Scotland's Hikikomori: Estimating Patterns of Extreme Social Withdrawal in Young People Following COVID19

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Chapter 1

A systematic review of biopsychosocial factors relevant to understanding and treating Hikikomori (extreme social withdrawal)

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Abstract

Background: Reported cases of people with Hikikomori (extreme social withdrawal) are on the increase throughout the world. This systematic review identified biopsychosocial factors that increased the risk of people developing and/or experiencing Hikikomori. Understanding biopsychosocial factors relevant to understanding Hikikomori should enable improved treatment development.

Aims: This review aims to investigate biopsychosocial factors associated with Hikikomori.

Method: This review provides a narrative synthesis of the literature. A search of five relevant electronic databases was completed following the PRISMA statement for reporting systematic reviews. Twelve studies met the inclusion criteria out of 1,471 records initially identified. Forward and backward searches were also undertaken with no additional papers identified. The methodology of each of the 12 papers were rated for quality using the AXIS tool.

Results: Twenty-one biopsychosocial factors were reported to be associated with Hikikomori behaviours across the twelve eligible studies. These included anxiety, coping skills, family psychiatric history and attachment. The biopsychosocial factors were grouped into four categories; Disorders, Interpersonal Dynamics, Psychological Capital, and Negative Experiences.

Conclusion: This review systematically identified and synthesised the current research looking at links between biopsychosocial factors and Hikikomori behaviours. The twelve included studies delineated twenty-one biopsychosocial factors associated with Hikikomori that could possibly help inform improved treatment and support for people who are withdrawing or on the path of withdrawing to the extreme level of Hikikomori.

Key words: Hikikomori, biopsychosocial factors, social withdrawal,

Introduction

Social withdrawal is defined as “voluntary isolation prolonged in time that involves the cessation of any form of social relationship and contact with people and the outside” (Morese, R. et al., 2020, p.1). The Japanese term “Hikikomori” is used to describe an extreme form of social withdrawal lasting for more than six months. Onset of Hikikomori happens predominantly in late adolescence, and disproportionately affects males (Teo, 2009). People with Hikikomori not only retreat into their bedrooms within the family home, but they also withdraw from family contact (Teo, 2009).

Hikikomori entered public awareness in the 1990s (Saitō, 2013) and is considered a relatively new condition but has been described as a “silent epidemic” (Teo, 2009). While originally recognised and characterised in Japan it is becoming a worldwide issue (Malagón-Amor et al., 2014). However, it is not considered an official psychiatric diagnosis in the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) and International Classification of Diseases (ICD-11) (Teo, 2009). In 2010, Japan's Ministry of Health, Labour and Welfare released management guidelines and a definition for Hikikomori. They defined Hikikomori as: “a situation where a person without psychosis is withdrawn into his/her home for more than six months and does not participate in society such as attending school and/or work” (Kato et al., 2018., p. 106). It is thought that 2.2% of the Japanese population aged between 20 and 64 years, have lifetime Hikikomori, suggesting increased prevalence (Nishi et al., 2019).

Current evidence on how to prevent or remediate Hikikomori is sparce. The effectiveness of psychosocial approaches that are commonly used in the treatment of mental ill health is unknown for people with Hikikomori. However, it is hypothesised that due to the interpersonal aspect of commonly used treatments, engagement in standard psychosocial therapies will be very challenging. The treatment studies that have been conducted often give treatment at the point where the person has withdrawn for longer than 6 months but is showing partial re-engagement with the outside world (albeit well below what is developmentally normal). Hence, the available treatment data does not accurately represent the whole Hikikomori spectrum (Yokoyama et al., 2023).

Given that the primary feature of Hikikomori is extreme avoidance of engaging with others, it is probable that indirect therapy approaches will need to be added to the range of intervention options (Yokoyama et al., 2023). Further understanding the psychosocial correlates and mechanisms of Hikikomori is an essential way of supporting treatments. Some psychosocial correlates that research has found so far in relation to Hikikomori are: fear of failure, self-esteem difficulties and family

dynamics (Malagón-Amor Á et al.,2015, Umeda et al., 2012, Kato et al., 2018). Continuing to gather and critically analysing this information should support the development of evidence-based treatments.

Most prevalence data have been collected in Japan, however, there is evidence of Hikikomori being observed across the world (Muris & Ollendick, 2023) and this has led to increased epidemiological and phenomenological research. Nonaka and Sakai's (2021) cross-sectional survey examining psychological factors relevant to extreme social withdrawal identified high overdependence, high psychological stress, and low self-compassion. Furthermore, Li and Wong's (2015) systematic review of quantitative and qualitative studies examining youth social withdrawal identified multiple intra-individual and interpersonal psychological factors that are relevant to extreme social withdrawal. However, the main the focus of Li and Wong's (2015) systematic review was summarizing existing information and gathering major findings to provide a greater under-standing of youth social withdrawal. Therefore, there is a need for a systematic review of the existing evidence and its quality. This review addresses the question: "*What biopsychosocial factors are associated with the development, maintenance, and remission of Hikikomori?".*Identifying and improving the understanding of the biopsychosocial factors relevant to Hikikomori development and maintenance can help guide targeted treatment and person focused interventions.

Scope of the review

- To identify biopsychosocial factors relevant to the development, maintenance and/or remission of Hikikomori
- To assess the quality of the available studies on Hikikomori using standardised quality assessment tools

Methods

Due to the nature of the literature and trial designs used to date, only elements of the PICO framework were used to organize the data extraction process.

Population: Hikikomori

Published findings on people matching the Hikikomori profile; "a person without psychosis is withdrawn into his/her home for more than six months and does not participate in society such as attending school and/or work" (Kato et al., 2018., p. 106) were found using a systematic search strategy. Search terms included: Hikikomori, Youth Social Withdrawal, Prolonged Social Withdrawal, Severe Social Withdrawal, Acute Social Withdrawal, Protracted Social Withdrawal, Primary Social Withdrawal and Hidden Youth. The term "social isolation" was initially included in the search but it the results were saturated in various unrelated research into the Covid-19 pandemic due to the overlapping of this term with research into the Covid-19 pandemic. Therefore, it was decided that the term "social isolation" would not be utilised when searching the databases. The limitation of doing this could result in a relevant paper being missed. Through forward and backward searching it was thought that this risk was reduced. All papers which reference Hikikomori or a subtype of social withdrawal were included in the Full Data Set due to the niche topic area.

Intervention Mechanisms: Biopsychosocial Factors

This review has used "biopsychosocial factors" as encompassed in Engels' Biopsychosocial model (1977). The Biopsychosocial model acknowledges that biological, psychological, and social factors, can present as separate and inter-related factors that are important when understanding illness (Engels, 1977). The World Health Organisation (WHO) International Classification of Functioning framework (ICF) definition for psychological factors is also rooted in the biopsychosocial model.

Search Strategy

This systematic review methodology followed the PRISMA statement for reporting systematic reviews (Page et al., 2021). The search strategy was developed in consultation with a research librarian. The following five databases were systematically searched for relevant studies: PsycINFO via OVID, Embase via OVID, PubMed, Web of Science Core Collection and ASSIA via ProQuest. Due to the niche topic area, the search sensitivity and specificity were kept broad by using search terminologies that were more inclusive and likely to detect all relevant papers. "Biopsychosocial factors" was not included in

the search terms but was considered manually at the screening stage. The search was done using Medical Subject Headings (MeSH) with free-text items added where necessary. Complete search strategies for each database provided (Appendix 1). Databases were first searched on the 16th of March 2023. No date restrictions were applied. Further citation searches of references and included articles were completed to check for any additional relevant studies. The last search of all five electronic databases was completed on the 21st of April 2023.

Design

This systematic review was not pre-registered on Prospero due to a notice on Prospero at the time stating a backlog of submissions and limited resources due to the Covid-19 pandemic. They also stated they did not have the resources to register reviews for students as part of their training course.

Table 1: Eligibility Criteria

Inclusion Criteria	Exclusion Criteria
○ Reports quantitative data on people with extreme social withdrawal matching the Hikikomori profile – i.e., >6 months ○ Published in English ○ Published in a peer-reviewed journal ○ Study quantitatively measures biopsychosocial factors relevant to social withdrawal/Hikikomori ○ Research design was prospective/longitudinal, observational or cross-sectional ○ Treatment studies were included if quantitative measures of change in social withdrawal were reported	○ the full article was unavailable ○ not published in a peer-reviewed journal (i.e., dissertations, conference articles, editorials) ○ Does not quantitatively measure biopsychosocial variables or correlates of Hikikomori ○ Does not measure social withdrawal/Hikikomori ○ Qualitative research or mixed methods design

Screening

Following de-duplication, titles were screened, followed by abstracts before full text reviews were screened using inclusion/exclusion criteria. To ensure articles were not missed, a hand search was undertaken of the reference lists of selected articles for additional studies of relevance. The primary researcher (AM) carried out the search, screening, and extraction of articles. To ensure a higher reliability, a co-rater (MS) reviewed a third of randomly selected texts against the eligibility criteria at full text screening.

Data Extraction

Data was extracted using general study information (author(s), date of publication, country), key inclusion and exclusion criteria, population characteristics (gender, age), prevalence of social withdrawal and any additional relevant information (such as recruitment). A table outlining the characteristics of the included and excluded studies was created (Appendix 1.2 pg.59).

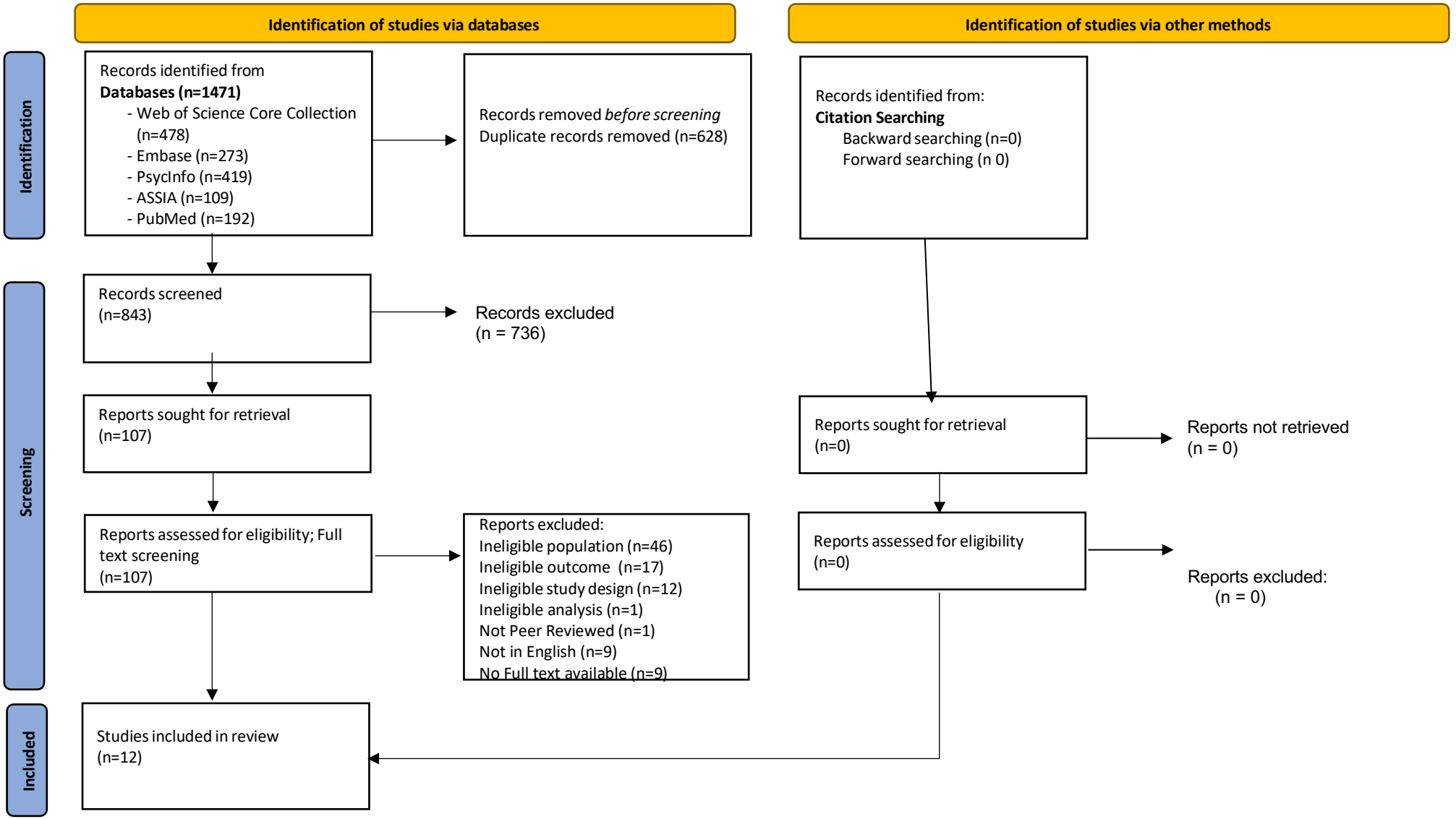
Methodological Quality Assessment

The Quality Appraisal Tool that was utilised was the Appraisal tool for Cross-sectional Studies (AXIS) (Downes et al., 2016) (See Appendix 1.3 pg.60). This tool was chosen following the screening process because it is specific to cross-sectional studies. The domains extracted and evaluated with the AXIS include: introduction, methods, results, discussion and other. To help with the critical appraisal decision making the supplementary guidance provided was used. Four papers were randomly assigned for independent appraisal (MS) and the degree of agreement was ascertained.

Results

The database search produced 1,471 records (See Figure 1). After duplicates were removed, title and abstract were screened against the eligibility criteria. One hundred and seven full texts were screened. RefWorks was used to facilitate de-duplication, screening, and co-rating procedures. Twelve studies were identified as eligible for inclusion. Backward and forward citation searching of the twelve included papers, found no additional studies not identified in the electronic search. Forward citation searching was completed using Google Scholar. In total, twelve studies were included for final review. Discrepancies between raters in relation to the critical appraisal was discussed and debated. These discrepancies were mainly related to whether the sample frame was taken from an appropriate population base and whether the selection process selected subjects/participants that were representative of the target/reference population under investigation.

Figure 1: PRISMA Flow Diagram



Narrative Synthesis

A meta-analysis was not feasible due to the heterogeneity of methods and outcomes used in the twelve included papers. Therefore, data was summarised narratively following published narrative synthesis guidelines and steps (Popay et al, 2006): explore patterns, group and describe similar findings, and develop a narrative account of the results. The included papers found little cross over of measures used.

Study & Sample Characteristics

Data relevant to the review question are presented in Table 2. The spread of country settings for the research suggests some generalisability of the findings to more diverse cultural and linguistic settings. Six studies had a Case Control study design, one used a Causal-Comparative design, two studies had a Cross-National design and there was one of each of the following study deigns; Descriptive, Observational and Cohort. The total participant number for all included studies was 2931 people. The mean age across eight studies who reported average ages was calculated to be 28 years old (five studies did not report mean age of sample; see Table 2). Sex of participants was not provided in all studies, however, on those that did provide this information participants were 2250 and recorded as male or female. Male participants in included studies represented 66% (N = 1478).

Table 2: Data Extraction Table

Author (Year) & Country	Study Design	Sample	Recruitment	Identified Relevant Biopsychosocial factor(s)	Measured Variables/Measures	Main Findings
Chan, G.H 2019 Hong Kong	Causal-comparative study	N=502 (male=384, female=118) Age Range= 12-21 years	Hong Kong residents who met Hikikomori criteria	• Empowerment • Psychological Capital	• Oiwa's (2006) five levels of social withdrawal • The empowerment scale • Psychological Capital Questionnaire (PCQ-24)	• Play Therapy was found to be significantly associated with increased psychological capital and empowerment • A positive predictor of psychological capital was significantly correlated to hours of Internet use (daily) • Empowerment above Play Therapy was the greatest predictor of psychological capital
Krieg et al. 2011 Japan	Case control study	Hikikomori N=24 (male=14, female=10) Contrast group N=59 (male=27, female=32) Hikikomori M Age= 23 years Contrast Group M Age = 21 years	Hikikomori group recruited from three programmes for *Hikikomori* sufferers Control group recruited from three universities (two in Tokyo and one in Kanagawa). All participants in both groups reside in the Kanto Region of Japan.	• Attachment • Shy temperament • Parental Rejection • Peer Rejection	• Trait Shyness Scale • Maternal Attachment Scale • Recollection of Parental Rejecting Behaviour Scale • Peer Rejection (Ijime) Scale • Maladjustment to School Scale	• Path models found both parental rejection and temperament independently impacted Hikikomori because of their effect on attachment and peer rejection.
Katsuki et al. 2020 Japan	Case control study	N=324 (male 221, female 103) M Age = 34 years	Through the Mood Disorder/Hikikomori Clinic at Kyushu University Hospital. Data was collected between May 2014 and July 2019	• Depression • Self-esteem • Poor problem solving • Autistic tendencies	• Semi-structured interview for *Hikikomori* • Japanese version of the Autism-Spectrum Quotient (AQ-J) • Hamilton Depression Rating Scale (HAMD-17) • Beck Depression Inventory II (BDI-II) • 22-item Tarumi's Modern-Type Depression Trait Scale: Avoidance of Social Roles, Complaint, and Low Self-Esteem (TACS-22) • Japanese version of the abbreviated Lubben Social Network Scale • Preference for Solitude Scale • Revised UCLA Loneliness Scale • Multidimensional Scale of Perceived Social Support	• Based on the AQ-J, Hikikomori cases have higher autistic tendencies • AQ-J showed Hikikomori cases with high Autism Spectrum Condition (ASC) were more likely to display higher traits of depression. • The TACS-22 reported more traits of reduced self-esteem in Hikikomori with high ASC. • Suggests Hikikomori with high ASC may have problem solving difficulties as a result of their difficulties in social interactions.
Katsuki et al. 2019 Japan	Case-control study	Hikikomori N=22 (male=12, female=10) Contrast group N=18 (male=11, female=7) M Age= 35.5 years	The Mood Disorder/Hikikomori Clinic at Kyushu University Hospital from June 2014 to March 2016	• Personality Traits • Attachment • Emotional Expression	• Semi-structured interview for *Hikikomori* • Self-Reported SCID-II Personality Questionnaire • The Rorschach Comprehensive System	• Found indirect emotion expression from those with Hikikomori and a potential to struggle becoming independent emotionally from their primary attachment • Scores for personality traits of avoidant, depressive, narcissistic, paranoid, passive aggressive, schizoid, and schizotypal in the SCID-II personality questionnaire were found to be significantly higher for the Hikikomori group

Malagón-Amor Á et al. 2015 Spain	Case study	N=164 (male=121, female=43) M Age= 40 years	Attended at home by the Crisis Resolution Home Treatment (CRHT) from years 2008 to 2013.	• Family; dynamics/ psychiatric history • Disorders i.e., Anxiety, /depression	• Diagnostic and Statistical Manual of Mental Disorders Text Revision (DSM-IV-TR) • Spanish version of the Severity of Psychiatric Illness (SPI) scale • Global Assessment of Functioning (GAF) • Clinical Global Impression (CGI) • Assault and Violence Assessment Tool (AVAT) • World Health Organization Disability Assessment (WHODAS)	• Family history of psychiatric illness was found in almost 60% of the Hikikomori group • Comorbidity with anxiety disorders, was found in up to 25% of the Hikikomori group • Those isolated from 1 to 4 years, were more frequent linked with personality disorders particularly cluster A
Teo et al. 2015 India, Japan, Korea, and the United States	Case study	N=72 (male 58, female 14) (Hikikomori N=36, Control N=36) M Age= not reported	Indian participants were referred from psychiatric outpatient clinics. Japanese and Korean participants were referrals from either a hospital or community mental health center. US participants responded to an online advertisement.	• Loneliness • Impaired Social Support	• Interview to assess for the presence of suspected Hikikomori • University of California, Los Angeles (UCLA) Loneliness Scale • Lubben Social Network Scale-6 (LSNS-6) • Sheehan Disability Scale (SDS) • Cornell Treatment Preferences Index (CTPI) • Sociodemographic Questionnaire	• High levels of loneliness were found using the UCLA Loneliness Scale • Descriptive data suggested those with Hikikomori more likely to be lonely. • Social support was found to be impaired
Hamasaki et al. 2022 France	Cross-national study	Japan Hikikomori N=20 (male=10, female=10) Japan Control group N=88 (male=56, female=32) France Hikikomori N=10 (male=7, female=3) France Control group N=115 (male=58, female=57) M Age= 14 years	Psychiatric outpatients who visited an adolescent outpatient clinic between December 2018 and May 2020 primarily for Hikikomori treatment and a healthy control group mainly comprised of siblings of student volunteers from the Paris Descartes University, and they were recruited using the snowball sampling method. Data compared to previous study in Japan	• Family; dynamics/ psychiatric history	• Child Behavior Checklist (CBCL4–18) • Hikikomori evaluated using an evaluation scale developed based on the definition of Hikikomori. • 5-point scale to measure environmental factors	• Parental psychiatric disorder, parental physical disorder and conflict between parents/child were found to be significantly higher in the Hikikomori group
Bellini et al. 2023 Italy	Case-control study	Hikikomori N=22 (11 males, 11 females) Control N=22 (8 males, 14 females) M Age= 15.2 years	Between 2017 and 2018, in a third level university center of Child and Adolescent Neuropsychiatry "Sapienza" University of Rome	• Anxiety • Loneliness • Attachment • Coping	• Child Depression Inventory test (CDI) • Hollingshead Index • Multidimensional Anxiety Scale for Children (MASC) • Louvain Loneliness Scale for Children and Adolescents (LLCA) • Wechsler Intelligence Scale for Children-IV (WISC-IV) or the Wechsler Adult Intelligence Scale-Fourth Edition (WAIS-IV)	• Prescence of Hikikomori in an Italian sample • A higher scoring for clinical cut-off was found in the subscale Anxious Coping and Anxiety Disorder Index for the Hikikomori group compared to the anxious/depressed group • The presence of an insecure attachment was suggested due to the significant score of separation anxiety in the Hikikomori group compared to the anxious/depressed group • Lack of social contact with peers was seen to affect mood and peer-related feelings of loneliness. • Intelligence

						• Social class
Nonaka et al. 2021 Japan	Case-control study	N=200 (male 107, female 93) M Age= 38.7 years	Large-scale web-based sample	• Psychological Stress • Coping • Self-Compassion	• Stress Response Scale (SRS-18) • Japanese version of the Brief Coping Orientation to Problems Experienced (COPE) inventory. • Japanese version of the Acceptance and Action Questionnaire-II (AAQ-II) • Japanese version of the Self-Compassion Scale Short Form (SCS-J-SF) • Adaptive Behaviors Scale for Hikikomori (ABS-H)	• Hikikomori was influenced by stress coping skills, low self-compassion and psychological stress, suggesting assessment and targeted therapy of these factors.
Chan, G.H et al. 2014 Hong Kong	Observational Study	N=588 (male 373, female 215) M Age = not reported	Online platforms	• Psychological Health • Social Relationships • Positive/ Negative Emotions	• World Health Organization Quality of Life – BREF Taiwan Version • Oiwa's (2006) five levels of social withdrawal • Positive and Negative Affect Schedule • Participants' involvement in online activities was assessed by one self-constructed question, i.e., the number of hours spent on online activities every week	• Avoidance based coping • Hikikomori as a subculture can help them re-define their self-concept and create a new identity through the internet as it provides peer support and recognition. • Social support levels were found to be associated with well-being.
Frankova, 2019 Ukraine	Case-control study	Primary Hikikomori n=13 (sex not reported) Secondary Hikikomori = 22 (sex not reported) Control group n=28 (sex not reported) M Age= 25.6 years	2014–2017 at the psychosomatic medicine and psychotherapy department of the Kyiv Railway Clinical Hospital and an online advertisement about checking the symptoms of Hikikomori was placed in social media. The age- and sex-matched control group participants were recruited among healthy volunteers	• Trauma/ Adverse events • Depression	• Toronto Alexithymia Scale (TAS-26) • Buss–Durkee Hostility Inventory (BDHI) • Quality of Life Scale (CQLS)] • Leonhard–Schmieschek Questionnaire (LEQ) • Semi-structured interview was administered to assess the presence of suspected Hikikomori according to research Hikikomori criteria	• Traumatic events over the life span were higher for the Hikikomori group. • Dysthymia levels were higher in Hikikomori population who were younger than 26 years. • Dysthymia, anxiety and excitability was significantly higher in Secondary Hikikomori compared to the control group, there was no significant difference between Primary Hikikomori and the control group
Imai et al. 2021 Japan	Cohort study	N=625 (sex not reported) M Age= not reported	Data used is from clinical records and interview sheet at the first visit, which had been collected as the daily practice at the clinics, data used was from February 1, 2020, to January 31, 2021, and from June 1, 2017, to May 31, 2018, respectively.	• Anxiety	• Overall Anxiety Severity and Impairment Scale • Three question survey to assess Hikikomori presence	• Anxiety is experienced more in those with current or past Hikikomori compared to the control group with anxiety being measured by the Overall Anxiety Severity and Impairment Scale • Current Hikikomori were shown to have higher anxiety than those with past Hikikomori • Current Hikikomori in those in their 20's and 50's and was lower in those in their 30'2 and 40's

Abbreviations: N= Number, M= Mean, R= Range, WMHJ= World Mental Health Survey Japan

Methodological Quality Appraisal

Methodological quality assessment of included papers is shown in Table 3. All studies were quality appraised using AXIS tool as the included papers mainly carried out on cross-sectional analysis. The AXIS tool was thought suitable and ensured a reliable method for quality appraisal.

Overall, the AXIS highlighted a generally good quality of reporting. Included studies were clear in their; aims and objectives, target population, limitations, and determinants of statistical significance. However, one study was less favorably appraised for its unclear aims and omission of consent (Katsuki et al., 2019).

None of the included studies provided sample size justification. Furthermore, none of the studies reported a sample size power calculation. Also missing from the papers was measures to address and categorise non-responders. This impacted the assessment of response rate bias. Additionally, 33% of the studies did not make it clear whether there was any conflict of interests for the author when interpreting the results.

Moreover, when assessing the selection process only three studies appeared to be representative of the target population (Chan, G.H, 2019, Chan, G.H, et al.,2014, Nonaka et al., 2021). Most studies recruited participants who were engaging in some form of treatment or support. However, the core feature of the target population is extreme social withdrawal so those recruited do not capture those who are not engaged in treatment or services, these studies are not representative of the target population.

Co-rater (MS) selected four included studies randomly and independently quality appraised the selected papers using the AXIS tool. Using the Cohen's Kappa calculator for inter-rater agreement, it showed good agreement between raters K =.761, p=<.001. Discrepancies related to sample frame and target/reference population under investigation which was understood through discussion of the spectrum of Hikikomori and the participants used.

Table 3: Methodological Quality (AXIS)

	Questions																		
	Intro	**Method**										*Results*					*Discussion*		*Other*	
Authors	*1*	*2*	*3*	*4*	*5*	*6*	*7*	*8*	*9*	*10*	*11*	*12*	*13*	*14*	*15*	*16*	*17*	*18*	*19*	*20*
Chan, G.H	☑	☑	X	☑	☑	☑	X	☑	?	☑	☑	☑	?	X	☑	☑	☑	☑	X	☑
Krieg et al.	☑	☑	X	☑	☑	X	X	☑	?	☑	☑	☑	?	X	☑	☑	☑	☑	?	?
Katsuki et al.	☑	☑	X	☑	☑	X	X	☑	☑	☑	☑	☑	?	X	?	☑	☑	☑	X	☑
Katsuki et al.	X	☑	X	☑	☑	X	X	☑	☑	☑	☑	☑	?	X	☑	☑	☑	☑	?	?
Malagón-Amor Á et al.	☑	☑	X	☑	☑	X	☑	☑	?	☑	☑	☑	?	☑	☑	☑	☑	☑	X	?
Teo et al.	☑	☑	X	☑	☑	X	X	☑	☑	☑	☑	☑	?	X	☑	☑	☑	☑	X	☑
Hamasaki et al.	☑	☑	X	☑	☑	X	X	☑	☑	☑	☑	☑	?	X	☑	☑	☑	☑	X	☑
Bellini et al.	☑	☑	X	☑	☑	X	X	☑	☑	☑	☑	☑	?	X	☑	☑	☑	☑	X	?
Nonaka et al.	☑	☑	X	☑	☑	☑	X	☑	☑	☑	☑	☑	?	X	?	☑	☑	☑	X	☑
Chan, G.H et al.	☑	?	X	☑	☑	☑	X	☑	?	☑	?	☑	?	X	X	☑	X	☑	?	?
Frankova,	☑	☑	X	☑	☑	X	X	☑	☑	☑	☑	☑	?	X	☑	☑	☑	☑	?	☑
Imai et al.	X	X	X	☑	X	X	X	X	X	☑	X	X	?	X	X	X	?	X	X	☑

X – No **☑ – Yes** **? – Don't know**

Co-rated studies highlighted in ***bold,*** *italics* and <u>underlined</u>

Biopsychosocial Factors

There were twenty-one biopsychosocial concepts classified as biopsychosocial factors relevant to the development and/or maintenance of Hikikomori. These factors were then taken and synthesised into categories to enable an easier understanding of common themes.

Biopsychosocial Factors Associated With Hikikomori

Biopsychosocial factors were grouped under the following terms: Disorders, Interpersonal Dynamics, Biopsychosocial Capital, and Negative Experiences. Due to the variation in findings between all twelve studies and data quality, some studies were referred to more than others either due to being the most common across the studies and/or due to them adding new information to the current research.

Disorders

Those grouped into the category of disorders were done so using the World Health Organisation (2022) definition of a mental disorder "is characterized by a clinically significant disturbance in an individual's cognition, emotional regulation, or behaviour. It is usually associated with distress or impairment in important areas of functioning." This included disorders such as. Anxiety and depression.

Psychiatric history in the family

Malagón-Amor Á et al. (2015) found almost 60% of the participants identified as Hikikomori had family with psychiatric histories, (44.4% - first-grade relatives, 55.7% - mother). The most predominant disorders for mothers were affective (46.2%) and anxiety (23.1%). While the father's common diagnoses were psychotic (40%) and drug use (40%). This high occurrence of family psychiatric history is important when thinking about how to identify and treat this population. Similarly,, Umeda et al. (2012) identified the connection between maternal mental disorders with Hikikomori. It was proposed that this connection could be due to parenting behaviours, reinforcing the child's anxiety and avoidant coping strategy. Arguing the case for family support and psychoeducation. Additionally, Hamasaki et al. (2022) also identified the Hikikomori patient group's scores were significantly higher than in the control group for "parental psychiatric disorder".

Depression

Bellini et al., (2023) present evidence to support the hypothesis that people with Hikikomori are at an increased likelihood to suffer from periods of depression as withdrawal duration increases. It was thought that the prolonged lack of peer social contact affected their mood. The results of this study also found Hikikomori repeatedly associated with social phobia suggesting a possible maladaptive defense to avoid social relations as a way of managing social phobia. This maladaptive defense thus leads to long term overall adaptive impairment increasing the risk of developing mood disorders and peer-related loneliness. The aforementioned study by Malagón-Amor Á et al.,(2015) found only three people (1.8%) with primary

Hikikomori, which is where Hikikomori is present without symptoms indicative of mental disorder.

Interpersonal Dynamics

Factors that were considered "Interpersonal Dynamics" were factors which indicated an interplay between people. These interplays included feelings, thoughts, and behaviors that promoted or hindered a relationships development and maintenance (Penn State, 2023).

Attachment

A significant score for "Separation Anxiety" within the Hikikomori participants was found by Bellini et al., (2023). Leading the researchers to hypothesise the existence of an insecure attachment in those with Hikikomori. This link with attachment was also noted in Katsuki et al. (2019). They found texture-related responses in the Rorschach Comprehensive System, which are related to attachment, dependence, and psychological reliance particularly between the child and their caregiver (primarily mother). Additionally, they found that people with high Coping Deficit Index in the Rorschach Comprehensive System have difficulties adjusting to social situations. The study found that 91% of those with Hikikomori lived predominantly with their parents, leading the researchers to suggest that some people with a high desire for basic affection and emotional support, might withdraw into their home as they are unable to fulfil their basic need for affection and emotional support in social situations like school and the work. Hamasaki et al. (2022) noted that while family factors contribute to the severity of Hikikomori worldwide, these factors can differ between countries, such as Europe and Japan.

Rejection

Krieg et al. (2011) results showed a link between attachment and peer rejection. They found that parental rejection had an effect on those with Hikikomori via attachment and peer rejection. Parental rejection was recalled by the Hikikomori participants and predicted bullying and rejection from peers in adolescence. This led researchers to propose that the risk of Hikikomori is increased due to combined effects of peer rejection and anxious attachment.

Psychological Capital

Factors grouped under Psychological capital referred to a person's mental reserves and their ability to sustain them through tough situations. These reserves are; Hope, Efficacy, Resilience and Optimism. Hope - future plans and goals, Efficacy - managing tasks successfully, Resilience - skill of adapting positively to difficulty or failure, Optimism – positivity for the future (Luthans et al., 2007).

Wellbeing/coping mechanisms

Many of the included studies identified a lack of psychological capital, more specifically efficacy with problem solving (Katsuki et al., 2020), coping and empowerment (Nonaka et al., 2021). Chan, G.H, (2019) looked at whether Play Therapy would be effective in enhancing the development of coping skills, a person's sense of well-being and any empowerment effect. Chan found a particular correlation between longer length of time on the internet and increased benefit from play therapy in relation to development of wellbeing and coping mechanisms. Similarly, Bellini et al., (2023) found those with Hikikomori presented significantly higher scoring in the subscale anxious coping than the anxious/depressed participant group. Potentially suggesting that people with Hikikomori have increased difficulty coping with distressing situations, for instance; school and interpersonal relationships, resulting in an avoidance strategy being utilised.

Negative Experiences

Various research into Hikikomori highlight the role of negative experiences prior to a person withdrawing into a level in line with Hikikomori. Negative experiences were understood to be an experience or situation that was deemed by the person to be negative, depressing unpleasant, or harmful (Collins online dictionary, 2023).

Frankova, (2019) found that, the earlier social withdrawal happened the greater impact trauma had on the person. Additionally, they found that people with primary Hikikomori (i.e., free from a psychiatric disorder) were found to have a higher number of traumatic events across their life span. The primary Hikikomori group were found to have more often faced psychological traumas than those with secondary Hikikomori and the control group. However, those with secondary Hikikomori were found to have witnessed a serious injury or death more commonly. Prevalent traumatic situations reported for those with Hikikomori were; “emotional insults or neglect (54.3%), learning about a serious life-threatening injury or an unexpected death of a close person (45.7%), emotional disturbances of significant others (42.9%), or parental divorce (34.3%)”. Chan, G.H et al. (2014) recognises a similar finding, suggesting those with Hikikomori decide to withdraw because of the stigmatisation for failing to meet the expectations of society.

Discussion

This systematic review aimed to investigate biopsychosocial factors associated with Hikikomori. Across all studies it identified twenty-one biopsychosocial factors that were significantly associated with Hikikomori. These factors showed some cross over (e.g., anxiety problems), hence grouping of the factors into themes gave a more synthesized account of the current literature. The review explored patterns identified and a narrative account of the results was developed.

Studies included were evaluated to be of overall good quality using AXIS, with high quality of reporting apparent in most of the included papers. However, generalisability issues were evident across almost all of the studies apart from three papers (Chan, G.H., 2019, Chan, G.H., 2014, Nonaka et al., 2021), this was due to recruitment procedures. Most of the studies recruited people with Hikikomori who were in some form, already linked in with support and treatment. Due to the reclusive nature of this population, it does not accurately represent those who are not involved in services. Future studies should take this into account and look to those studies who have managed to recruit a more gerneralisable participant group to identify ways of gaining their collaboration (Malagón-Amor Á et al.,2015).

Hikikomori has often been seen as a culture-bound syndrome, yet the available literature for this review yielded results not only from Japan but also Spain, Hong Kong, the Ukraine and the United States. This identifies the presence of Hikikomori outside of Japan going against earlier theories that identified Hikikomori as a culture-bound syndrome (Teo & Gaw, 2010) suggesting that the cultural environment on the emergence and persistence of Hikikomori plays a smaller role than previously thought. The idea of it being a cultural bound syndrome could reduce recognition of it outside of Japan and potentially lead to misdiagnosis in other countries. Resulting in a potential delay of appropriate treatment (Bommersbach & Millard, 2019). Hence, it is important to understand Hikikomori outside of Japan, so that its prevalence rate is understood, and a common diagnostic language is used (Bommersbach & Millard, 2019).

In cross cultural studies there are patterns of Extreme Social Withdrawal that are suggestive of Hikikomori. However, examinations of the studies have shown some variations of duration and pervasiveness of the withdrawal in some countries. These variations reflect the fact that even in Japan these nuances are being defined (Teo et al., 2013). Therefore, the definition of Hikikomori is not fully consistent across studies. This causes implications for the state of our current understanding of morbidity effect and impact on not only the person but their family

and society (Teo et al., 2013). Furthermore, it hinders research and advancement on this topic (Nonaka & Sakai, 2023). This lack of congruency between countries could result in less individuals meeting the hikikomori definition and therefore not having access to the appropriate treatments (Nonaka & Sakai, 2023). Further research into the phenomenon of hikikomori and its variations could increase accuracy and eventually consistency of definition and treatment across counties.

As found in some studies (Malagón-Amor Á et al.,2015, Umeda et al., 2012, Hamasaki et al., 2022), family psychiatric difficulties have a significant association with Hikikomori. This finding is important to consider when thinking about early detection. Increased support and mindfulness of descendants and relatives of those with mental illness could aid in the prevention of Hikikomori through early interventions such as parent-child interventions or psychotherapy (Kato et al., 2018). Subsequently, findings of the high comorbidity of people with Hikikomori with anxiety disorders in this review, have also been seen in previous studies (Nagata et al., 2013; Teo, Lerrigo, & Rogers, 2013), particularly social anxiety. Included studies also found comorbidity with affective disorders and autism spectrum disorders (Malagón-Amor Á et al.,2015, Katsuki et al., 2020). Again, this is consistent with previous literature (Kato, Shinfuku, Sartorius, & Kanba, 2011; Koyama et al., 2010; Teo, 2013) and reinforces the need for prevention of Hikikomori through early interventions for mental health difficulties.

Due to the challenge of operationalising biopsychosocial factors and the accompanying heterogeneity within the wider literature, sensitivity was prioritized for the search strategy, resulting in the screening of all available papers that incorporated Hikikomori. This permitted an extensive coverage of the available literature. As a result, the papers that were included were heterogeneous in measures and statistical methods, making it more difficult to compare or interpret study findings.

Future Research Directions

There were a number of case-control designs, however the included study designs varied. Due to the questions around cultural aspect of Hikikomori, future research could further explore biopsychosocial factors across a range of cultures using cross-national study designs. This increased understanding of factors that are seen in various cultures could potentially identify targets for treatment. Most of the studies outside of Japan that identified biopsychosocial factors were the first of its kind in the country such a Spain, Italy etc., thus replication is required to

support interpretation.

Lastly, as highlighted by the quality assessment tool, studies better representing the target population and not solely those in the population that are engaged in treatment, or some form of service support would increase understanding of the most hidden Hikikomori population. As seen in other studies (Chan, G.H, 2019) through online engagement and recruitment it is possible to get a more accurate and diverse range of people with Hikikomori.

This systematic review was not pre-registered on Prospero, which is a limitation as pre-registering supports transparency, prevents unintended duplication of reviews and can reduce potential bias. Registration offers advantages to many stakeholders in return for modest additional effort from the researchers registering their reviews.

The PRISMA flow diagram shows that after duplicates were removed 843 records were left identified for screening. The approach taken was to first screen the titles of the 843 papers to rapidly exclude those that were ineligible. This relied on the title conveying enough information for this judgement to be made accurately. Once the title screening was done the abstracts of the remaining potentially eligible papers (n=513) were then screened and further ineligible papers were discarded. A possible limitation arising from this approach is that the titles may have conveyed insufficient information to inform a robust inclusion-exclusion decision. However, the sensitivity checks done (e.g. forward and backward citation searching) suggest that the eligible pool of papers was successfully identified. Concerns about this possible limitation could be avoided in future studies by conducting concurrent screening of titles and abstracts.

Conclusion

This review is the first to systematically identify and synthesise the current research looking at links between biopsychosocial factors and Hikikomori. The twelve included studies found twenty-one biopsychosocial factors associated with Hikikomori that could possibly help inform appropriate treatment and support. Further research could build on the existing evidence base and investigate whether the identified factors can be used when developing targeted interventions. Findings of this review indicate that future research should (1) further explore biopsychosocial factors across a range of cultures using cross-national study designs to estimate worldwide prevalence rates and develop a common diagnostic language, (2) undertake

replication studies to confirm reliability and consistency and (3) be more aware of their selective process to ensure generalization to the Hikikomori population.

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Chapter 2

Scotland's Hikikomori: Estimating Patterns of Extreme Social Withdrawal in Young People Following COVID19

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Plain Language Summary

Title

Scotland's Hikikomori: Estimating Patterns of Extreme Social Withdrawal in Young People Following COVID19

Background

Hikikomori is a form of extreme social withdrawal, they do not attend a job or school, rarely have friends, and even withdraw contact with family (Teo, 2009). Adolescent and young adult men are thought to be at higher risk. This form of social withdrawal is on the rise worldwide (Kato et al., 2019) and it is thought that the recent COVID-19 pandemic will result in a worldwide increase of people withdrawing socially (Gavin & Brosnan, 2022).

Glasgow City Council have noticed a rise in school absences, specifically unexplained school absences since the start of the COVID-19 pandemic. Understanding of this rise is important to identify any social impact this may have and implement targeted interventions.

Hikikomori has a low detection rate making it a hidden mental health issue with those suffering falling through the nets of services. Delayed access to appropriate mental health support can lead to multiple negative outcomes such as family distress and avoidable disability (Teo, 2009).

Aims

The primary aim of the study is to estimate rates of extreme withdrawal and social isolation in high school aged children in Glasgow. The secondary aim is to understand the impact of the COVID-19 pandemic on social withdrawal in adolescence. The final aim is to identify data systems and collection processes for distinguishing this population.

Methods

Linking anonymised Children and Adolescent Mental Health Services (CAMHS) records with educational absence records to estimate rates of withdrawal and social isolation in young people in Glasgow and those who are/are not receiving CAMHS input.

Main findings and conclusion

Data suggests that 2228 young people are slipping through current safety nets. Furthermore, pre-mid-and post pandemic patterns suggest worsening absenteeism. Additionally, when

taking deprivation into account, 38 young people are withdrawing from society without explanation, 20 of these were male which, based on previous observational studies, is a risk factor for those progressing to a state of Hikikomori.

This study provides valuable information in relation to the estimated rates of withdrawal and social isolation in Glasgow. It also identified data systems and collection processes that could help to catch this population who are currently being missed by the current safety nets. This may help inform targeted service delivery, aid identification of young people for further assessment, improve early detection, and reduce the long-term impact specifically for those young people withdrawing to the extreme socially. Lastly, it aided in understanding the impact of the COVID-19 pandemic on increasing withdrawal in adolescence, potentially increasing some young people's vulnerability to withdrawing to a level consistent with Hikikomori.

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Abstract

Aims

Hikikomori is an extreme form of social withdrawal. Since those with Hikikomori withdraw from work/school, it is thought that a high school absentee rate could be an early predictor for some young people, persisting in their withdrawal and conforming to the Hikikomori pattern.

Additionally, it was thought that the COVID-19 restrictions may also worsen the Hikikomori pattern social withdrawal for those already at risk of withdrawing from society.

Methods

Data collected from Glasgow City Council secondary schools identified young people whose attendance fell below 60% of school days. Of the absences, 50% or more were recorded as unauthorized absences. Identifiers were extracted for linkage to Children and Adolescent Mental Health Services user data. Data was compiled for August to February: pre COVID-19 pandemic (2019-20), mid COVID-19 pandemic (2020-2021) and post COVID-19 pandemic (2021-2022). Those identified due to their duration of school non-attendance were then screened for any involvement with Children and Adolescent Mental Health Services (CAMHS). A lack of referral to or contact with CAMHS reduced the chance that the school absence was explained by a mental health condition.

Supplementary data collected such as deprivation and gender, was then screened against the main population of interest to see if it enabled any further identification of young people at an enhanced risk of progressing to a state of Hikikomori.

Results

Data showed that out of 2454 young people meeting criteria only 226 are known to CAMHS. This suggests that 2228 young people are slipping through current safety nets. Furthermore, pre-mid-and post pandemic patterns suggest worsening absenteeism.

Conclusion

Approximately 1% of high school aged children in Glasgow may be at risk of extreme social withdrawal. Furthermore, an increase post-COVID-19 in those withdrawing from school was identified. Additionally, many benefits were demonstrated to using existing data systems and collection processes in research. It is hoped that this study could influence future research into

understanding the phenomenon of Hikikomori in Scotland.

Key words; COVID-19, data linkage, Hikikomori, withdrawal

Introduction

Social withdrawal is "voluntary isolation prolonged in time that involves the cessation of any form of social relationship and contact with people and the outside" (Morese, R. et al., 2020, p.1). It is a transdiagnostic symptom which, if persistent, can worsen mental health problems.

Hikikomori is a form of extreme social withdrawal marked by minimal contacts outside the home lasting for more than six months. This relatively new syndrome rose to public awareness in Japan in the 1990s (Saitō, 2013). It is thought to affect hundreds of thousands in Japan and is seen as a "silent epidemic" (Teo, 2009). Although first identified and characterised in Japan it is becoming an increasing issue worldwide (Malagón-Amor et al., 2014).

Despite Hikikomori's rise in prominence, it is not an "official" psychiatric diagnosis as there is no definition set out in the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) and International Classification of Diseases (ICD-11) (Teo, 2009). In 2010, The Japanese Cabinet Office provided the following operational definition for Hikikomori: "a situation where a person without psychosis is withdrawn into his/her home for more than six months and does not participate in society such as attending school and/or work" (Kato et al., 2018., p. 106). Hikikomori is broken down further into "Primary" and "Secondary" Hikikomori. Primary Hikikomori is where the person is "free from a psychiatric disorder" and is less common. Secondary Hikikomori is "secondary to other psychopathology" (Malagón-Amor et al., 2014). However, social withdrawal and social disinterest is a symptom of multiple mental health conditions in the DSM-5 and ICD-11. Due to recognition in relation to other mental health difficulties it has led to the conclusion that it might not be a new diagnosis but associated with the presence of another mental illness (Malagón-Amor et al., 2014).

The onset of Hikikomori is predominantly in late adolescence and disproportionately affects young adult males who withdraw into their bedrooms within the family home, also withdrawing contact with family (Teo, 2009). Research into prior economic recessions like the 2007-08 Financial Crisis, identified younger people to be at increased likelihood of suffering the consequences such as unemployment (Wong, 2020). Research has shown in general that increased absence from employment and education increases the experience of marginalisation, impacting negatively on a young person's mental health, increasing vulnerability to dependence, self-harm, and suicidal behaviour (Wong, 2020). Past societal shocks such as the economic crisis in Japan in the 1990s had an effect on withdrawal rates (Wong, 2020). Similarly, it is plausible that the COVID-19 pandemic may increase the risk of

Hikikomori. Comparing data pre, mid and post COVID-19 pandemic could identify whether there has been an increase in extreme withdrawal since the pandemic, enabling exploration of the impact of the pandemic on extreme social withdrawal. Thus, identifying whether social and economic shocks are a factor that may worsen marginalization and withdrawal behaviors.

Hypotheses of Origin

Japanese culture follows a particular parent-child relationship where fathers are notably absent, but an exceptionally lengthy bond is formed with the mother (Kato et al., 2018). This dynamic could lead to difficulty developing the interpersonal skills and independence needed in adolescence/adulthood. Research in Japan (Iwakabe, 2021; Kato et al., 2018) has questioned whether this dynamic could potentially lead to these young people developing a vulnerability to school/workplace stress, triggering the desire to escape social situations. Although western culture does not generally follow this type of parent-child relationship, the fact that Hikikomori presentations have been observed across cultures suggests that factors related to the attachment relationship may be relevant to the development of these types of behaviours.

Individuals presenting with Hikikomori often share the experience of adverse events leading to low self-esteem (Iwakabe, 2021). This adverse experience thus leads this population to socially withdraw, avoiding re-traumatization (Rooksby, Furuhashi & McLeod, 2020). Additionally, in Japan, there is shame associated with mental health problems. Although, Hikikomori has a high prevalence rate, families will often hide it “because of a strong sense of societal shame” (Iwakabe, 2021).

One factor implicated in the rise of Hikikomori is change in communication due to the rapid evolution of technology such that virtual interaction often replaces face to face communication (Kato et al., 2018). This enables an easier withdrawal from social situations physically, allowing it to go unnoticed for some time, and so advances in technology have been blamed for reduction in social activity for some people (Dong, Li and Baker, 2022). However, the way in which technology is used impacts the likelihood of social withdrawal, with online gaming emerging as a risk factor for worse social withdrawal compared to using technology for social media which decreases social withdrawal risk (Dong, Li and Baker, 2022).

COVID-19 restrictions may have worsened Hikikomori-pattern social withdrawal in those already at risk of withdrawing from society (Gavin & Brosnan, 2022). While people with Hikikomori voluntarily avoid social situations and COVID-19 restrictions were government

forced social isolation, this change in lifestyle might exacerbate the risk of young people choosing not to re-join society (Gavin & Brosnan, 2022). Loss of opportunity to participate in socially meaningful roles like work/education during adolescence could increase social anxiety leading to withdrawal from society and development of Hikikomori (Gavin & Brosnan, 2022). During similar epidemics (SARS/MERS) research showed a corresponding increase in mental health difficulties such as anxiety and depression post epidemic (Karakasi, Kevrekidis and Pavlidis, 2020; McAlonan et al., 2007; Brooks et al., 2020). Similarly post-COVID-19 life will be changed both economically and socially causing substantial mental health difficulties. Research into the COVID-19 pandemic has similarity to the SARS/MERS epidemics research identified an increase in mental health issues such as anxiety and depression. However, research is likely to miss those who continue to withdraw socially, due to Hikikomori's central feature of their desire to be hidden from society (Rooksby et al., 2020). Consequently, an increase in Hikikomori is a strong possibility after the COVID-19 pandemic (Kato et al., 2020; Roza et al., 2021).

Linking Withdrawal to School Attendance Patterns

Research has shown that school dropout and absenteeism are linked to various difficulties throughout the lifespan (Gubbels et al., 2019, p1). School avoidance may occur for various reasons; however, it could indicate life-course problems. Research has shown increased avoidance/absence from education increases the experience of marginalisation, impacting negatively on a young person's mental health (Wong, 2020; Knollmann et al., 2010). Although not conclusive, research has found a link between school avoidance and poor mental health (Knollmann et al., 2010). Within this overall behaviour of not going to school there will be some young people who will persist in their withdrawal and so conform to the Hikikomori pattern. Since one of the fundamental parts of Hikikomori is school absenteeism, with limited data on how to identify young people at risk of Hikikomori and how to estimate the rates of Hikikomori type withdrawal, non-attendance could provide a guide of general health and functional problems in young people. So, school non-attendance could indicate a transitional sign to the development of Hikikomori.

Due to the lack of a globally agreed definition and varying cultural views of Hikikomori, there is often a delay in sufferers accessing appropriate mental health support (Kato et al., 2018). This delayed access can lead to a major loss of potential, increased family distress, and avoidable disability. There has been increasing recognition that this extreme social withdrawal can lead to those with Hikikomori becoming invisible to normal safety nets within health and education systems. Hikikomori also has a negative influence on wider systems such as education and

workforce (Kato et al., 2019). This combined with a potential increase in cases post COVID-19 make it a priority issue to understand and create a more solid evidence base for identification and treatment (Teo, 2009).

The Role of Health Service Data

There is very little research examining young people who are no longer in school, have withdrawn from society and are unknown to other services such as CAMHS. These young people will not seek help themselves and parents might delay seeking help for various reasons until the problem has progressed to an advanced stage, making it harder to get the young person back into training or education. Thus, it is important to develop new methods to identify and quantify the problems of social withdrawal as a potential early marker of risk.

Young people are referred to CAMHS for a variety of different mental health reasons and varying severities of mental health difficulties. Some will be seen within specialist teams such as Forensic CAMHS, others will be seen in outpatient CAMHS and for very severe mental health difficulties, an inpatient unit under the mental health act. By linking school attendance rates to CAMHS data, it is possible to aid estimation of this hidden population of young people who are becoming invisible to society and to address their needs (Cotton, et al., 2022; Mansfield, Gallacher, Mourby and Fazel, 2020). Moreover, considering technological advances and available data over the last decade, this area is a potentially under utilised resource especially regarding linking educational data (Mansfield, Gallacher, Mourby and Fazel, 2020). Additionally, it is a resource which is likely to continue to develop and provide increased information that could aid research. Use of such a resource has the potential to control variables, observe additional outcomes, and enrich information available.

Aims

The overarching aim of this project was to develop and test ways of using administrative data on school attendance and CAMHS service contact to improve the estimation of rates of students at risk of developing hikikomori in school age adolescents.

Our primary objective was to describe profiles of withdrawal (i.e., withdrawal for >6 months with/without input from CAMHS). Further, exploration was then undertaken to understand whether risk of extreme withdrawal is associated with socioeconomic factors such as deprivation (based on the Scottish Index of Multiple Deprivation; SIMD, 2020) and gender.

Our secondary objective was to compare rates of school withdrawal across three main timepoints - pre, mid, and post COVID19 pandemic to examine rates of extreme school withdrawal across the Covid-19 timeline.

Methodology

Design

This investigation adopted a quantitative data linkage design to estimate rates of school withdrawal in young people in Glasgow with/without input from CAMHS. Demographic data collected was screened against population findings to detect any further identification of young people at an enhanced risk of progressing to a state of Hikikomori. Administrative and health data from government funded schools and NHS CAMHS services were compiled for three periods: August to February for 2019/20, 2020/21 and 2021/22. The data extracted was restricted to the start of the school year in Scotland (August) until February (6 months), which was just before the COVID19 pandemic restrictions came into effect in the United Kingdom (March). This was done for two reasons, one the six month time period also fits with the Hikikomori presentation of withdrawal for 6 months or more and two it is hoped to allow for a more accurate representation of pre-COVID19 attendance. Again, in order to increase comparison, the criteria of August to February was utilised in the mid- and post-pandemic data.

Cohort Description

Administrative data on attendance at government funded schools in the GCC catchment was processed to identify the names of young people whose attendance was below 60% of available school days. Of the absences, 50% or more needed to be recorded as unauthorized absences. Names, date of birth and post code were used to achieve linkage to CAMHS service use data.

Data Sources

GCC school attendance data is gathered daily, collating information about young people's attendance and absences from school. This information is stored centrally in the internal GCC Content Management Interoperability Services (CMIS) system and stored throughout pupils' school years. Data meeting the stipulated attendance threshold was made available from GCC Psychology Services and transferred securely to National Health Service Greater Glasgow and Clyde (NHSGG&C) CAMHS servers for processing under the terms of the existing Data Sharing Agreement between GCC and NHSGGC.

Transfer of data from GCC servers was done via encrypted file transfer and compiling of the

data base completed on NHSGGC computers. To prevent unauthorized exposure to personal information all data were processed in a private space on NHSGGC premises.

CAMHS data taken from the internal NHS GG&C Egton Medical Information Services (EMIS) electronic health records system was added to the study data file. The list of names was cross-checked with the EMIS health record to ensure accurate case linkage and data was compiled on service use and referral status. Specific service use data extracted from the health record:

- Referral status (ended, rejected, or currently active referral)
- Service utilization (attendance at appointments, missed appointments, type of appointment)

Linkage between the school attendance data and CAMHS service use data was completed using the following data:

- Date of birth
- First name and surname
- Recorded postcode of home address

The SEEMiS Identification Number (SEEMiS is an Education Management Information System) was also given by GCC however this identification number is different to the CHI Number (Case Reference Number) used by CAMHS.

Once the linkage between the school attendance data and CAMHS service use data was completed the SEEMiS Identification Number was replaced with a study code. The log of study code and SEEMiS data is stored in a password protected file on NHSGGC servers with access restricted to those involved in the study. The pseudonymized database was transferred to secure Glasgow University IT systems for analysis to ensure anonymity was preserved.

Population

The population of interest is young people exhibiting extreme social withdrawal not explained by health or mental health factors. We attempted to sample this population by identifying young people with erratic or persistent non-attendance at publicly funded secondary schools in the catchment of Glasgow City Council Education authority who did not have involvement with CAMHS.

Recruitment

Extraction of pupils with attendance lower than 60%, with more than 50% of their absences being unauthorized, brought the number down from approx. 28,000 to 2454 combined over the three time points. Due to the number of pupils, it is thought that the study is indicative of the general school population.

Research Procedures

Attendance data provided by GCC was linked to CAMHS records resulting in a single database using name, date of birth and postcode to link cases.

Due to the information available a probabilistic data linkage application was used. This was done in 3 steps:

- Pre-linkage: editing and data cleaning, parsing fused strings such as first and last name or post code, and standardizing matching variables so that they have the same formats and definitions.
- Linkage: bringing pairs together for comparison and determining correct matches, i.e., the pair belongs to the same entity.
- Post-linkage: checking residuals for the unmatched, determining error rates and other quality indicators and carrying out analysis taking into account linkage errors." (Gabriele, 2019).

Triangulating school attendance records with CAMHS attendance in time epochs before, during and after the pandemic.

Analysis

The study adopted two analytical approaches. The first analytical strategy sought to explore estimated rates of withdrawal and social isolation currently observed in this sample and identify data systems and collection processes for distinguishing this population. Descriptive information on the patterns of withdrawal over a six-month period in line with Hikikomori, description of sub-groups found and how much crosses over with contact/non-contact with CAMHS.

The second analytical strategy was to understand the impact of the COVID-19 pandemic on social withdrawal patterns. Data was grouped according to time scales to explore the proportion of withdrawal across epochs of the COVID-19 pandemic.

When determining the deprivation scale the Scottish Index of Multiple Deprivation Scale (2020) was used. Scores range from 1 to 5 with 1 being the top quintile of deprived areas in Scotland and 5 being the least deprived quintile.

Statistical considerations

Sample Size

Number of high school pupils in Glasgow council schools per year is approx. 28,000. Extraction of pupils brought the number down to 2454 in total over the three time points. Due to the number of pupils, the study should be indicative of the population enrolled in Glasgow secondary schools. However, adequate sampling frame is unknown as there is no known rate in the Scottish population.

Inclusion Criteria

Participants were included in the study database if they were:

- aged 11-19 years
- enrolled at a secondary school in the GCC Education Authority
- resident within NHS GG&C health board catchment
- have an unexplained attendance record below 60% with 50% of their absences unauthorized

Exclusion Criteria

- Those with school non-attendance below the 60% threshold but more than 50% of their absences were authorized.
- Those with school non-attendance below the 60% threshold were excluded if their absence was explained by factors such as long-term sickness.

Data analysis

Descriptive data was presented to characterize the patterns of non-attendance over time (e.g., proportions and frequencies). Moreover, the extent demographic (e.g., sex, deprivation, age) affect relationships regarding CAMHS was investigated. These patterns were compared across the three timepoints that are linked to the pre-, mid-, and post-pandemic phases. A series of Pearson's chi-square tests of association and one way analysis of variance (ANOVA) were conducted to address the studies research questions. Additionally, any significant effects found was followed up by Bonferroni Post-hoc tests due to its flexibility and use with all types of

statistical tests.

Although it was possible that the same young people could be included in the data across the three time periods, this was not the case. Through examination of unique identifiers, it was determined that the young people at each time point consisted of different pupils.

Ethics (See Appendix 2.1, 2.2, 2.3: Pg. 62-67)

NHS ethical approval for using patient data was obtained from the NHS Research Ethics Committee (REC). The NHS GG&C Research and Innovation (R&I) department granted approval to use a GG&C's research site. Given the use of administrative data, it was not necessary to gain consent from participants, but permission was obtained from a CAMHS Caldicott Guardian to access NHS GG&C patient data collected for routine clinical purposes. Additionally, an Education Support Research Group (ESRG) application was made to GCC for approval to access and use routinely collected GCC data. An existing data sharing agreement between GCC and the NHS enabled the sharing of the data. Data was collected, pseudonymized and stored in line with current regulations and policies (General Data Protection Regulation (2018) and NHS GG&C policies).

Results

Data linking was undertaken by combining four variables to identify 226 matches, but this approach can result in a considerable number of unmatched cases due to incomplete or missing data (see table 1 below), and variations/errors.

Table 1.

Referral linkage rate using different linkage variables.

Note Name refers to First Name and Surname and are considered separate variables.

Year	Council Count	Name Match	Name & D.O.B Match	Name, D.O.B & Postcode Match	% of complete linkage
19-20	760	78	67	46	6%
20-21	694	48	42	32	5%
21-22	1000	61	53	36	4%

It is unknown whether the numbers included in this study represent an increase in withdrawal rates or are proportional with changes in overall numbers of students. This issue will be explored in the discussion.

Unique Identifiers

Data was screened by reviewing unique identifiers, to determine if any young person was included in more than one data collection time period. Though probable, this was not the case, samples at each time point contained different pupils.

Estimated School Withdrawal

GCC provided 2454 records for young people who met the inclusion criteria over the three time periods. Of the 2454 provided by GCC a count of 226 young people were known to CAMHS. This leaves 2228 young people who are withdrawing from school that are also unknown to CAMHS. Frequency data broken down over three time periods; pre-pandemic (August to February 2019-20) was 760, mid-pandemic (August to February 2020-2021) was 694, and post-pandemic (August to February 2021-2022) was 1000. Furthermore, frequency data for those with 100% unauthorized absence broken down over three time periods; pre-pandemic (August to February 2019-20) was 216, mid-pandemic (August to February 2020-2021) was 210, and post-pandemic (August to February 2021-2022) was 235.

In order to determine whether unauthorized absence varied according to the time periods, a One-way between-subjects analysis of variance (ANOVA) was conducted with time period (Aug19-Feb20, Aug20-Feb21, Aug21-Feb22) as the independent variable and average percentage of unauthorized absence for all pupils with and without CAMHS input as the dependent variable. The ANOVA indicated that there was a significant difference in average unauthorized absence F (2, 2451) = 5.096, p = .006. The reason for this significant effect was because unauthorized absence was considerably larger post-pandemic (M = .854, SE = .005) as explained by Bonferroni Post-hoc tests, compared to pre-pandemic epoch (M = .871, SE = .005, p = .049). Similarly, the average rate of unauthorized absence mid-pandemic (M = .875, SE = .006) was considerably larger compared to post-pandemic (p = .01).

Data gathered after full linkage identifier between GCC and CAMHS resulted in a count of; 83 known to CAMHS pre-pandemic, 72 known to CAMHS mid- pandemic and 71 known to CAMHS

post-pandemic.

Table 2.
Proportions of Pupils with more than 50% Unauthorised School Absences with CAMHS Service Contact

	Pre-Pandemic		**Mid-Pandemic**		**Post-Pandemic**	
	Contact with CAMHS		Contact with CAMHS		Contact with CAMHS	
	No	Yes	No	Yes	No	Yes
	n/%	n/%	n/%	n/%	n/%	n/%
Male	365 (48%)	35 (4.6%)	319 (46%)	30 (4%)	457 (46%)	32 (3%)
Female	312 (41%)	48 (6.3%)	303 (44%)	42 (6%)	472 (47%)	39 (4%)
Total	677 (89%)	83 (11%)	622 (90%)	72 (10%)	929 (93%)	71 (7%)

Note: CAMHS refers to those who had an appointment and/or referral into CAMHS. No CAMHS refers to those who did not have an appointment and/or referral into CAMHS.

The percentage of those with a high school absentee rate with no CAMHS input is higher pre-pandemic 89.1% as compared to post-pandemic 92.9%.

The chi-square test indicated a significant association between time period and CAMHS input across all three time periods x^2 (2) = 9.112, p = .01. As seen in Table 2. the percentage receiving CAMHS input varied across each time epoch, with a decrease of 4% in those having input from CAMHS and increase of 4% without CAMHS input.

Deprivation

The deprivation scale went from 1 to 5 with 1 being the most deprived areas in Scotland and 5 being the least deprived areas. Four postcodes did not have a deprivation number associated with them; therefore, they were excluded from this part of the analysis. Analysis of deprivation revealed that 72.4% of young people whose attendance fell below 60% were in the most deprived areas. While those in the least deprived areas only accounted for 1.7% of the 2454 young people provided by GCC.

Table 3.

Deprivation levels and frequency

	Frequency	*%*	*No CAMHS Input*	*CAMHS Input*	*CAMHS %*
Deprivation 1	1776	72.4	1617	159	9
Deprivation 2	376	15.3	343	33	8.8
Deprivation 3	177	7.2	156	21	11.9
Deprivation 4	79	3.2	70	9	11.4
Deprivation 5	42	1.7	38	4	9.5
Total	2450	99.8	2224	226	11.4

Additionally, while there is a steady increase of each level (approximately double) in young people going from Deprivation level 5 to 2, there is a large jump (approximately five times) from deprivation level 2 to level 1.

The chi-square test indicated no significant association between CAMHS contact and deprivation level x^2 (4) = 2.017, p = .733.

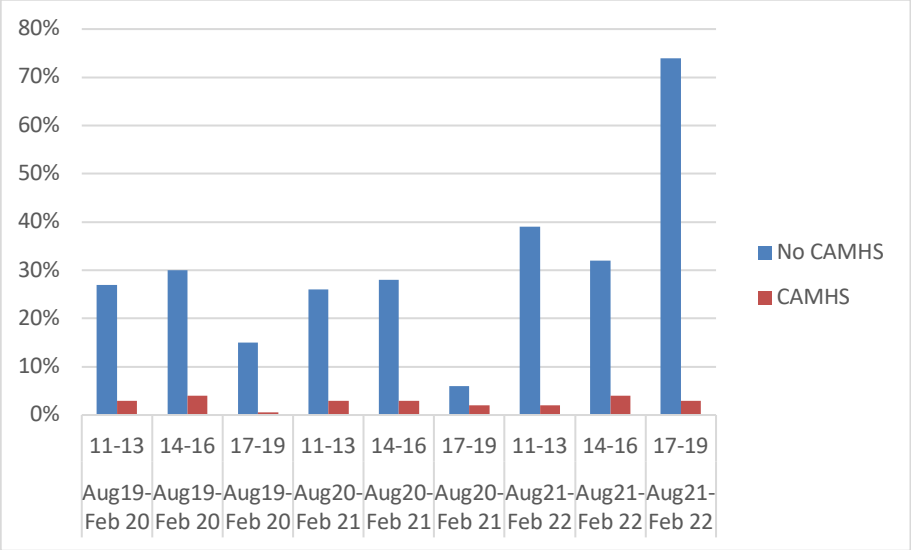
In order to determine whether there was a significant difference in unauthorized absences and deprivation a One-way between-subjects analysis of variance (ANOVA) was conducted, as the independent variable (deprivation status, five categories) and average percentage of unauthorized absence for all pupils with and without CAMHS input as the dependent variable. The ANOVA did not indicate a significant difference in average unauthorized absence among the deprivation groups, F (4, 2445) = 2.267, p = .06.

Age

There is a significant association between age and CAMHS input/time period, x^2 (10) = 130.260, p < .001. The numbers of those involved in CAMHS and not involved in CAMHS was consistent, pre and mid pandemic up until age 16. However, post-pandemic the proportion of 17-19 year old's who were not in contact with CAMHS is considerably higher from 14.8% to 74.3%.

Figure 1

Age, time period and CAMHS input



Sex

The data records comprised of a frequency of 1238 males (50.4%) and 1216 females (49.6%). To determine whether gender had an effect on CAMHS input between the time periods, separate Pearson Chi-Square tests of association were conducted using CAMHS input (i.e., had a referral and/or appointment) by time period for males and females. The test indicated that there was no significant association between CAMHS input and time period for males, x^2 (2) = 1.872, p = .392. However, the test indicated a significant association between CAMHS input and time period for females, x^2 (2) = 8.484, p = .014. CAMHS input for females; pre-pandemic 13%, mid-pandemic 12% and post-pandemic 8%. CAMHS input for males; pre-pandemic 9%, mid-pandemic 9% and post-pandemic 7%.

Discussion

The present study used administrative data on school attendance and CAMHS service contact to estimate rates of those at a higher risk of withdrawing to the extremes of Hikikomori in school age adolescents. Moreover, it compared rates of unauthorized absences across three main timepoints - pre, mid, and post COVID19 pandemic to compare rates of withdrawal across the time periods. Lastly, the study described profiles of withdrawal (i.e., withdrawal for >6 months with/without input from CAMHS) and explored whether unauthorized absences are associated with factors such as gender and socioeconomic factors i.e., living in a deprived postcode (based on the Scottish Index of Multiple Deprivation; SIMD, 2020).

Data extracted suggests that in the years across the pandemic the number of young people who are not in school nor are they in contact with CAMHS ranges between 622 and 929. This reflects a change in unexplained school absence in the post-pandemic period. Refining the methods for identifying true Hikikomori is needed, this could be done by reviewing the school attendance data of the 226 young people identified further to ascertain if there is any additional data on the duration, consistency, or previously unrecognised reasons for their absences. However, it appears that approximately 3% of young people in Glasgow are absent from school for more than 40% of the time and this isn't explained by a mental health condition that has put them in contact with CAMHS. This highlights a number of young people not being referred to CAMHS services and so a more robust way of identifying them is needed.

The data shows a change in unauthorised absenteeism post-pandemic compared to pre-pandemic, 760 pre-pandemic and 1000 post-pandemic. Without knowing whether the overall number of students changes, it is difficult to determine whether the numbers included in this study represent an increase in proportions or are proportional with changes in overall numbers of students. However, those with 100% unauthorized absence broken down over three time periods; pre-pandemic (August to February 2019-20) was 216, mid-pandemic (August to February 2020-2021) was 210, and post-pandemic (August to February 2021-2022) was 235. While these unauthorised absences will not all be explained by Hikikomori, some might be due to extreme social withdrawal. In order to improve the precision and usefulness of this data in the future, school records could be revisited to further characterise the patterns of absence (e.g., erratic attendance versus persistent non-attendance that lasts beyond the Hikikomori cut-off of 6 months). The data also highlighted a change in those young people being known to CAMHS with 11% known pre-pandemic and only 7% known to CAMHS post-pandemic. While this may be partially explained by an increase in referral numbers it is still notable that there

has been a change in CAMHS input. The combination of changes in school absenteeism and CAMHS input post-pandemic compared to pre-pandemic raises the question of the impact that the COVID-19 pandemic has had on young people withdrawing. While not all of these young people will progress to developing Hikikomori there is a potential for a number of them to progress to such an extreme withdrawal stage.

Deprivation

Analysis of the deprivation data revealed that 72.4% of young people whose attendance fell below 60% lived in the most deprived areas. Considering, risk of social withdrawal is associated with socioeconomic factors such as deprivation, this study supports current findings with young people in the most deprived area withdrawing from school and also unknown to CAMHS, 486 pre-pandemic, 461 mid-pandemic and 669 post-pandemic.

Research into socioeconomic factors for those who fit the Hikikomori profile in Japan found Hikikomori is more prevalent in middle- and upper-class families where parents have high levels of education (Umeda et al., 2012). This would suggest a greater vulnerability for those in the least deprived areas. Young people in the least deprived area only accounted for 1.7% of young people provided by GCC, yet 9.5% of them were known to CAMHS. This leaves 38 young people who are withdrawing from school and not involved with CAMHS. A descriptive feature of this group is that they lived in the least deprived post-codes, it will be for future studies to ascertain the relevance of this pattern to the development of extreme social withdrawal. Therefore, future research into youth at risk of developing Hikikomori could prioritise screening young people in the least deprived post-codes first.

For those young people where withdrawal from school is less likely to due to Hikikomori or living in the most deprived areas, their school withdrawal could be explained by NEET (Not in Education, Employment or Training). NEET is frequently recognized as a hopelessness to participate in education and employment, due to a low ability academically, lacking experience, and having low confidence (Thompson, 2011).

Age

Data showed that the numbers of young people with and without CAMHS input between the ages of 11 and 16 was consistent both pre and mid pandemic. Post-pandemic saw an increase in young people aged 11-16 without CAMHS input. However, the most notable finding was the number of 17-19 year old's post-pandemic, who were not involved in CAMHS, increased almost

fivefold from 15% to 74%. Similarly, the data showed a sixfold increase in CAMHS input for those aged between 17 and 19 from .5% pre pandemic to 3% post-pandemic. Therefore, regardless of CAMHS input, those between the ages of 17 and 19 years old have shown a change in unauthorized absences from school after the pandemic. Again, it is difficult to determine whether the numbers included in this study represent an increase in proportions or are proportional with changes in overall numbers of students. However, this finding highlight a potential impact of the pandemic on all age groups, particularly those aged between 17 and 19. Research into Hikikomori identified onset of Hikikomori being predominantly in late adolescence and disproportionately affects young adult males (Teo, 2009) this change of withdrawal post-pandemic for older adolescence could suggest a rise in Hikikomori post-pandemic. It must be noted however, that waiting times for NHS CAMHS access is a major challenge so reduced contact may be due to service issues (McNicholas et al., 2021). Also, proportions reported are skewed by the general increase in absenteeism.

Sex

The equal proportion of females to males seen in this study enables a greater generalisability to the wider school aged population. Moreover, it also supports previous finding from the UK Government's Department for Education (2018) that males and females have similar collective levels of absence. In this study sex was a statistically significant factor regarding differences of input/no input for CAMHS for females but was not statistically significant for males. It is thought that the difference in statistical significance of sex could be due to CAMHS input for females changing over the three time periods, while there was little variations for males.

Due to CAMHS reduced resources mid-pandemic and reduced access to services such as schools and GPs to identify a need for CAMHS involvement, it would be understandable that input from CAMHS may have reduced for both sexes. However, once services began to return it would seem females were more likely to seek help than males. Since adolescent to young adult males are at highest risk of withdrawing to the extremes of Hikikomori this finding is a concern for those males with both a low attendance rate and not receiving input from CAMHS. However, there is a possibility of delinquency (Blyth & Milner, 1999) rather than Hikikomori as an explanation for some of these young people. Without more fine-grained data on the patterns of school absence (e.g., erratic attendance vs. absence for 6 months or more) research cannot be certain what these young people are doing when they are not attending school.

As mentioned, Hikikomori is more predominant in middle- and upper-class families where

parents have high levels of education (Umeda et al., 2012), which in this study resulted in 38 young people being identified as having a high absentee rate in school, from the least deprived areas and not known to CAMHS. Of this 18 were female and 20 were male. This narrows down the number of individuals at a higher risk of withdrawing to the point of meeting the Hikikomori profile.

Consistent young people who are withdrawing

Surprisingly there were no young people in the data provided by GCC that were identified as meeting the unauthorized absence threshold in more than one time period. It would be reasonable to predict that if some young people early in their education had a high absentee rate during one time period that they would show a similar pattern the following year. Allowing for changes to a person's life improving/worsening and changing schools outside of GCC it could still be hypothesized that there would still be a number of young people who throughout their school years consistently have a high absentee rate. This leads to the question of school dropout and its current estimated rates.

Strengths & Limitations

This study is the first to attempt to estimate young people who may be a potential risk of withdrawing to the extreme of fitting the Hikikomori profile in a comprehensive sample of young people taken from a city wide public school network. It is hoped that this study will lead to further research into identifying those who are being missed by current services.

Another strength of this study is its utilisation of existing data. Data linkage enables researchers to harness and use huge quantities of preexisting data which continues to be collected and stored. This pre-existing data is beneficial in many areas of research. It has the ability to provide extensive data, link multiple data sources relating to an individual, an event or a location. Furthermore, it can aid pattern identification across whole populations and provide a broader picture, leading to an increased level of understanding. Studies can recognize previously undetected associations, give increased clarity and understanding than is feasible from the analysis of a single data source. Additionally, it is more cost-effective and efficient than it would be to collect the comparable data in other ways and is also less invasive. People who are socially withdrawn are very difficult to recruit into studies (almost by definition). By using administrative and health data sources we can begin to get a more precise estimate of the sub-population of interest (i.e., young people who are not in regular schooling and are not under the care of CAMHS). This data also increases feasibility of certain projects leading to better

health and wellbeing, service improvements and more-informed policies. Also, the process of data linkage can enable recognition and/or exclude irregularities for instance duplication, thus improving quality and accuracy of the data. For the specific population being examined here, data linkage not only increases access to a large number of difficult to reach young people, but it also creates opportunities to research this phenomenon.

A limitation of data linkage found in this study is in relation to unique identifiers across services. As unique identifiers do not exist for individuals in both CAMHS and GCC, data linking was undertaken by combining four variables to identify matches, but this approach can result in a considerable number of unmatched cases due to incomplete or missing data (see Table 3 in results section), and variations/errors. It is thought that future research might utilise less variables to identify matches. For instance, table 3 (results section) shows name and date of birth being the most reliable linkage variables. Additionally, variables used could be focused on ones that are less prone to error or changes, for instance date of birth does not change in comparison to address which might not get updated across all systems.

As previously noted whilst analysis addressed the study question, it is problematic to determine whether the numbers included in this study represent an increase in proportions or are proportional with changes in overall numbers of students. Further exploration of the data with exact student numbers for each time period; pre-, mid- and post- pandemic would enable an accurate comparison in regards to changes in withdrawal from school across the time epochs.

Findings are based on young people who have been identified by GCC as having attendance that falls below 60%. Of the absences, 50% or more were recorded as unauthorized absences. However, although attempting to increase specificity, due to the variation of how absences are coded in each school this research maybe overestimating the level of withdrawal in Glasgow as some absences maybe explained by other issues such as caring responsibilities. In order to improve estimation of withdrawal future research could follow up on those who have been identified in this study and a more precise number of young people be identified. Future research could also go further and identify young people who not only have a low attendance rate but also those who have dropped out of education completely.

Conclusion

One interpretation of the available analysed data is that 20 young males from the least deprived areas may potentially have an increased vulnerability to withdraw to a level consistent with Hikikomori. Considering the current study was conducted over a short time period and only looked at one area, it suggests that extreme social withdrawal throughout the United Kingdom could indicate a significant public health issue. The findings provide an insight into a potential impact of the COVID-19 pandemic on the withdrawal rates of young people, showing simultaneously changes post-pandemic, in those withdrawing from school and young people being known to CAMHS. Furthermore, the study illustrated multiple benefits to using existing data systems and collection processes in research, such as how existing data systems and collection processes can be used particularly to understand populations that are difficult to engage or to detect populations who are currently being missed by the normal safety nets available within health and education systems. It is hoped that this research could guide future research endeavors to further understand the phenomenon of Hikikomori in Scotland and inform how best to improve safety nets, help inform target delivery, improve early detection, and reduce the long-term impact for people with who are currently withdrawing themselves to the extreme socially.

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Appendices

Appendix 1:1 - Search Strategy Documentation Template

Search strategy for 1. PsycINFO via OVID, 2. Embase via OVID, 3. PubMed, 4. Web of Science Core Collection and 5. ASSIA via ProQuest using PICO framework

Database Name	Platform	Date Coverage	Date of Search	# of results
1. PsycINFO	Ovid	Not listed on database	21.04.23	419
2. Embase	Ovid	Not listed on database	21.04.23	273
3. PubMed		Not listed on database	21.04.23	192
4. Web of Science Core Collection		Not listed on database	21.04.23	478
5. ASSIA	ProQuest	Not listed on database	21.04.23	109

Total Records = 1471 **Total Records after deduplication = 845**

1. PsycINFO (Ovid)

Date of Search: 21.04.23 Number of results: 419

PsycINFO OVID	**Subject Headings**	**Title and Abstract Search Terms**
	social withdrawal	("Hikikomori" or "socially withdrawn youth" or "youth social withdrawal" or "severe social withdrawal" or "acute social withdrawal" or "protracted social withdrawal" or "prolonged social withdrawal" or "primary social withdrawal" or "hidden youth").tw

#	Search string	# of results
1	social withdrawal/	265
2	("Hikikomori" or "socially withdrawn youth" or "youth social withdrawal" or "severe social withdrawal" or "acute social withdrawal" or "protracted social withdrawal" or "prolonged social withdrawal" or "primary social withdrawal" or "hidden youth").tw.	231
3	1 or 2	419

2. EMBASE (Ovid)

Date of Search: 21.04.23 Number of results: 273

Embase OVID	**Subject Headings**	**Title and Abstract Search Terms**
	Hikikomori	("Hikikomori" or "socially withdrawn youth" or "youth social withdrawal" or "severe social withdrawal" or "acute social withdrawal" or "protracted social withdrawal" or "prolonged social withdrawal" or "primary social withdrawal" or "hidden youth").tw

#	Search string	# of results
1	Hikikomori	141
2	("Hikikomori" or "socially withdrawn youth" or "youth social withdrawal" or "severe social withdrawal" or "acute social withdrawal" or "protracted social withdrawal" or "prolonged social withdrawal" or "primary social withdrawal" or "hidden youth").tw.	262
3	1 or 2	273

3. PubMed

Date of Search:

21.04.23

Number of results:

192

PubMed	**Subject Headings**	**Title and Abstract Search Terms**
		("Hikikomori"[Title/Abstract] OR "socially withdrawn youth"[Title/Abstract] OR "youth social withdrawal"[Title/Abstract] OR "severe social withdrawal"[Title/Abstract] OR "acute social withdrawal"[Title/Abstract] OR "protracted social withdrawal"[Title/Abstract] OR "prolonged social withdrawal"[Title/Abstract] OR "primary social withdrawal"[Title/Abstract] OR "hidden youth"[Title/Abstract])

4. Web of Science Core Collection

Date of Search:

21.04.23

Number of results:

478

Web of Science Core Collection	**Subject Headings**	**Title and Abstract Search Terms**
	N/A	("Hikikomori" or "socially withdrawn youth" or "youth social withdrawal" or "severe social withdrawal" or "acute social withdrawal" or "protracted social withdrawal" or "prolonged social withdrawal" or "primary social withdrawal" or "hidden youth")

5. ASSIA (ProQuest)

Date of Search:

21.04.23

Number of results:

109

ASSIA via ProQuest	**Subject Headings**	**Title and Abstract Search Terms**
	N/A	("Hikikomori" or "socially withdrawn youth" or "youth social withdrawal" or "severe social withdrawal" or "acute social withdrawal" or "protracted social withdrawal" or "prolonged social withdrawal" or "primary social withdrawal" or "hidden youth")

Appendix 1.2 - Data Extraction Checklist

Extrapolate data from included studies related to;

a. Author
b. Year
c. Country
d. Study setting
e. Sample size
f. Age
g. Demographics (e.g., male/female)
h. Recruitment base
i. Sampling Method
j. Eligibility Criteria
k. Psychological Factors
l. Data Analysis/Statistical Methods
m. Main Findings

Appendix 1.3 – AXIS Quality Appraisal Tool

The final AXIS tool following consensus on all components by the Delphi panel

		Yes	No	Do not know/comment
Introduction				
1	Were the aims/objectives of the study clear?			
Methods				
2	Was the study design appropriate for the stated aim(s)?			
3	Was the sample size justified?			
4	Was the target/reference population clearly defined? (Is it clear who the research was about?)			
5	Was the sample frame taken from an appropriate population base so that it closely represented the target/reference population under investigation?			
6	Was the selection process likely to select subjects/participants that were representative of the target/reference population under investigation?			
7	Were measures undertaken to address and categorise non-responders?			
8	Were the risk factor and outcome variables measured appropriate to the aims of the study?			
9	Were the risk factor and outcome variables measured correctly using instruments/measurements that had been trialled, piloted or published previously?			
10	Is it clear what was used to determined statistical significance and/or precision estimates? (eg, p values, CIs)			
11	Were the methods (including statistical methods) sufficiently described to enable them to be repeated?			
Results				
12	Were the basic data adequately described?			
13	Does the response rate raise concerns about non-response bias?			
14	If appropriate, was information about non-responders described?			

		Yes	No	Do not know/comment
15	Were the results internally consistent?			
16	Were the results for the analyses described in the methods, presented?			
Discussion				
17	Were the authors' discussions and conclusions justified by the results?			
18	Were the limitations of the study discussed?			
Other				
19	Were there any funding sources or conflicts of interest that may affect the authors' interpretation of the results?			
20	Was ethical approval or consent of participants attained?			

Appendix 2.1 – REC/IRAS Ethical Approval Letters

West Midlands - Black Country Research Ethics Committee
The Old Chapel
Royal Standard Place
Nottingham
NG1 6FS

Appendix 2.2 –NHS GG&C Board/R&I Approval Letter

NHS
Greater Glasgow and Clyde

Research & Innovation
Dykebar Hospital, Ward 11
Grahamston Road
Paisley, PA2 7DE
Scotland, UK

Appendix 2.3 – Caldicott Approval Letter

Data Protection Officer
Information Governance Department
NHS Greater Glasgow & Clyde
2nd Floor, 1 Smithhills Street
Paisley PA1 1EB

Date: 27/02/2023

Appendix 2.4 – MRP Proposal

Final Approved MRP Proposal can be accessed at the following link;

https://osf.io/ks25g/

Appendix 2.5 – JARS–Quant Checklist

APA Style **JARS**
Journal Article Reporting Standards

JARS–**Quant** | Table 1
Information Recommended for Inclusion in Manuscripts
That Report New Data Collections Regardless of Research Design

Title and Title Page

Title

- Identify main variables and theoretical issues under investigation and the relationships between them.
- Identify the populations studied.

Author Note

- Provide acknowledgment and explanation of any special circumstances, including
  - registration information if the study has been registered
  - use of data also appearing in previous publications
  - prior reporting of the fundamental data in dissertations or conference papers
  - sources of funding or other support
  - relationships or affiliations that may be perceived as conflicts of interest
  - previous (or current) affiliation of authors if different from location where the study was conducted
  - contact information for the corresponding author
  - additional information of importance to the reader that may not be appropriately included in other sections of the paper

Abstract

Objectives

- State the problem under investigation, including main hypotheses.

Participants

- Describe subjects (nonhuman animal research) or participants (human research), specifying their pertinent characteristics for the study; in animal research, include genus and species. Participants are described in greater detail in the body of the paper.

Study Method

- Describe the study method, including
  - research design (e.g., experiment, observational study)
  - sample size
  - materials used (e.g., instruments, apparatus)
  - outcome measures
  - data-gathering procedures, including a brief description of the source of any secondary data. If the study is a secondary data analysis, so indicate.

Findings

- Report findings, including effect sizes and confidence intervals or statistical significance levels.

Conclusions

- State conclusions, beyond just results, and report the implications or applications.

Introduction

Problem

- State the importance of the problem, including theoretical or practical implications.

Review of Relevant Scholarship

- Provide a succinct review of relevant scholarship, including
  - relation to previous work
  - differences between the current report and earlier reports if some aspects of this study have been reported on previously

Hypothesis, Aims, and Objectives

- State specific hypotheses, aims, and objectives, including
  - theories or other means used to derive hypotheses
  - primary and secondary hypotheses
  - other planned analyses
- State how hypotheses and research design relate to one another.

Method

Inclusion and Exclusion

- Report inclusion and exclusion criteria, including any restrictions based on demographic characteristics.

Participant Characteristics

- Report major demographic characteristics (e.g., age, sex, ethnicity, socioeconomic status) and important topic-specific characteristics (e.g., achievement level in studies of educational interventions).
- In the case of animal research, report the genus, species, and strain number or other specific identification, such as the name and location of the supplier and the stock designation. Give the number of animals and the animals' sex, age, weight, physiological condition, genetic modification status, genotype, health–immune status, drug or test naiveté, and previous procedures to which the animal may have been subjected.

Sampling Procedures

- Describe procedures for selecting participants, including
  - sampling method if a systematic sampling plan was implemented
  - percentage of sample approached that actually participated
  - whether self-selection into the study occurred (either by individuals or by units, such as schools or clinics)
- Describe settings and locations where data were collected as well as dates of data collection.
- Describe agreements and payments made to participants.
- Describe institutional review board agreements, ethical standards met, and safety monitoring.

Sample Size, Power, and Precision

- Describe the sample size, power, and precision, including
  - intended sample size
  - achieved sample size, if different from the intended sample size
  - determination of sample size, including
    - › power analysis, or methods used to determine precision of parameter estimates
    - › explanation of any interim analyses and stopping rules employed

Measures and Covariates

- Define all primary and secondary measures and covariates, including measures collected but not included in the report.

Data Collection

- Describe methods used to collect data.

Quality of Measurements

- Describe methods used to enhance the quality of measurements, including
  - training and reliability of data collectors
  - use of multiple observations

Instrumentation

- Provide information on validated or ad hoc instruments created for individual studies, for individual studies (e.g., psychometric and biometric properties).

Masking

- Report whether participants, those administering the experimental manipulations, and those assessing the outcomes were aware of condition assignments.
- If masking took place, provide a statement regarding how it was accomplished and whether and how the success of masking was evaluated.

Psychometrics

- Estimate and report values of reliability coefficients for the scores analyzed (i.e., the researcher's sample), if possible. Provide estimates of convergent and discriminant validity where relevant.
- Report estimates related to the reliability of measures, including
  - interrater reliability for subjectively scored measures and ratings
  - test–retest coefficients in longitudinal studies in which the retest interval corresponds to the measurement schedule used in the study
  - internal consistency coefficients for composite scales in which these indices are appropriate for understanding the nature of the instruments being used in the study
- Report the basic demographic characteristics of other samples if reporting reliability or validity coefficients from those samples, such as those described in test manuals or in norming information for the instrument.

Conditions and Design

- State whether conditions were manipulated or naturally observed. Report the type of design as per the JARS–Quant tables:
  - experimental manipulation with participants randomized
    - › Table 2 and Module A
  - experimental manipulation without randomization
    - › Table 2 and Module B
  - clinical trial with randomization
    - › Table 2 and Modules A and C
  - clinical trial without randomization
    - › Table 2 and Modules B and C
  - nonexperimental design (i.e., no experimental manipulation): observational design, epidemiological design, natural history, and so forth (single-group designs or multiple-group comparisons)
    - › Table 3
  - longitudinal design
    - › Table 4
  - *N*-of-1 studies
    - › Table 5
  - replications
    - › Table 6
- Report the common name given to designs not currently covered in JARS–Quant.

Data Diagnostics

- Describe planned data diagnostics, including
  - criteria for post-data-collection exclusion of participants, if any
  - criteria for deciding when to infer missing data and methods used for imputation of missing data
  - definition and processing of statistical outliers
  - analyses of data distributions
  - data transformations to be used, if any

Analytic Strategy

- Describe the analytic strategy for inferential statistics and protection against experiment-wise error for
  - primary hypotheses
  - secondary hypotheses
  - exploratory hypotheses

Results

Participant Flow

- Report the flow of participants, including
  - total number of participants in each group at each stage of the study
  - flow of participants through each stage of the study (include figure depicting flow, when possible; see the JARS–Quant Participant Flowchart)

Recruitment

- Provide dates defining the periods of recruitment and repeated measures or follow-up.

Statistics and Data Analysis

- Provide information detailing the statistical and data-analytic methods used, including
  - missing data
    - › frequency or percentages of missing data
    - › empirical evidence and/or theoretical arguments for the causes of data that are missing—for example, missing completely at random (MCAR), missing at random (MAR), or missing not at random (MNAR)
    - › methods actually used for addressing missing data, if any
  - descriptions of each primary and secondary outcome, including the total sample and each subgroup, that includes the number of cases, cell means, standard deviations, and other measures that characterize the data used
  - inferential statistics, including
    - › results of all inferential tests conducted, including exact *p* values if null hypothesis significance testing (NHST) methods were used, and reporting the minimally sufficient set of statistics (e.g., *df*s, mean square [*MS*] effect, *MS* error) needed to construct the tests
    - › effect-size estimates and confidence intervals on estimates that correspond to each inferential test conducted, when possible
    - › clear differentiation between primary hypotheses and their tests–estimates, secondary hypotheses and their tests–estimates, and exploratory hypotheses and their test–estimates

Statistics and Data Analysis (*continued*)

  - complex data analyses—for example, structural equation modeling analyses (see also Table 7), hierarchical linear models, factor analysis, multivariate analyses, and so forth, including
    - › details of the models estimated
    - › associated variance–covariance (or correlation) matrix or matrices
    - › identification of the statistical software used to run the analyses (e.g., SAS PROC GLM or the particular R package)
  - estimation problems (e.g., failure to converge, bad solution spaces), regression diagnostics, or analytic anomalies that were detected and solutions to those problems.
  - other data analyses performed, including adjusted analyses, if performed, indicating those that were planned and those that were not planned (though not necessarily in the level of detail of primary analyses).
- Report any problems with statistical assumptions and/or data distributions that could affect the validity of findings.

Discussion

Support of Original Hypotheses

- Provide a statement of support or nonsupport for all hypotheses, whether primary or secondary, including
  - distinction by primary and secondary hypotheses
  - discussion of the implications of exploratory analyses in terms of both substantive findings and error rates that may be uncontrolled

Similarity of Results

- Discuss similarities and differences between reported results and work of others.

Interpretation

- Provide an interpretation of the results, taking into account
  - sources of potential bias and threats to internal and statistical validity
  - imprecision of measurement protocols
  - overall number of tests or overlap among tests
  - adequacy of sample sizes and sampling validity

Generalizability

- Discuss generalizability (external validity) of the findings, taking into account
  - target population (sampling validity)
  - other contextual issues (setting, measurement, time; ecological validity)

Implications

- Discuss implications for future research, program, or policy.

Appendix 2.6 – ISA Glasgow City Council

Data Sharing Agreement can be accessed at the following link;

https://osf.io/ks25g/