-.32$, $n=45$, $p=.035$) and Verbal IQ ($\rho=-.34$, $n=44$, $p=.023$). No significant associations were found between observed RAD scores at T2 and age ($p=.67$; $p=.26$), placement moves ($p=.73$), Performance IQ ($p=.34$), SDQ total difficulties ($p=.63$), internalising problems ($p=.10$), externalising problems ($p=.71$), emotional problems ($p=.09$), behavioural problems ($p=.55$), hyperactivity ($p=.31$), peer problems ($p=.37$), and prosocial behaviour ($p=.56$).

Spearman's Rho correlations were used to investigate the relationships between carer reported RAD symptoms and the variables including mental health difficulties and cognitive functioning mentioned above. Carer reported RAD symptoms at T1 were significantly associated with higher levels of prosocial behaviour ($\rho=.36$, $n=37$, $p=.03$) only. At T2, carer reported RAD symptoms were associated with externalising problems ($\rho=.33$, $n=37$, $p=.046$), behavioural problems ($\rho=.46$, $n=37$, $p<.001$) and prosocial behaviour ($\rho=.41$, $n=37$, $p=.01$) only. See Appendix 2.4 for all associations explored, including non-significant findings ($p>.05$).

A multiple linear regression was calculated to predict observed RAD symptoms at T2 based on variables that were found to significantly correlate with RInAB scores (observed symptoms) at T1 and T2. Therefore predictor variables included in the regression analyses were prosocial behaviour, FSIQ and Verbal IQ; SDQ emotional problems was also included as a potential predictor as it was the only other association somewhat approaching significance ($p=.090$). The regression controlled for observed RAD symptoms at T1, although this did not account significantly for any observed variance ($p=.12$). Regression assumptions were met. An analysis of standard residuals was carried out, which showed that the data contained no outliers (Std. Residual Min= -1.56, Std. Residual Max= 2.12). Tests to see if the data met the assumption of collinearity indicated that multicollinearity was not a concern (Tolerance was $>.1$ and VIF was <10 for all variables). The data met the assumption of independent errors (Durbin-Watson value = 1.76). The histogram of standardised residuals indicated that

the data contained approximately normally distributed errors, as did the normal P-P plot of standardised residuals. The scatterplot of standardised residuals showed that the data met the assumptions of homogeneity of variance and linearity. The data also met the assumption of non-zero variances. A significant regression equation was found ($F(5, 30) = 4.59, p = .003$), with an R^2 of .43 (adjusted $R^2 = .34$). Participants' observed RAD symptoms at T2 were found to be significantly predicted by Verbal IQ ($\beta = -.73, p = .023$) but not any of the other included variables (prosocial behaviour $\beta = -.301, p = .132$; emotional problems $\beta = -.083, p = .622$; FSIQ $\beta = .204, p = .486$; and T1 RInAB scores $\beta = -.236, p = .119$).

A further multiple linear regression was calculated to predict carer reported RAD symptoms at T2 based on variables that were significantly associated with DAI scores (carer reported symptoms) at T1 and T2. Predictor variables included in the regression analyses were prosocial behaviour, externalising problems and behavioural problems. As SDQ total difficulties score was approaching significance ($p = .078$), it was also included in the model. The regression controlled for carer reported RAD symptoms at T1, although this did not account significantly for any observed variance ($p = .389$). The steps outlined above were again considered and regression assumptions were met. A significant regression equation was found ($F(30) = 3.94, p = .007$), with an R^2 of .39 (adjusted $R^2 = .29$). Participants' carer reported RAD symptoms at T2 were significantly predicted by prosocial behaviour ($\beta = .479, p = .005$) and not significantly predicted by any other included variables (externalising problems $\beta = -.310, p = .396$; behavioural problems $\beta = .440, p = .086$; SDQ total difficulties $\beta = .162, p = .588$; T1 DAI $\beta = -.140, p = .389$).

Discussion

Prevalence and stability of RAD

It was hypothesised that the level of RAD symptoms would reduce over time but clinical levels of symptoms would remain for some. It was difficult to ascertain if this hypothesis was supported. A larger sample size with fewer dropouts, along with diagnoses informed by fully validated measures would allow for more certainty in regard to the perseverance of RAD. Based on clinical diagnoses, it was found that RAD was prevalent in 7.3% (n=3) at T1 and 4.3% (n=2) met a borderline diagnosis of RAD at T2. Only one child (1.8%) met both observational and carer report diagnostic criteria for RAD at T1 and 0% (n=0) at T2. The child who met diagnostic criteria on both measures at T1 no longer met criteria on either measure at T2. It is important to note however that one child continued to meet observational criteria for RAD at T2. This may begin to address one of the most controversial aspects of the field, whether or not RAD can be persistent, perhaps demonstrating that it can be and is not simply a 'state' associated with current maltreatment. The prevalence of RAD appeared to be very low, even in a high-risk cohort and the confidence limits of the prevalence in the study were very large, suggesting a much larger sample would be needed to ascertain a reliable prevalence of RAD. It is possible that previous studies with relatively small sample sizes finding no persistent cases would have detected persistence with a larger sample size.

Upon further investigation of RAD symptoms at T2, according to observed criteria 65.2% (n=30) showed an improvement and 34.8% (n=16) had worsened; according to carer reported criteria, 35.6% (n=16) improved, 33.3% (n=15) remained stable and 31.1% (n=14) worsened. Children whose observed RAD symptoms had worsened were found to be significantly older at T2; this fits in with the widely accepted concept of early intervention in child maltreatment being more effective. It should be noted that RAD symptoms were fairly infrequent in the current study and although RAD has been found to discriminate from other mental health presentations in children (Minnis et al., 2007), changes could be due to the measure picking up on symptoms that were not specific to RAD. The utilisation of a study design comparing maltreated children with RAD to a control group of maltreated children without RAD symptoms over time would be helpful in addressing this reliably.

Overall, observed RAD symptoms decreased significantly from when children were first placed in to care whereas carer reported symptoms did not change significantly. The findings suggest a discrepancy between observed symptoms of RAD and what carers reported. This may be due to measures lacking sensitivity. In order to explore this further, a much larger sample would be required whereby sensitivity of the RInAB and DAI would be determined against multimodal diagnosis using both measures; ideally also ratified by an experienced clinician. It is possible that at T1, carers did not know the child well enough to be providing an informed rating (the DAI states the carer should know the child well, after one month this may be unlikely) but this does

not explain the continued discrepancy between observed and carer reported symptoms at T2. Given the apparent rareness of the disorder, it is also possible that foster carers lack awareness of symptoms. RAD symptoms have a subtle nature in comparison to conduct problems for instance, which are difficult to ignore. It was noted in the SDQ scales that foster carers consistently rated behavioural problems as higher than emotional problems, perhaps indicating more of an awareness of symptoms that are often easier to recognise.

RAD and associated difficulties

It was hypothesised that symptoms of RAD would be significantly associated with other mental health difficulties and lower cognitive functioning. This hypothesis was partly supported. Carer reported RAD symptoms at T2 were associated with externalising problems ($r=.33$) and behavioural problems ($r=.46$). More prosocial behaviour was associated with observed symptoms of RAD at T1, and carer reported RAD symptoms at T1 and T2 ($r=.47$, $r=.36$, $r=.41$, respectively). Significant correlations were found between observed symptoms at T1 and younger age at T2 ($r=-.32$). At T2, significant associations were found between observed RAD symptoms and lower FSIQ ($r=-.32$) and Verbal IQ ($r=-.34$). A regression showed that verbal IQ was the strongest predictor of observed RAD symptoms at T2, accounting for 18% of variance. Carer reported RAD symptoms at T2 were significantly predicted by prosocial behaviour, with the model accounting for 39% of variance.

The unexpected and perhaps counterintuitive findings in relation to age and prosocial behaviour may be due to the relatively small sample size and small number of children presenting with notable observed or carer reported symptoms of RAD. It may be the case that this would also account for the unexpected non-significant findings in relation to other mental health and cognitive functioning difficulties. The unexpected findings in relation to carer reported symptoms could suggest that the difficulties of these children are going unnoticed by their carers. It was observed that some infants with RAD symptoms were demonstrating hyper-compliance and it might be that carers were mistaking this for prosocial behaviour. The only other known study exploring a relationship between RAD and prosocial behaviour (in a Scottish youth justice population) found a negative association ($R^2 = -.59$) (Moran, unpublished).

Limitations

The current study has a number of limitations. For instance, children may have moved placement between T1 and T2 and a small number of children had returned back to birth parents. Although number of placements or returning to birth parents had no statistically significant impact, it is possible that a larger sample size may have illustrated a difference. In consideration of the assessment of observed RAD symptoms, it was difficult at times to rate items relating to attachment due to children showing no

apparent distress. The authors of the RInAB state that children with RAD fail to demonstrate key attachment behaviours such as seeking or signalling to their caregiver when experiencing distress. Although some participants were subject to a procedure, which is purposefully designed to elicit distress and activate attachment behaviours (their carer leaving them alone in a room; SPP), noticeable distress was often still lacking. Perhaps a lack of noticeable distress is in itself an indicator of RAD, given that signalling distress is a crucial part of forming an attachment with a caregiver. It could be considered a limitation that at T1, children had not been with their carer for more than 6 months as advised by the RInAB authors. However we did not view this as a limitation of the study since the aim was to explore change in RAD symptoms soon after placement in foster care. The DAI also recommends carers know the child well. At T1, children had been with their foster carer for approximately 3 weeks, which may not be long enough for them to know each other well. However, the alternative of waiting longer (e.g. six months) may mean a crucial insight in to maltreated children coming in to care is missed.

The recently developed RInAB scale is the only known observational assessment of RAD. However, the version used is still in preliminary stages and has not yet been fully validated, it may be that the change in observed RAD scores over time is due to lack of test-retest reliability data for the scale rather than an actual change in level of RAD symptoms. In addition, the scale has not been normed on non-maltreated populations. It should be noted that statistical validity may be limited in the current study, as higher RInAB scores do not necessarily indicate higher levels of RAD symptoms given it the utilisation of three critical items. On a methodological level, it was not always possible to be blinded to whether the video footage was T1 or T2, despite attempts.

The study is further statistically limited due to the fairly small sample size. Additionally, there was a relatively low prevalence of RAD symptoms across the sample and only singular cases of children meeting diagnostic criteria. Fairly large confidence intervals demonstrate that for a rare disorder such as RAD, a much larger sample size would be required in order to be confident about prevalence rates and correlates. In terms of the statistical methods, by conducting numerous associations, the risk of a type I error is increased and as with any correlational design, associations rather than causal relationships are elicited. Furthermore, the regression was likely to be underpowered and the fairly low level of variance accounted for suggests other factors that were not investigated may be of importance.

Clinical Implications

If persistent, symptoms of RAD are likely to have profoundly negative effects on children's development as children who are emotionally withdrawn and inhibited are unlikely to elicit the kind of parental support needed for development (Prior & Glaser, 2006). Findings from the current study hopefully begin to provide a greater insight in to the occurrence and correlates of RAD, thus improving awareness of the disorder and

any associated difficulties. It is important that professionals working with children, particularly those who may have been maltreated, are aware of the clinical symptoms and potential correlates of RAD; especially given emotional withdrawal is at the core of the disorder and such children are easily missed.

Future Directions

It is clear from the findings of this study that, it is difficult to draw firm conclusions about RAD and its correlates given its rarity. Larger samples and/or the pooling of samples across studies are required in order to address important questions, as is further validation of the available observational measure. It would be helpful if future research could distinguish risk factors specific to RAD and therefore inform which children are at risk of persistent RAD. This would mean a further step towards developing effective interventions for and ultimately preventing RAD, a disorder with a potentially significant impact on individuals and society.

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APPENDICES

Appendix 1.1

Infant mental Health Journal: Author Guidelines

The *Infant Mental Health Journal* (IMHJ) is the official publication of the World Association for Infant Mental Health (WAIMH) and is copyrighted by the Michigan Association for Infant Mental Health.

Information for Contributors

Reflecting the interdisciplinary nature of the field, its international focus, and its commitment to clinical science, the IMHJ publishes research articles, literature reviews, program descriptions/evaluations, clinical studies, and book reviews on infant social-emotional development, caregiver-infant interactions, and contextual and cultural influences on infant and family development. The Journal is organized into three sections: Research, Clinical Perspectives, and Book Reviews. Research focuses on empirical research. Clinical Perspectives allows for more diversity in types of submissions and is designed to advance infant mental health practice and scholarship. Requests for book reviews should be sent by the author or publisher to the Editor In Chief. Please do not send a copy of the book until the request is approved.

The Journal welcomes a broad perspective and scope of inquiry in infant mental health and has an interdisciplinary and international group of associate editors, consulting editors, and reviewers who participate in the peer review process. In addition to regular submissions to the Journal, proposals for special issues or sections are also welcome. These should be discussed with the Editor In Chief prior to submission.

MANUSCRIPTS for submission to the *Infant Mental Health Journal* should be forwarded to the Editor as follows:

1. Go to your Internet browser (e.g., Netscape, Internet Explorer).
2. Go to the URL <http://mc.manuscriptcentral.com/imhj>
3. Register (if you have not done so already).
4. Go to the Author Center and follow the instructions to submit your paper.
5. Please upload the following as separate documents: the title page (with identifying information) and all remaining files without any identifying information, including the body of your manuscript, and each table and figure. Please note that the cover letter is uploaded directly into a field in the on-line submission platform.
6. The Title Page should include a discussion of any conflicts of interest, human subjects approvals, and funding. Acknowledgements may also appear here. The *Infant Mental Health Journal* complies with all relevant recommendations from the International Committee of Medical Journal Editors in these areas.
7. Your abstract should be uploaded into the appropriate field at the submission website and should also be included in the main text of the manuscript. The abstract in the manuscript must include 3-5 key words listed at the end of the text.
8. Please note that this journal's workflow is double-blinded. Authors must prepare and submit files for the body of the manuscript and any accompanying files that are anonymous for review (containing no name or institutional information that may reveal author identity).
9. All related files will be concatenated automatically into a single .PDF file by the system during upload. This is the file that will be used for review. Please scan your files for viruses before you send them, and keep a copy of what you send in a safe place in case any of the files need to be replaced.
10. Style must conform to that described by the American Psychological Association *Publication Manual*, Sixth Edition, 2009 (American Psychological Association, 750 First Street, N.E., Washington, D.C. 20002-4242). Authors are responsible for final preparation of manuscripts to conform to the APA style.

Manuscripts generally do not exceed 10,000 words and will be assigned for peer review by the Editor or Associate Editor(s) and reviewed by members of the Editorial Board and invited reviewers with special knowledge of the topic addressed in the manuscript. The Editor retains

the right to reject articles that do not meet conventional clinical or scientific ethical standards. Normally, the review process is completed in 3 months. Nearly all manuscripts accepted for publication require some degree of revision. There is no charge for publication of papers in the *Infant Mental Health Journal*. The publisher may levy additional charges for changes in proofs other than correction of printer's errors. Authors have the option to participate in Wiley's OnlineOpen program which allows authors of primary research articles to make their article available to non-subscribers on publication and archive the final version of their article. With OnlineOpen, the author, the author's funding agency, or the author's institution pays a fee to ensure that the article is made available to non-subscribers upon publication via Wiley Online Library, as well as deposited in the funding agency's preferred archive. For more information, please visit the OnlineOpen page.

Proofs will be sent to the corresponding author and must be read carefully because final responsibility for accuracy rests with the author(s). Author(s) must return corrected proofs to the publisher in a timely manner. If the publisher does not receive corrected proofs from the author(s), publication will still proceed as scheduled.

Additional questions with regard to style and submission of manuscripts should be directed to the Editor: Paul Spicer, PhD, at paul.spicer@ou.edu

Appendix 1.2

Table 2. Search Results, conducted January 2016.

Database	Search term	Results	Articles excluded following removal of duplicates and title screening	Articles excluded following abstract screening
Pubmed	“reactive attachment disorder” in title/abstract	n = 79	36 excluded (6 reviews, 3 case studies, 1 animal study, 26 not relevant) (43 remained for abstract review)	26 excluded (Unrelated=10 Case study=6 Not empirical=9 Qualitative=0) Non English=1 17 remained
Web of science	“reactive attachment disorder” in title	n= 66	50 excluded (22 not relevant, 28 duplicates) (16 remained for abstract review)	14 excluded Unrelated=3 Case study=3 Not empirical=4 Qualitative=0 Paper not found=2 2 remained
PsychINFO	“reactive attachment disorder” all fields	n = 166	142 excluded (3 qualitative, 29 not relevant, 30 not empirical study, 18 case studies, 62 duplicates) (24 remained for abstract review)	7 excluded Unrelated=4 Case study=0 Not an empirical study=1 Qualitative=2 17 remained
Ethos	“reactive attachment disorder” in all fields	n = 4	2 excluded (2 not relevant) (2 remained for abstract review)	2 excluded Unrelated= 2 2 remained
Google Scholar	“reactive attachment disorder”	n=1	1 remained for abstract review	1 remained
				Total remaining, n= 39 (n=40 including paper from research conference)

Appendix 1.3

Table 3. Articles included and excluded following full paper screening

<i>Study</i>	<i>Included/Excluded</i>	<i>Reason for Exclusion *</i>	<i>Double Rated</i>
Mizuno et al. (2015)	Excluded	7	
Shimada et al. (2015)	Included	n/a	<input type="checkbox"/>
Jonkman et al. (2014)	included	n/a	<input type="checkbox"/>
Pritchett, Rochat, Tomlinson, Minnis (2013)	Excluded	1	
Lehmann, Havik, Havik, et al. (2013)	Excluded	1	
Giltaij, Sterkenburg and Schuengel (2015)	Included	n/a	
Pritchett et al. (2013)	Excluded	1	
Minnis et al. (2013)	Excluded	1	
Kocovska et al. (2013)	Excluded	2	
Smyke, Zeanah, Gleason, Drury, Fox, Nelson and Guthrie (2012)	Included	n/a	<input type="checkbox"/>
Ayaz et al. (2012)	Excluded	2	
Sadiq et al. (2012)	Excluded	1	
Raaask et al. (2011)	Excluded	1	
Gleason, fox, drury et al. (2011)	Included	n/a	<input type="checkbox"/>
Minnis, Green, O'Connor, Liew, Glaser, et al. (2009)	Excluded	1	
Zeanah, Smyke, Coga and Carlson (2005)	Included	n/a	
Zeanah, Scheeringa, Boris, Heller, Smyke and Trapani et al. (2004)	Included	n/a	<input type="checkbox"/>
Raaska· Elovainio, Sinkkonen, et al. (2013)	Excluded	1	
Minnis, Fleming, and Cooper (2010)	Excluded	1	
Sheaffer, Golden, Bridgers and Hall (2009)	Excluded	1	
Elovainio, Raaska, Sinkkonen, Sanna, Makipaa and Lapinleimu (2015)	Included	n/a	
Woolgar and Baldock (2015)	Excluded	2	
Raaska Elovainio, Sinkkonen, et al. (2015)	Excluded	1	
Raaska, Lapinleimu, Sinkkonen, Salmivalli, Matomaki, Makipaa and Elovainio (2012)	Excluded	1	
Termini, Golden, lyndon, and Sheaffer (2009)	Excluded	1	
Sheaffer (2010)	Included	n/a	
Copp (unpublished, 2012)	Excluded	7	
Marr (unpublished, 2015)	Excluded	7	
Huletz (unpublished, 2012)	Excluded	7	
Thompson (unpublished, 2011)	Excluded	7	
Moorer (unpublished, 2007)	Excluded	7	

Lehmann, Breivik, Heiervang, Havik and Havik (2015)	Included	n/a	
R. Millward , E. Kennedy , K. Towlson and Minnis, H. (2006)	Excluded	1	
Pakdaman (2004)	Excluded	6	
Schraft, C. V., and Franklin, R. (unpublished, 2015)	Excluded	7	
Pérez, Di Gallo, Schmeck, and Schmid (2011)	Excluded	6	
Moran (unpublished thesis, 2014)	Included	n/a	
Coughlin (unpublished thesis, 2011)	Excluded	1	
Minnis, Reekie, Young, O’connor, Ronald, Gray and Plomin (2007) (from citation search)	Included	n/a	<input type="checkbox"/>
McGoron, Gleason, Smyke, Drury, Nelson III, Gregas, Fox, and Zeanah, (2012) (from google scholar search)	Included	n/a	
Zimmerman, A. J (2015) unpublished doctoral thesis (research conference)	Included	n/a	<input type="checkbox"/>

Appendix 1.4

*Reason for Exclusion Key

11. Does not discriminate between inhibited and disinhibited forms of RAD.
12. Unrelated to question; does not focus on Reactive attachment disorder (inhibited type) and associated factors.
13. Case studies
14. Not an empirical study: previous reviews/book chapters/commentaries/letters/conference abstracts
15. Qualitative design
16. Non-English journal, no translation available
17. Unpublished thesis or dissertation with no access/no access

Appendix 1.5

Adapted Quality appraisal checklist – quantitative studies reporting correlations and associations (NICE, 2012)

1.1 Is the source population or source area well described? <ul style="list-style-type: none"> Was the country (e.g. developed or non-developed, type of health care system), setting (primary schools, community centres, etc.), location (urban, rural), population demographics etc. adequately described? 	3 2 0 1 NA
1.2 Is the eligible population or area representative of the source population or area? <ul style="list-style-type: none"> Was the recruitment of individuals, clusters or areas well defined (e.g. advertisement, birth register)? Was the eligible population representative of the source? Were important groups underrepresented? 	3 2 0 1 NA
1.3 Do the selected participants or areas represent the eligible population or area? <ul style="list-style-type: none"> Was the method of selection of participants from the eligible population well described? What % of selected individuals or clusters agreed to participate? Were there any sources of bias? Were the inclusion or exclusion criteria explicit and appropriate? 	3 2 0 1 NA
2.1 Selection of exposure (and comparison) group. How was selection bias minimised? <ul style="list-style-type: none"> How was selection bias minimised? 	3 2 0 1 NA
2.2 Was the selection of explanatory variables based on a sound theoretical basis? <ul style="list-style-type: none"> How sound was the theoretical basis for selecting the explanatory variables? 	3 2 0 1 NA
2.3 Was the contamination acceptably low? <ul style="list-style-type: none"> Did any in the comparison group receive the exposure? If so, was it sufficient to cause important bias? 	3 2 0 1 NA
2.4 How well were likely confounding factors identified and controlled? <ul style="list-style-type: none"> Were there likely to be other confounding factors not considered or appropriately adjusted for? Was this sufficient to cause important bias? 	3 2 0 1 NA
2.5 Is the setting applicable to the UK? <ul style="list-style-type: none"> Did the setting differ significantly from the UK? 	3 2 0 1 NA
3.1 Were the outcome measures and procedures reliable? <ul style="list-style-type: none"> Were outcome measures subjective or objective (e.g. biochemically validated nicotine levels ++ vs self-reported smoking -)? How reliable were outcome measures (e.g. inter- or intra-rater reliability scores)? Was there any indication that measures had been validated (e.g. validated against a gold standard measure or assessed for content validity)? 	3 2 0 1 NA
3.2 Were the outcome measurements complete? <ul style="list-style-type: none"> Were all or most of the study participants who met the defined study outcome definitions likely to have been identified? 	3 2 0 1

	NA
3.3 Were all the important outcomes assessed? <ul style="list-style-type: none"> Were all the important benefits and harms assessed? Was it possible to determine the overall balance of benefits and harms of the intervention versus comparison? 	3 2 0 1 NA
3.4 Was there a similar follow-up time in exposure and comparison groups? <ul style="list-style-type: none"> If groups are followed for different lengths of time, then more events are likely to occur in the group followed-up for longer distorting the comparison. Analyses can be adjusted to allow for differences in length of follow-up (e.g. using person-years). 	3 2 0 1 NA
3.5 Was follow-up time meaningful? <ul style="list-style-type: none"> Was follow-up long enough to assess long-term benefits and harms? Was it too long, e.g. participants lost to follow-up? 	3 2 0 1 NA
4.1 Was the study sufficiently powered to detect an intervention effect (if one exists)? <ul style="list-style-type: none"> A power of 0.8 (i.e. it is likely to see an effect of a given size if one exists, 80% of the time) is the conventionally accepted standard. Is a power calculation presented? If not, what is the expected effect size? Is the sample size adequate? 	3 2 0 1 NA
4.2 Were multiple explanatory variables considered in the analyses? <ul style="list-style-type: none"> Were there sufficient explanatory variables considered in the analysis? 	3 2 0 1 NA
4.3 Were the analytical methods appropriate? <ul style="list-style-type: none"> Were important differences in follow-up time and likely confounders adjusted for? 	3 2 0 1 NA
4.4 Was the precision of association given or calculable? Is association meaningful? <ul style="list-style-type: none"> Were confidence intervals or p values for effect estimates given or possible to calculate? Were CIs wide or were they sufficiently precise to aid decision-making? If precision is lacking, is this because the study is under-powered? 	3 2 0 1 NA

Appendix 1.6

Adapted Scoring of guidance for Quality appraisal checklist – quantitative studies reporting correlations and associations (NICE, 2012)

3	Indicates that for that particular aspect of study design, the study has been designed or conducted in such a way as to minimise the risk of bias.
2	Indicates that either the answer to the checklist question is not clear from the way the study is reported, or that the study may not have addressed all potential sources of bias for that particular aspect of study design.
0	Should be reserved for those aspects of the study design in which significant sources of bias may persist.
1	Should be reserved for those aspects in which the study under review fails to report how they have (or might have) been considered.
(NA)	Should be reserved for those study design aspects that are not applicable given the study design under review (for example, allocation concealment would not be applicable for case-control studies).

Appendix 1.7

Table of Quality appraisal scoring, based on quantitative studies reporting correlations and associations checklist (NICE, 2012)

Items Study	1.1 source	1.2 recruit-ment	1.3 selection	2.1 exposure	2.2 theory	2.3 contamin-ation	2.4 confoun-ding	3.1 measures	3.2 complete	3.3 harm	3.4 follow up	3.5 time	4.1 power	4.2 variables	4.3 methods	4.4 precision	Total Score
Shimada et al., (2015)	3	3	2	NA	3	NA	3	1	2	NA	NA	NA	1	3	3	2	81.8%
Jonkman et al., (2014)	3	3	1	NA	3	2	3	1	2	NA	NA	NA	1	2	3	3	75.0%
Giltaij, et al., (2015)	3	3	3	NA	3	NA	2	1	2	NA	NA	NA	1	2	3	3	78.8%
Zeanah et al., (2005)	3	2	2	NA	3	3	3	2	2	NA	NA	NA	1	3	3	3	83.3%
Gleason et al., (2011)	3	2	3	2	3	2	2	2	2	3	3	3	1	3	3	2	81.3%
Smyke et al., (2012)	3	3	3	3	3	2	2	1	2	3	3	3	1	3	3	3	85.4%
McGoron, et al.,(2012)	3	2	2	NA	3	2	3	2	2	2	3	3	1	2	3	3	80.0%
Zeanah et al., (2004)	3	2	3	NA	3	NA	2	2	2	3	NA	NA	1	2	3	2	77.7%
Elovainio et al., (2015)	3	2	1	NA	3	NA	2	1	2	3	2	2	1	3	3	3	73.8%
Sheaffer (unpub. 2010)	3	3	3	NA	3	NA	2	1	2	NA	NA	NA	1	2	3	2	75.8%
Lehmann et al., (2015)	3	3	2	NA	3	NA	2	1	2	3	NA	NA	1	3	3	2	77.8%
Moran (unpub. 2013)	3	3	3	NA	3	NA	2	1	2	NA	NA	NA	2	3	3	2	81.8%
Minnis et al., (2007)	3	3	2	NA	3	NA	2	1	2	NA	NA	NA	1	3	3	3	75.8%
Zimmerman (unpub. 2015)	3	3	1	NA	3	NA	2	2	2	NA	3	2	1	3	3	2	79.5%

Appendix 2.1 Participant Consent Form



(Form to be on local hospital headed paper)

Title of Project: The Best Services Trial (BeST?)

Patient Identification Number for this trial: (to be obtained at randomisation)

Please make sure you understand everything about the project before you sign the consent form. If you have any questions, please contact **Professor Helen Minnis** on **0141 201 9239**. More information can also be found on the study website http://www.bestservicestrial.org.uk/best_services_trial/home.html

	<i>Please tick as appropriate</i>	
	YES	NO
▶ I have read and understood the information sheet (version 2: 21 Dec 2015) and have had the chance to ask questions.	<input type="checkbox"/>	<input type="checkbox"/>
▶ I understand that I do not have to take part, that I am free to withdraw at any time without giving any reason, and without my child's medical care and legal rights being affected.	<input type="checkbox"/>	<input type="checkbox"/>
▶ I agree to take part in the BeST? Services Trial	<input type="checkbox"/>	<input type="checkbox"/>
▶ I agree to the making of a video of my child's assessment	<input type="checkbox"/>	<input type="checkbox"/>
▶ I agree that the research team can review data stored on me and my child in other parts of health or social services (e.g. GP records)	<input type="checkbox"/>	<input type="checkbox"/>
▶ I agree that my GP can be informed about my family's involvement in the study	<input type="checkbox"/>	<input type="checkbox"/>
▶ I agree that the study sponsor can access my data for monitoring and auditing purposes	<input type="checkbox"/>	<input type="checkbox"/>
▶ I would like to be sent a summary of the study results	<input type="checkbox"/>	<input type="checkbox"/>
▶ I agree I can be contacted for future research studies.	<input type="checkbox"/>	<input type="checkbox"/>
▶ I agree that my videos/data can be used for teaching and training purposes	<input type="checkbox"/>	<input type="checkbox"/>

Name of participant	date	signature
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Name of child	Relationship to child
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Name of Researcher/person taking consent	date	signature
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Appendix 2.2 Letters of ethical approval

see tape

WoSRES
West of Scotland Research Ethics Service

NHS
Greater Glasgow
and Clyde

West of Scotland REC 5
Ground Floor – The Tennent Institute
Western Infirmary
38 Church Street
Glasgow
G11 6NT

Dr Helen Minnis
University of Glasgow
Section of Psychological Medicine
Caledonia House, RHSC Yorkhill
Dalnair Street, Glasgow
G3 8SJ

Date 25 November 2011

Direct line 0141 211 2482
Fax 0141 211 1847
E-mail rose.gallacher@ggc.scot.nhs.uk

Dear Dr Minnis

Study title: Evaluation of the New Orleans Intervention Model for Infant Mental Health in Glasgow

REC reference: 10/S1001/37

This study was given a favourable ethical opinion by the Committee on 26 October 2010.

Research Ethics Committees are required to keep a favourable opinion under review in the light of progress reports and any developments in the study. You should submit a progress report for the study 12 months after the date on which the favourable opinion was given, and then annually thereafter. Our records indicate that a progress report is overdue. It would be appreciated if you could complete and submit the report by no later than one month from the date of this letter.

Guidance on progress reports and a copy of the standard NRES progress report form is available from the National Research Ethics Service website.

The NRES website also provides guidance on declaring the end of the study.

Failure to submit progress reports may lead to the REC reviewing its opinion on the study.

10/S1001/37:	Please quote this number on all correspondence
--------------	--

Yours sincerely

R Gallacher

Mrs Rose Gallacher
Committee Assistant Co-ordinator

Copy to: Dr. Michael Barber, NHSGGC R&D department

Improving better health
nhsggc.org.uk

Dr Helen Minnis
Senior Lecturer in Child Psychiatry
University of Glasgow
Caledonian House,
Yorkhill Hospital,
Glasgow
G3 8SJ

West of Scotland REC 5
Ground Floor - Tennent Building
Western Infirmary
38 Church Street
Glasgow
G11 6NT

Date 03 April 2014
Direct line 0141 211 2102
E-mail WoSREC5@ggc.scot.nhs.uk

Dear Dr Minnis

Study title: Evaluation of the New Orleans Intervention Model for
Infant Mental Health in Glasgow
REC reference: 10/S1001/37
Amendment number: 5
Amendment date: 05 March 2014
IRAS project ID: 46400

The above amendment was reviewed by the Sub-Committee in correspondence.

Ethical opinion

The Sub-Committee were happy to approve the amendment but wished to advise the researchers of two minor points that they may like to consider:

There was concern that the text messages every two months may be intrusive. As a suggestion only, the researchers may want to consider sending a text message at one time point prior to the study appointment.

In the "New Parent Participant Consent Form", it states "In Scotland we routinely collect a range of information..." but it is not clear who "we" is. It suggests that it is the researchers collecting data about people. The researchers may wish to reword this sentence. If this is changed, this would be a minor amendment to be submitted to the Committee.

The members of the Committee taking part in the review gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

Approved documents

The documents reviewed and approved at the meeting were:

Document	Version	Date
New Parent Letter of Invitation	1.0	04 March 2014
Participant Information Sheet: New Parent	1.0	04 March 2014

Appendix 2.3 RInAB rating and scoring sheets

Rating of Inhibited Attachment Behavior – RInAB

Version 3.0

ID: _____

Strange Situation Procedure:

Caregiver-Child Interaction:

Other procedure: _____

1 – Not at all characteristic of this child
 3 – Somewhat characteristic of this child
 5 – Very characteristic of this child
 NA – Not applicable

	1'	2'	3'	4'	5'	NA'
A. Attachment Behavior						
1. When in distress, the child does not search for comfort with the caregiver!	!	!	!	!	!	!
2. When in distress, the child does not respond to the comfort offered by the caregiver!	!	!	!	!	!	!
3. The child shows lack of preference: unconcerned with who is present or interacting with him/her!	!	!	!	!	!	!
4. The child fails to show evidence of heightened arousal on caregiver's departure or reunion!	!	!	!	!	!	!
5. The child's behavior does not tend to elicit care and nurturing behavior from the caregiver!	!	!	!	!	!	!
B. Exploratory Behavior						
1. The child is discomforted with new situations and with the presence of strangers!	!	!	!	!	!	!
2. The quality of play decreases in the presence of the caregiver!	!	!	!	!	!	!
3. The child gives preference and seem to be more comfortable on solitary or parallel play instead of interactive play!	!	!	!	!	!	!
4. The child explores out of synchrony with the caregiver's behavior and availability!	!	!	!	!	!	!
C. Socioemotional Behavior						
1. The child shows withdrawing behaviors (e.g., lack eye contact, no interest on interacting) with the caregiver!	!	!	!	!	!	!
2. The child shows a reduced or absent social and emotional reciprocity (e.g., reduced eye contact, turn taking, social referencing, not sharing excitement)!	!	!	!	!	!	!

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 RInAB & Version 3.0, June 2014

enjoyment) with the caregiver!						
3. The child exhibits hyper-compliance!	!	!	!	!	!	!
4. The child shows false positive affect or discrepant simultaneous behavior (e.g., smiles simultaneously with a tense posture and movements) with the caregiver!	!	!	!	!	!	!
5. The child shows aggressive reactions or irritability!	!	!	!	!	!	!
6. The child shows an apparent misery, sadness, apathy and/or passivity!	!	!	!	!	!	!
7. The child is hypervigilant and/or fearful!	!	!	!	!	!	!
8. The child shows limited positive affect when expected!	!	!	!	!	!	!
9. The child shows unpredictable behavior!	!	!	!	!	!	!

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Raquel Corval, Joana Baptista, Inês Fachada, Ana Beiramar & Isabel Soares!!!
 RInAB & Version 3.0, June 2014!

!

RInAB Scoring sheet

Scoring Procedures

1. For considering the presence of Inhibited Attachment Behavior using a cut-off point, and based on the DSM – 5:

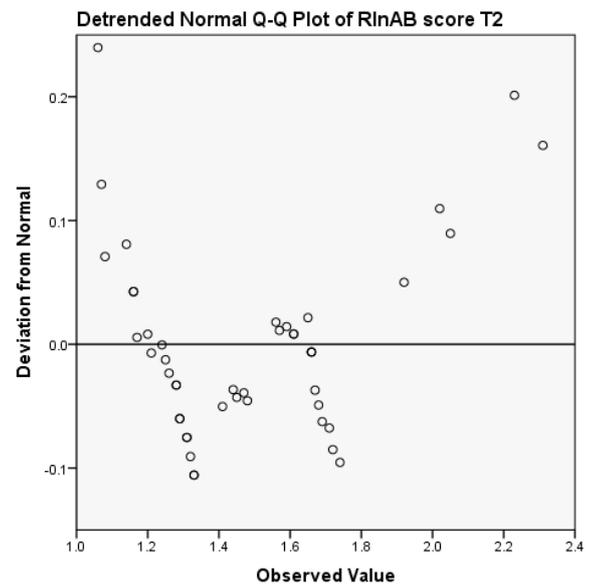
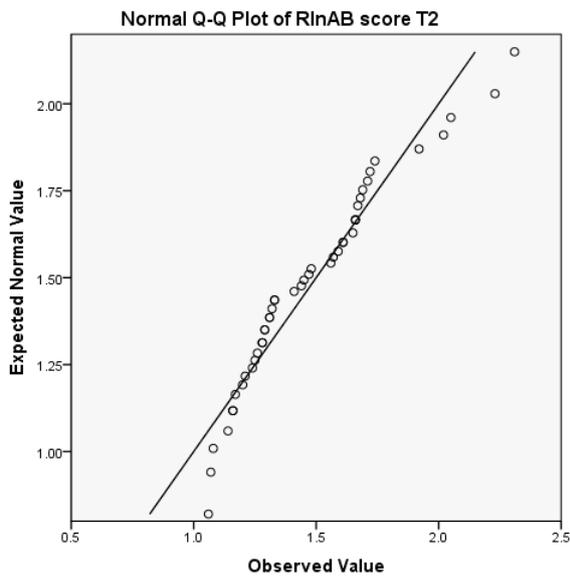
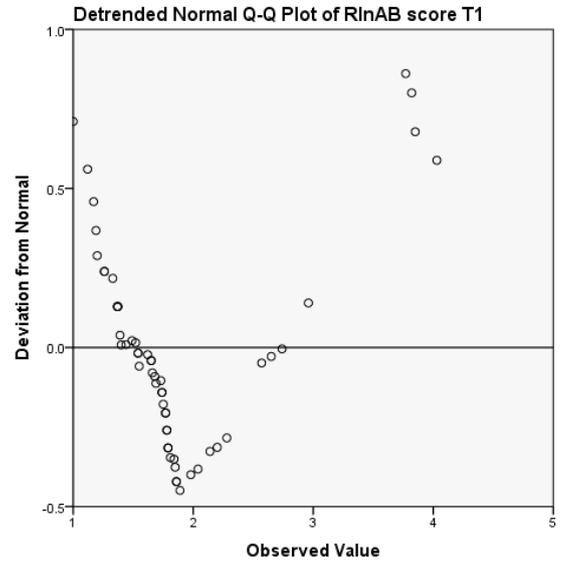
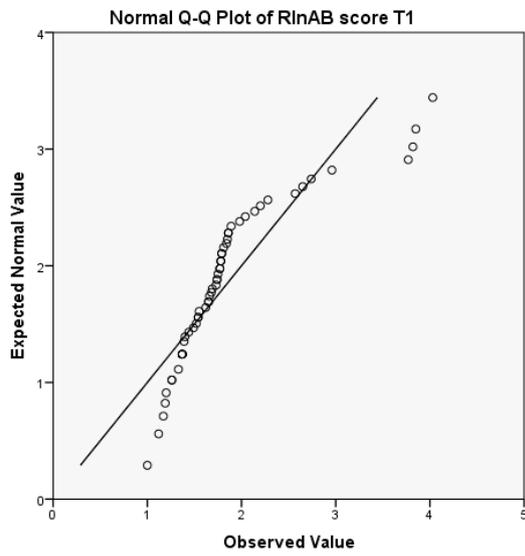
(i) According to Criteria A, given the core feature of the RAD related to attachment, the **items 1 and 2** of the **Attachment Behavior Sub-scale** should have a score of, at least, 3.

(ii) According to Criteria B, **two items** should have a score of, at least, 3 in what concerns to the **Socioemotional Behavior Subscale**.

2. The **final score of RInAB** is the mean of all the scored items (excluding NA), with a higher mean score indicating more inhibited attachment disordered behavior.

A. Attachment Behavior		
1		
2		
3		
4		
5		
Mean		Cut-off point for InAB:
B. Exploratory Behavior		
1		
2		
3		
4		
Mean		
C. Socioemotional Behavior		
1		
2		
3		
4		
5		
6		
7		
8		
9		
Mean		Cut-off point for InAB:
T Mean		Total cut-off point:

Appendix 2.4 QQ plots demonstrating distribution of RAD symptoms at T1 and T2



Appendix 2.5 Nonparametric Spearman's Rho Correlations

		1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18
1. DAI score T2	Correlation Coefficient	1.000	.160	.095	-.079	.051	.027	-.023	.294	.180	.330*	.038	.456**	.048	.159	.405*	.187	.079	.072
	Sig. (2-tailed)	.	.295	.534	.606	.742	.864	.885	.078	.286	.046	.824	.005	.777	.348	.013	.218	.605	.644
	N	45	45	45	45	44	44	43	37	37	37	37	37	37	37	37	45	45	44
2. RInAB score T2	Correlation Coefficient	.160	1.000	.037	-.053	-.316*	-.341*	-.149	-.083	-.272	.064	-.283	-.101	.171	-.152	-.100	-.123	.064	.171
	Sig. (2-tailed)	.295	.	.806	.725	.035	.023	.340	.627	.103	.708	.090	.553	.313	.368	.555	.415	.671	.262
	N	45	46	46	46	45	44	43	37	37	37	37	37	37	37	37	46	46	45
3. RInAB score T1	Correlation Coefficient	.095	.037	1.000	.203	.088	.088	.121	.017	-.042	.019	.123	.178	-.097	-.160	.472**	.177	.212	
	Sig. (2-tailed)	.534	.806	.	.149	.567	.572	.441	.922	.804	.913	.467	.292	.566	.344	.003	.213	.127	n/a
	N	45	46	54	52	45	44	43	37	37	37	37	37	37	37	37	51	53	
4. No of placement moves	Correlation Coefficient	-.079	-.053	.203	1.000	.225	.141	.160	-.250	-.221	-.259	-.051	-.172	-.285	-.172	.057	-.042	-.057	-.106
	Sig. (2-tailed)	.606	.725	.149	.	.138	.362	.307	.135	.190	.121	.764	.308	.087	.308	.739	.772	.686	.478
	N	45	46	52	53	45	44	43	37	37	37	37	37	37	37	37	50	53	47
5. WPPSI fullscale IQ	Correlation Coefficient	.051	-.316*	.088	.225	1.000	.850**	.816**	.095	.066	-.010	.127	.075	-.066	.164	-.037	-.083	.142	.024
	Sig. (2-tailed)	.742	.035	.567	.138	.	.000	.000	.581	.701	.955	.461	.665	.704	.340	.831	.586	.352	.880
	N	44	45	45	45	45	43	43	36	36	36	36	36	36	36	36	45	45	44
6. WPPSI verbal IQ	Correlation Coefficient	.027	-.341*	.088	.141	.850**	1.000	.484**	.125	.162	-.029	.214	.039	-.053	.186	-.253	-.127	-.011	-.077
	Sig. (2-tailed)	.864	.023	.572	.362	.000	.	.001	.461	.339	.863	.202	.820	.754	.270	.132	.410	.942	.622
	N	44	44	44	44	43	44	43	37	37	37	37	37	37	37	37	44	44	43
7. WPPSI performance IQ	Correlation Coefficient	-.023	-.149	.121	.160	.816**	.484**	1.000	.109	-.046	.112	-.055	.120	.061	.189	.172	.074	.152	.038
	Sig. (2-tailed)	.885	.340	.441	.307	.000	.001	.	.526	.789	.515	.751	.486	.726	.270	.316	.637	.329	.809
	N	43	43	43	43	43	43	43	36	36	36	36	36	36	36	36	43	43	42
8. SDQ scores T2	Correlation Coefficient	.294	-.083	.017	-.250	.095	.125	.109	1.000	.752**	.867**	.505**	.705**	.666**	.451**	.186	.104	.087	.135
	Sig. (2-tailed)	.078	.627	.922	.135	.581	.461	.526	.	.000	.000	.001	.000	.000	.005	.270	.539	.609	.434
	N	37	37	37	37	36	37	36	37	37	37	37	37	37	37	37	37	37	36
9. SDQ internalising score	Correlation Coefficient	.180	-.272	-.042	-.221	.066	.162	-.046	.752**	1.000	.378*	.760**	.330*	.225	.520**	.112	.119	.116	.156
	Sig. (2-tailed)	.286	.103	.804	.190	.701	.339	.789	.000	.	.021	.000	.046	.182	.001	.510	.484	.494	.363
	N	37	37	37	37	36	37	36	37	37	37	37	37	37	37	37	37	37	36
10. SDQ externalising score	Correlation Coefficient	.330*	.064	.019	-.259	-.010	-.029	.112	.867**	.378*	1.000	.164	.802**	.779**	.277	.216	.114	-.063	.011
	Sig. (2-tailed)	.046	.708	.913	.121	.955	.863	.515	.000	.021	.	.333	.000	.000	.097	.198	.503	.710	.947
	N	37	37	37	37	36	37	36	37	37	37	37	37	37	37	37	37	37	36

11. SDQ emotional problems	Correlation Coefficient	.038	-.283	.123	-.051	.127	.214	-.055	.505**	.760**	.164	1.000	.125	.094	-.068	.252	.065	-.079	-.044
	Sig. (2-tailed)	.824	.090	.467	.764	.461	.202	.751	.001	.000	.333	.	.460	.582	.690	.132	.704	.643	.798
	N	37	37	37	37	36	37	36	37	37	37	37	37	37	37	37	37	37	36
12. SDQ behavioural problems	Correlation Coefficient	.456**	-.101	.178	-.172	.075	.039	.120	.705**	.330*	.802**	.125	1.000	.286	.224	.320	.289	-.061	-.101
	Sig. (2-tailed)	.005	.553	.292	.308	.665	.820	.486	.000	.046	.000	.460	.	.086	.183	.053	.083	.718	.557
	N	37	37	37	37	36	37	36	37	37	37	37	37	37	37	37	37	37	36
13. SDQ hyperactivity	Correlation Coefficient	.048	.171	-.097	-.285	-.066	-.053	.061	.666**	.225	.779**	.094	.286	1.000	.197	.059	-.159	.024	.134
	Sig. (2-tailed)	.777	.313	.566	.087	.704	.754	.726	.000	.182	.000	.582	.086	.	.243	.729	.346	.886	.435
	N	37	37	37	37	36	37	36	37	37	37	37	37	37	37	37	37	37	36
14. SDQ peer problems	Correlation Coefficient	.159	-.152	-.160	-.172	.164	.186	.189	.451**	.520**	.277	-.068	.224	.197	1.000	-.221	.074	.311	.300
	Sig. (2-tailed)	.348	.368	.344	.308	.340	.270	.270	.005	.001	.097	.690	.183	.243	.	.189	.663	.061	.076
	N	37	37	37	37	36	37	36	37	37	37	37	37	37	37	37	37	37	36
15. SDQ prosocial behaviour	Correlation Coefficient	.405*	-.100	.472**	.057	-.037	-.253	.172	.186	.112	.216	.252	.320	.059	-.221	1.000	.357	-.085	-.174
	Sig. (2-tailed)	.013	.555	.003	.739	.831	.132	.316	.270	.510	.198	.132	.053	.729	.189	.	.030	.617	.309
	N	37	37	37	37	36	37	36	37	37	37	37	37	37	37	37	37	37	36
16. DAI T1	Correlation Coefficient	.187	-.123	.177	-.042	-.083	-.127	.074	.104	.119	.114	.065	.289	-.159	.074	.357	n/a	-.151	-.147
	Sig. (2-tailed)	.218	.415	.213	.772	.586	.410	.637	.539	.484	.503	.704	.083	.346	.663	.030	.	.295	.324
	N	45	46	51	50	45	44	43	37	37	37	37	37	37	37	37		50	47
17. Age T1	Correlation Coefficient	.079	.064	.212	-.057	.142	-.011	.152	.087	.116	-.063	-.079	-.061	.024	.311	-.085	-.151	1.000	.905**
	Sig. (2-tailed)	.605	.671	.127	.686	.352	.942	.329	.609	.494	.710	.643	.718	.886	.061	.617	.295	.	.000
	N	45	46	53	53	45	44	43	37	37	37	37	37	37	37	50	53		47
18. Age T2	Correlation Coefficient	.072	.171	n/a	-.106	.024	-.077	.038	.135	.156	.011	-.044	-.101	.134	.300	-.174	n/a	.905**	1.000
	Sig. (2-tailed)	.644	.262		.478	.880	.622	.809	.434	.363	.947	.798	.557	.435	.076	.309		.000	.
	N	44	45		47	44	43	42	36	36	36	36	36	36	36	36		47	47

* Correlation is significant at the 0/05 level (2-tailed) ** Correlation is significant at the 0.01 level (2-tailed)

Appendix 2.6 Major Research Project Proposal

Prevalence and Stability of Inhibited Reactive Attachment Disorder Symptoms in Maltreated Infants and Associated Mental Health and Cognitive Functioning Difficulties

Abstract

Reactive attachment disorder has been described as one of the least researched and most poorly understood mental disorders (Chaffin *et al.*, 2006). There is very little research in to inhibited reactive attachment disorder (I-RAD) specifically but given what is known about attachment, it is likely that I-RAD has profound consequences for child development. Very little is known about the stability of I-RAD symptoms over time. This study will explore the stability of I-RAD symptoms in a maltreated infant sample and investigate associations between symptoms of I-RAD and other mental health problems and cognitive functioning. The sample has been recruited for the on going Best Services Trial. Assessments were administered one month after a child became accommodated into care and again at a one-year follow up. Findings from the study will add to the developing understanding of I-RAD symptoms and associated difficulties.

Introduction

Attachment is a fundamental instinct across species, whereby to protect itself from threat an infant instinctively seeks to be close to its caregiver when distressed (Bowlby, 1969). In humans, the formation of secure attachment relationships allows for positive social development and emotional regulation, which protects against mental health problems (Prior & Glaser, 2006). Children with an attachment disorder such as Inhibited Reactive Attachment Disorder do not appear to demonstrate important attachment behaviours such as seeking or signalling to their caregiver when experiencing distress. There appears to be a deactivation of the attachment system in that these children do not seek and accept comfort or signal distress when frightened or hurt (Prior & Glaser, 2006). Furthermore, children with I-RAD may be socially and emotionally withdrawn in a wide range of situations, which is likely to prevent them from making use of love or care from others and limiting their opportunities for learning; therefore, having a considerable impact on the development of these children (Prior & Glaser, 2006).

Reactive attachment disorder was first defined in 1980 and has been revised several times since (Zeanah & Gleason, 2010), it has been described as one of the least researched and most poorly understood disorders listed in the DSM (Chaffin *et al.*, 2006). There was previously two forms of the disorder as defined by the DSM-IV and DSM-IV-TR (APA, 1994; 2000), these being 'inhibited reactive attachment disorder (I-RAD)' and 'disinhibited reactive attachment disorder (D-RAD)'. However, the where as the DSM-V (APA, 2013) has more recently updated the classification of I-RAD to simply 'Reactive Attachment Disorder' and changed the classification of disinhibited reactive attachment disorder to 'social engagement disorder'. The DSM-V, defines reactive attachment disorder (or I-RAD) as:

- A consistent pattern of inhibited, emotionally withdrawn behaviour toward adult caregivers.
- Persistent social and emotional disturbance.
- The child has experienced patterns of extremes of insufficient care.

The international classification of diseases 10th edition (ICD-10; WHO, 1992) details that children with reactive attachment disorder (or I-RAD) may exhibit misery, huddling, clinginess, an inappropriate lack of response, or aggression. For the purposes of this proposal, given the recent transition of terms, Inhibited Reactive Attachment Disorder (I-RAD) will continue to be used as opposed to the newer DSM-V term of 'Reactive Attachment Disorder'. It is hoped that this will avoid confusion with disinhibited reactive attachment disorder, now referred to as 'social engagement disorder (DSM-V), due to stark differences in their clinical presentation.

Researchers are fairly confident about the prevalence of disinhibited reactive attachment disorder (D-RAD); however the prevalence of I-RAD is less known but appears to be a rarer disorder (Corval, Baptista, Fachada, Beiramar & Soares, 2014; Gleason et al., 2011). The DSM-V (APA, 2013) states that less than 10% of children who have been severely neglected develop I-RAD (and about 20% develop D-RAD).

I-RAD is considered to be more common within children with an experience of institutionalization (Corval, Baptista, Fachada, Beiramar & Soares, 2014). Studies exploring RAD in children in care (Millward, Kennedy, Towlson & Minnis, 2006; Minnis, Everett, Polosi Dunn & Knapp, 2006) found that the sample scored higher on measures of RAD when compared to children not in care; however these studies were not specific to the inhibited form of reactive attachment disorder. In a deprived population, a UK based study found that RAD (D-RAD, I-RAD or mixed disorder) had a prevalence of 1.4% (Minnis *et al.*, 2013). Other studies have reported variable prevalence rates: as cited in Corval et al., (2014), studies involving children with an experience of pathogenic care have found prevalence of I-RAD to vary between 0% and 35% (Boris, Hinshaw-Fuselier, Smyke, Scheeringa, Heller & Zeanah, 2004; Egger Erkanli, Keeler, Potts, Walter & Angold 2006; Zeanah, Scheeringa, Boris, Heller, Smyke & Trapani, 2004). A study involving a youth justice population found from a sample of 29 found that over half of adolescents met the criteria for a RAD or Borderline RAD diagnosis, with 10% having I-RAD (Moran, unpublished doctoral thesis). In conducting the Bucharest Early Intervention Project with previously institutionalised Romanian children, Gleason et al., (2011) found that those meeting a diagnostic criteria for I-RAD varied at each time point (4.6% at baseline, 3.3% at 30 months, 1.6% at 42 months and 4.1% at 54 months).

Some researchers argue that further clarity around the definition of RAD is needed. Zeanah and Gleason (2010) propose that the symptoms of RAD are signs of current maltreatment rather than signs of an attachment disorder. Zeanah, Mammen and Lieberman (1993) assert that the frozen watchfulness associated with the inhibited nature of I-RAD, is in fact a response when confronted by an abusive caregiver rather than an expressed sign of attachment disorder. In order to be a true disorder, I-RAD would need to be pervasive across different situations. If I-RAD were simply a "state" associated with current maltreatment, then it may be expected to disappear once a child is placed in a stable, nurturing foster family.

Historically, there has been some uncertainty around the reliability of diagnosing mental health difficulties in very young children, however, Stovgarrd, Houmann, Christiansen, Landorph,

Jørgensen, Olsen, Heering, Kaas-Nielsen, Samberg, and Lichtenberg (2007) carried out a study exploring psychopathology in infants. The study found that prevalence and distribution of mental health problems in 18-month-old children seemed to correspond to the distributions among older children. Until recently, it has been difficult to investigate the presence of I-RAD due to limited measures for informing a diagnosis specific to I-RAD. However, a new tool has recently been developed (The Rating of Inhibited Attachment Behavior; RInAB) (Corval, Baptista, Fachada, Beiramar & Soares, unpublished manuscript, 2014).

There is limited research exploring the mental health of children with I-RAD, however behaviours indicative of attachment disorders have been shown to be distinct from conduct problems, emotional problems and hyperactivity (Minnis, Reekie, Young, O'connor, Ronald, Gray & Plomin, 2007). Yet, given the link between early childhood psychopathology and difficulties in parent-child relationships (Stovgarrd et al, 2007), it is likely that children with symptoms of I-RAD would have a higher likelihood of experiencing mental health difficulties. Millward et al (2006) found a significant correlation ($r = 0.84$) between reactive attachment disorder (RAD) and other mental health symptoms. However, this study was not specifically exploring inhibited reactive attachment disorder. Minnis et al. (2009) found that a RAD sample had significantly more mental health symptoms as indicated by lower SDQ total difficulties scores in comparison to children without RAD, likewise this did not distinctly consider I-RAD. It would be useful to explore an association with mental health difficulties investigating I-RAD symptoms specifically.

Pritchett et al., (2013b) found that of those meeting the diagnostic criteria for I-RAD or D-RAD over 85% were identified as having a comorbid diagnosis, including a likely diagnosis of ADHD (52%), PTSD (19%), and specific phobia (14.3%). However, it is likely that some of these comorbidities (e.g. ADHD) are more associated with the disinhibited form of RAD. Again, it would be useful to explore if similar associations are identified when focussing solely on I-RAD. Moran (2014, unpublished doctoral thesis) explored both types of RAD in a youth justice population (12-17 years) and found a strong association between I-RAD and other mental health symptoms (accounting for 36% of variance with a large affect size, $r_s=.6$). Looking particularly at I-RAD in previously institutionalised Romanian children, Gleason et al., (2011) found that symptoms were associated with higher levels of depressive symptoms.

Research has found both forms of RAD (Pritchett, et al., 2013b) and specifically I-RAD (Gleason et al., 2011) to be associated with below average intellectual functioning. Furthermore, studies have demonstrated that both forms of RAD are particularly associated with language difficulties (Minnis et al., 2009; Sadiq, Slate, Skuse, Law, Gillberg & Minnis, 2012). It would be useful to investigate this further in a UK based maltreated sample.

In summary, very little is known about the prevalence and stability of I-RAD symptoms in maltreated infants coming into foster care, and virtually nothing about the associations between I-RAD, other mental health problems and cognitive functioning. This study has a unique opportunity to investigate maltreated children over a one year period and address some of these gaps.

Aims and hypotheses

Aims

- To establish the stability of I-RAD symptoms in a maltreated sample, comparing symptoms shortly after placement in foster care (time 1) and to level of symptoms after one year of being placed in foster care (Time 2).
- To establish the association between symptoms of I-RAD and mental health difficulties and cognitive functioning.

Hypotheses

- It is hypothesised that the level of I-RAD symptoms will reduce somewhat over time but clinical levels of symptoms will remain for some.
- It is hypothesised that symptoms of I-RAD will be significantly associated with other mental health difficulties and lower cognitive functioning.

Plan of Investigation

Participants

The sample will consist of approximately 100 children aged between 12 and 60 months who have been accommodated into care in the city of Glasgow and are involved in an RCT of an infant mental health intervention (the BeST? Services Trial).

Methods/Design

Inclusion and Exclusion Criteria

Inclusion/exclusion criteria from original study (BEST Services Trial; Pritchett et al., 2013a) from which data is being used.

All parents (or recognised parental guardians) with a child aged between 6 and 60 months who come into a period of care due to child protection concerns are invited to take part in the study. Children are excluded from the study if:

- they have a profound learning disability (as assessment outcome measures would not be appropriate), and/or
- their primary caregiver is unavailable to take part in the intervention (such as long-term imprisonment, death, or being uncontactable by services or research team for 3 months or more).

Additional exclusion criteria for the proposed study:

- Children under 12 months old as “typical development, selective attachment behaviours develop between 6 and 9 months (Schofield & Beek, 2006).
- Children who have not been followed up at T2.

Recruitment Procedures

The sample has already been recruited; videotape footage is stored on a hard drive in Yorkhill Royal Hospital for Sick Children.

Information on the original study (Pritchett al., 2013a):

Recruitment was between December 2011 and April 2013. Each eligible child who entered care due to maltreatment during this period was considered. Consent from parents and foster carers to be approached by the research team to discuss the study was obtained by the social worker who gave potential participants an information leaflet and a video explaining the study. Informed consent from those agreeing to be contacted was obtained by the study's recruitment officer.

Measures

Baseline assessment were administered at a minimum of one month after the child was received into care to allow the child and carer to get to know each other and for the child to settle into the carer's home. Follow up assessment was completed one year later. At baseline, the assessment was completed with all children and their foster carers. At follow up, the assessment was completed with the child's primary caregiver at that time who may have been the birth parent, adoptive parent, or a foster carer.

Measures already collected include:

- The Strengths and difficulties questionnaire (SDQ; Goodman, 1997) was administered at T1 and T2. The Strengths and Difficulties Questionnaire (SDQ) is a brief behavioural screening questionnaire about 2-17 year olds. It includes 25 items psychological attributes, divided between five scales: emotional symptoms, conduct problems, hyperactivity/inattention, peer relationship problems, prosocial behaviour. The SDQ has been well validated across a wide age range by various studies (Goodman, 2001).
- The Early Years Development and wellbeing assessment (DAWBA, Goodman, Ford, Richards, Gatward & Meltzer, 2000) was administered at T2. The DAWBA was completed by carers and is used to generate International Classification of Diseases (ICD) and Diagnostic and Statistical Manual of Mental Disorders (DSM) codes. It has been validated by several studies (e.g. Moya et al., 2005).
- A cognitive assessment of the child has been undertaken at T1 and T2. Children under 2.5 years were assessed with the Bayley Scales of Infant Development (Emmy & Bayley, 1966), while children 2.5 years and over were assessed using the Wechsler Pre-school and Primary Scale of Intelligence (WPPSI IV) (Wechsler, 1967).
- The Disturbances of Attachment Interview (DAI; Smyke & Zeanah, 1999) was administered for those who were likely to meet a diagnosis of RAD based on the other measures. The DAI is a semi-structured interview designed to be administered by clinicians to caregivers. Smyke, Dumitrescu and Zeanah (2002) stated that the DAI scales to demonstrate strong internal validity for both types of RAD (Cronbach's alpha

0.83 for D-RAD and 0.80 for I-RAD). Furthermore, inter-rater reliability for the DAI was demonstrated to be excellent ($\kappa=0.88$).

Measures that will be administered for the study in hand are as follows:

- The Rating of Inhibited Attachment Behavior (RInAB) Scale will be administered to rate videos. This is a scale recently developed by Corval, Baptista, Fachada, Beiramar and Soares (unpublished manuscript, 2014). The authors state that the RInAB was developed based on a scientific literature review, DSM-5 criteria for RAD, and repeated observations of interactions of child-caregiver dyads. The RInAB aims to assess the behaviors considered more relevant to identify the inhibited attachment in the preschool years. This scale contains 18 items rated between 1 (not at all characteristic) and 5 (very characteristic) and grouped in three sub-scales: attachment behavior, exploratory behaviour and Socioemotional Behaviour (Corval, Baptista, Fachada, Beiramar & Soares, unpublished manuscript, 2014). The authors are currently investigating the tool's validity.

Design

This is a prospective longitudinal cohort study using a within groups design. A correlation/regression approach will be taken to explore associated variables.

Research Procedures

Video clips taken at T1 and T2 will be rated. Each clip is approximately 10-15 minutes in duration and includes approximately 5 minutes of the infant playing and approximately 5 minutes of the infant having lunch (provided by the research team).

Existing data gathered from interviews and the video clips will be accessed from the York Hill research office (stored on a hard drive).

The RInAB scale will be administered for each participant using the video footage, it is expected that this will take around 20 minutes. Video footage will be analysed at T1 and T2. The main researcher will be blind to whether the footage is showing T1 or T2. A BeST[?] Trial administrator will order and label each clip using a computer-generated random number sequence so that half of the sample are rated at T1 first and the other half are rated at T2 first. A BeST[?] Research assistant will provide inter-rater reliability by scoring 20% of the sample. Training supervised by HM on video rating according to the RInAB will be received which will include an experienced research assistant rating five of the same video clips and individual scoring being compared and discussed to ensure rater reliability.

Data Analysis

Aim 1.

In order to explore the stability of I-RAD symptoms over one year, the mean RInAB, Waiting Room Observation for RAD and DAI scores at both time points will be compared using a one-way analysis of variance (ANOVA). This will establish if there is a significant difference in I-RAD symptoms at time 1 and time 2.

Aim 2.

In order to explore variables that may be associated with symptoms of I-RAD, correlations will be carried out. At T2, the level of I-RAD symptoms will be correlated with other mental health difficulties (as defined by SDQ scores); and level of cognitive functioning (WPPSI IV scores, including a break down of separate domains including verbal comprehension). Correlations will be provided using either Pearson's product-moment correlation or Spearman's rank order correlation depending on the distribution of the data. Where correlations exist, a regression analysis will be used to identify factors that predict I-RAD at T2. Potential confounders, such as child age, will be considered.

A Statistical Package for the Social Sciences (SPSS) version 22.0 will be used to investigate the aims and hypotheses.

Justification of sample size

In exploring the stability of I-RAD symptoms, using the statistical programme G*Power (Faul, Erdfelder, Lang and Buchner, 2007), a sample size of 100, a power of 0.8 and a significance level of 0.05, it was demonstrated that the study has the power to identify a small effect size of 0.25.

A power calculation was made based on the hypothesis that there will be an association between RAD symptoms and mental health symptoms as indicated by the Strengths and Difficulties Questionnaire (SDQ) scores. A previous study (Millward, Kennedy, Towlson & Minnis, 2006) found a correlation of ($r = 0.84$) between RAD and SDQ scores. Using G*Power (Faul, Erdfelder, Lang and Buchner, 2007) and inputting a large effect size of ($r = 0.5$), setting power at 0.8 and alpha at 0.05, it was calculated that a sample size of 29 was adequate.

A power calculation was made based on the hypothesis that mental health symptoms level of cognitive functioning will predict I-RAD symptoms. Using G*Power (Faul, Erdfelder, Lang and Buchner, 2007), estimating a medium effect (0.15), a power of 0.8 and alpha at 0.05, it was calculated that a sample size of 68 was adequate.

Health and Safety Issues

Please refer to the Health and Safety form for more information (Appendix 1).

Researcher Safety Issues

Researched safety issues will be minimal. Data has been collected and video footage will be analysed in an office within Yorkhill Hospital.

Participant Safety Issues

There are minimal participant safety issues. Ethical guidelines were followed when obtaining data. Informed consent was sought from relevant parties for the participants involved and they were made aware they could withdraw any time.

Please refer to the Health and Safety form for more information (Appendix 1).

Ethical Issues (including where submissions will be made)

As discussed with Prof Helen Minnis and Bridie Fitzpatrick (Trial Manager), the project will not require an ethics amendment as it is covered by current ethical approval (approved by: NHS West of Scotland Research Committee number 5, 26/10/2010). However, an IRAS form will be completed and submitted for my academic file.

It has been made clear to the carers of the participants that participation was entirely voluntary and will not affect any aspect of their care or management.

The researcher will report any information observed that highlights risk to the young person or others. All data will be anonymous and kept confidential, it will be stored on an NHS or password protected computer. The time period of data storage will be in accordance with NHS GG&C policies and the confidentiality and use of participant data will be determined by the Data Protection Act 1998, it will only be used for the purposes outlined. Any publications arising from the study will only contain non-identifiable data.

Timetable

Jan 2015-	submit research proposal
Mid 2015-	begin data analysis
Late 2015-	complete data analysis
Early 2016-	begin write up of systematic review and thesis
Spring 2016-	systematic review submission
July 2016-	thesis submission

Practical Applications

If persistent, symptoms of I-RAD are likely to have profoundly negative effects on children's development as children who are emotionally withdrawn and inhibited are unlikely to elicit the kind of parental support needed for development (Prior & Glaser, 2006). Findings from the study in hand will hopefully provide a greater insight in to the occurrence and correlates of I-RAD, thus improving awareness of the disorder and any associated mental health difficulties. This would be pertinent for professionals working with children, particularly those children who may have been maltreated and given emotional withdrawal is at the core of the disorder, this is particularly important as such children are easily missed.

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