#### Psychopathology profiles in children with congenital visual impairment

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#### Contains: Executive summary, systematic review, empirical paper, integration, impact and dissemination, references, appendices.

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1. Executive Summary
   1. Background

* Improving mental health in children is an international (World Health Organisation) and UK governmental priority (Department of Health and Department for Education, 2017).
* Children with paediatric disorders are at increased risk of psychopathology (Royal College of Paediatrics and Child Health, 2018).
* Congenital visual impairment (VI) has a major impact on development, learning and autonomy, and children with VI may be at increased risk of psychopathology.
* An informed understanding of psychopathology profiles in children with VI is required, to plan appropriate support and intervention.
  1. Previous Research

Previous studies have produced mixed findings regarding psychopathology symptoms and associated risk and protective factors, within children with VI. Mixed findings may be related to methodological limitations:

* VI is an extremely heterogeneous condition that is frequently comorbid with Intellectual Disability (ID) and other neurological impairments. Many previous studies have included children with known comorbid impairment, leading to potentially confounded and contradictory findings.
* Most studies are limited to specific geographical areas and have recruited from specialist VI schools only, which may reduce generalisability. Children who attend specialist schools may have additional needs compared to children who attend mainstream schools, though educational inclusion policies vary between countries.
* The vision characteristics (e.g. severity of VI, type of vision disorders) of samples often lack sufficient detail, limiting replicability and generalisability.
* Samples often include a broad age range spanning multiple developmental periods, so findings may be confounded by age related developmental changes.
* Previous studies have used a wide variety of measures of psychopathology, which may also explain the mixed findings.
* There has been a lack of multivariate analysis to investigate the relative importance of risk and protective factors for psychopathology, in children with VI.

* 1. Project Aims

The aim of this project was to increase current understanding of psychopathology profiles and associated risk and protective factors, in children with VI.

* + 1. Systematic review research question:

To inform the design of the study, a systematic review of the available literature was undertaken. The key question addressed was:

Which risk and protective factors are associated with psychopathology in children and adolescents with VI?

* + 1. Empirical study research questions:

Drawing on the systematic review findings, the empirical study set out to answer the following questions:

1. Are school aged children with VI at increased risk of psychopathology compared to typically developing (TD) children and what patterns of psychopathology are found in this group?
2. How do parent report and child self-report of internalising symptoms and quality of life, compare in children with VI?
3. Are children who are ‘blind’ at higher risk of psychopathology than children who are less visually impaired and have ‘low vision’?
4. How do the potential risk factors of more severe VI, gender, lower socioeconomic status, social communication difficulties and lower adaptive functioning relate to psychopathology patterns in children with VI?
   1. Methodology
      1. Systematic Review.

A systematic review of articles published between 1984 and 2018, investigating risk or protective factors associated with psychopathology in children with VI, was conducted.

* + - 1. Inclusion Criteria.
* VI sample aged 4-19 years.
* At least one measure of internalising or externalising symptoms.
* At least one research question related to possible risk or protective factors.
* Correlational, controlled and uncontrolled studies.
  + - 1. Exclusion Criteria.
* Unpublished studies.
* Grey literature.
* Qualitative design.
* Case Studies.
* Full text not available in English.
* Total sample has additional comorbid disabilities.
  + 1. Procedure

Studies were identified by searching two major electronic psychology and clinical databases: PSYCHOINFO and MEDLINE. Relevant data was extracted, and a narrative synthesis was written.

* + 1. Empirical Study
       1. Inclusion Criteria.
* Children aged 8 to 11.11 years.
* ‘Low vision’ (logMAR 0.5 to 1.3) or ‘blindness’ (logMAR 1.4 or worse), according to the International Classification of Diseases 2010 (ICD-10).
* Congenital VI originating in ophthalmological disorders, referred here as ‘potentially simple’ Congenital Disorders of the Peripheral Visual System (CDPVS).
  + - 1. Exclusion Criteria.
* Cortical visual disorders (CVI), which originate in the brain.
* ‘Potentially Complicated’ CDPVS, with known additional neurological involvement.
* Comorbid ID, hearing impairment or motor impairment.
* Insufficient English to complete questionnaire measures.
  + 1. Procedure
* Participants were recruited through two national tertiary paediatric ophthalmological services in London.
* Recruitment took place from September 2017 to February 2018.
* Parents (*n*=34) completed a questionnaire battery of standardised measures (assessing internalising and externalising symptoms, social communication skills, adaptive functioning, and quality of life) to investigate psychopathology profiles in children.
* Children completed related self-report questionnaires which assessed internalising symptoms and quality of life.
* Medical and demographic information were obtained, from parent report and medical records.
* A cross-sectional within-group design was used to examine psychopathology profiles in the sample and to investigate potential associated risk factors.
* Between-group analyses were used to compare the sample with normative childhood population scores.
  1. Findings
     1. Systematic Review
* 18 publications met the inclusion criteria.
* Risk and protective factors for psychopathology were identified in different studies, but findings were often contradictory between studies or required replication.
* The most frequently studied risk factors were gender, severity of VI and cognition.
* No unique risk or protective factors for psychopathology were found for children who have VI, compared to sighted samples.
* The only consistent risk factor for psychopathology identified in this review was ID.
* Mixed findings may have been related to methodological differences between studies.
* There was a lack of focus on protective factors in the literature.
* The overall quality of articles was ‘moderate’.
  + 1. Empirical Study.
* Children with VI in the sample were found to be at increased risk of psychopathology, compared to TD peers.
* 11 children (33%) were at ‘high risk’ of an internalising disorder and 7 children (21.2%) were at ‘high risk’ of an externalising disorder, compared to 10% of the TD population.
* 10 children (29%) scored in the clinical range for separation anxiety.
* Risk factors found to be associated with psychopathology were social communication difficulties, more severe VI, and adaptive functioning difficulties.
* Gender, socioeconomic status, and age were not associated with psychopathology symptoms in the sample.
* Social communication accounted for 56% of the variance in psychopathology symptoms.
* There was a moderate agreement between parent report and child self-report.
  1. Strengths and Limitations
     1. Systematic Review.
        1. Strengths.
* First systematic review of risk and protective factors for psychopathology, in children with VI.
* Comprehensive search strategy, including supplementary hand searching.
* Focus on psychopathology rather than broader constructs of mental health.
* Good agreement between two raters in relation to the applied search strategy.
* Quality assessment of all included studies.
  + - 1. Limitations.
* Exclusion criteria may have reduced representativeness.
* Included studies used different inclusion criteria, so the samples (e.g. VI severity, comorbid impairment) varied considerably, which may have impacted validity.
* Broad age range may have affected the validity of findings.
  + 1. Empirical Study
       1. Strengths.
* Inclusion limited to children who had ‘potentially simple’ CDPVS and ‘low vision’ or ‘blindness’ (ICD-10) with no known additional comorbidities. This enabled a more clearly specified investigation of the relationship between VI and psychopathology.
* Recruitment from two paediatric ophthalmological departments and the use of detailed medical report ophthalmic information increased, clarity of visual acuity bandings.
* Focus on middle childhood, which has been under researched in VI studies.
* Valid and reliable standardised measures that are used to assess psychopathology.
* Comparison of parent report and child self-report, to investigate reliability of perspective and ratings.
* Appropriate statistical methods, including multivariate regression analyses.
* Post hoc power analyses indicated that the analyses were appropriately powered to detect large effects.
  + - 1. Limitations.
* Target sample size (*n*=50) was not reached (68% attained) due to time constraints.
* There was a risk of Type 1 (false positive) errors due to multiple comparisons, though Bonferonni corrections were applied.
* The design did not permit consideration of causality.
* Positively skewed socioeconomic status and low response rate may have led to a sample bias and therefore reduced representativeness.
* Differences in data collection between children who were ‘blind’ (oral administration by parents) and those with ‘low vision’ (self-completed using enlarged font) may have impacted on the validity of data, due to social desirability effects.
  1. Implications
* Children in the sample were particularly at risk of separation anxiety and interventions may need to be offered to target internalising symptoms for some children with VI.
* Children with VI should be regularly and routinely screened for psychopathology symptoms.
* Parents and professionals may benefit from psychoeducation about potential psychopathology symptoms in children with VI.
* Where there are concerns, the child should have timely access to intervention.
* More priority for children with VI may be needed at a service commissioning and policy level, in the context of limited Child and Adolescent Mental Health Services (CAMHS).
* The most effective kind of service delivery that is also cost effective may need further consideration, such as a National Blind CAMHS (along the lines of the national ‘deaf’ CAMHS).
* ID, more severe VI (‘blindness’), adaptive functioning difficulties and social communication difficulties may all be risk factors for psychopathology in children with VI.
* Interventions to promote adaptive functioning and social communication skills in children with VI may be helpful.
* Early identification of ASD in children with VI might lead to improved psychological outcomes, particularly in relation to externalising disorders
  1. Future Research
* Clarity and consistency of sampling methodology and measurement tools is needed to improve reliability and consensus across research.
* More multivariate research using longitudinal designs are needed to understand the relative importance of risk and protective factors in children with VI.
* Future research is needed to develop and test theoretical VI specific models, to further understand risk and protective factors related to psychopathology in this group.
* Future research is particularly needed to investigate the role of protective factors, in relation to parental and family factors.
* Future research could investigate whether more secure attachments and reduced separation anxiety can be supported by targeting parenting style and the development of child independence skills.
* Programmes to support the development of social communication skills in children with VI may be worth exploring to see if this reduces the risk of externalising symptoms.
* Future research is needed to examine how to tailor preventative and intervention approaches to reduce or ameliorate psychopathology symptoms in children with VI.
  1. Impact

The main impact is to assist children with VI and their families by increasing the recognition of their needs. This may lead to a greater recognition of these needs, within health and education services. The scope of the impact is that professionals supporting children with VI will be informed by the research, which may be used to inform practice inputs. Findings may inform both a psychoeducation approach and VI specific interventions, based on identified need and risk factors. The practical yield of the research for families and practitioners could be maximised by using a consultation model, and auditing in relation to changes in service protocols and understanding of child mental health, in this population.

* 1. Dissemination
* The systematic review and the empirical study are being prepared for publication. They are planned to be submitted to the Developmental Medicine and Child Neurology journal or the Journal of Consulting and Clinical Psychology.
* The research will be presented (oral and poster presentation) at the Mary Kitzinger Trust (GOSH/UCL) conference, London, July 2018. The title of the conference is: ‘Childhood visual impairment and mental health: Science into practice’.
* An abstract has been submitted to the British Psychological Society, Faculty for Children, Young People, and their Families Annual Conference, Liverpool, September 2018.
* Findings will also be disseminated via presentation to clinicians who work with children who have VI, in the services involved with the project.
* A written summary of findings will also be produced to highlight implications and recommendations and will be published on the affiliated research website.
* Findings will also be disseminated to families who took part in the project, in an end of project report.
* Findings may also be disseminated to other families of children who have VI, via an online blog.
  1. Conclusion
* Findings add to the growing evidence base that children with VI are at increased risk of psychopathology.
* The systematic review findings indicated that ID is a risk factor of psychopathology in children with VI. There was a lack of consensus regarding other risk factors and a lack of research investigating protective factors.
* Some potentially important risk factors (e.g. socioeconomic status, autism traits, adaptive functioning) have been under-researched and so were addressed in the empirical study, within a multivariate design.
* The systematic review identified the methodological limitations of previous research and these were addressed in the empirical study. Only children who had ‘potentially simple’ CDPVS with banding of VI according to ICD-10 criteria, and no additional known comorbidities, were included.
* Children in the sample were at increased risk of psychopathology compared to TD peers. They were particularly at risk of separation anxiety.
* The most severe level of VI (‘Blindness’), reduced adaptive functioning skills, and weaker social communication skills were possible risk factors for psychopathology in the sample.
* Gender, socioeconomic status, and age were not associated with psychopathology in the sample.
* Social communication explained 56% of the variance in psychopathology in the sample and particularly explained the variance in externalising symptoms.
* Early parent-child relationships may be affected by VI and families may require support in developing secure attachment styles.
* Future research is needed to investigate the efficacy of preventative and interventionist approaches to psychopathology in children with VI.

1. Risk and Protective Factors for Psychopathology in Children and Adolescents with Visual Impairment: A Systematic Review
   1. Abstract

This is the first systematic review of risk and protective factors associated with psychopathology in children with visual impairment (VI). It builds on a recent systematic review that concluded that children with VI were at higher risk of psychopathology, compared to typically developing children (Augestad, 2017).

A systematic review of articles published between 1984 and 2018, investigating risk and protective factors associated with psychopathology in children with VI, was conducted. Studies were identified by searching two major electronic psychology and clinical databases: PSYCHOINFO and MEDLINE. A total of 18 publications met the inclusion criteria, of which 15 had a cross-sectional design.

No unique risk or protective factors for psychopathology were found for children with VI, compared to typically developing peers. The most frequently studied risk factors were gender, severity of VI, and cognitive ability. Co-occurring Intellectual Disability (ID) was found to be a risk factor for psychopathology. There was limited research investigating potential protective factors. The overall quality of articles was ‘moderate’ in relation to the developed criteria for quality research with VI samples.

This review only identified ID as a risk factor for psychopathology in children with VI. Group heterogeneity was concluded to be an important limitation of previous research. Multivariate research within valid samples is needed to examine the relative importance of other potentially important risk factors, such as autism traits and adaptive functioning ability. Future research is needed to investigate potential protective factors, such as parental and family functioning. A cumulative risk approach could be a helpful framework for understanding the high incidence of psychopathology in children who have VI.

* 1. Introduction

Improving mental health in children is an international (World Health Organisation; WHO) and UK governmental priority (Department of Health and Department for Education, 2017). In the UK, ten percent of children aged 5-16 years have a diagnosable mental health disorder (Green, McGinnity, Meltzer, Ford & Goodman, 2004). Children with paediatric disorders, such as Intellectual Disability (ID) (Allington-Smith, 2006), epilepsy (Reilly, Agnew & Neville, 2011), and chronic kidney disease (Assadi, 2013) are at elevated risk of mental health difficulties (Royal College of Paediatrics and Child Health; RCPCH, 2018). International research suggests that children who have visual impairment (VI) may also be at elevated risk of mental health difficulties, compared to typically developing (TD) peers (Augestad, 2017).

VI is defined as vision acuity (the finest detail that can be seen) within categories of ‘low vision’ (logMAR 0.5 or worse) or ‘blindness’ (logMAR 1.4 or worse) (WHO; ICD-10). Based on this criterion, it is estimated that 2 out of every 1,000 individuals under the age of 25 have VI, in the UK. Four out of every 10,000 children in the UK are ‘blind’ by age one (Rahi & Cable, 2003; Cumberland, Pathai & Rahi, 2010). In line with WHO recommendations, many studies investigating psychological distress in children with VI have focused on broad concepts, such as psychological wellbeing and quality of life.

Whilst this research is valuable, critics have highlighted that an exploration of psychopathology allows for a more targeted focus on specific symptoms, allowing for more targeted screening and intervention (Martikainen, Bartley & Lahelma, 2002). The two main presentations of psychopathology in childhood are ‘internalising’ and ‘externalising’ disorders (Patalay et al., 2015). Internalising symptoms consist of excessive control over one’s behaviours, emotions, and thoughts, whereas externalising symptoms consist of a lack of control over these (Nunes, Faraco, Vieira & Rubin, 2013). Internalising psychopathology may be expressed in the form of depression or anxiety, whereas externalising psychopathology may be expressed by aggressive, impulsive, and challenging behaviours. Both forms of psychopathology may lead to limited social experience, creating obstacles for psychological development.

As children with VI may be at increased risk of childhood psychopathology and because this is highly predictive of psychiatric disorder later in life (Blanken et al., 2018), an understanding of risk and protective factors specific to children with VI may be needed to aid early identification and intervention. In the literature on sighted children, Zeanah, Boris and Larrieu (1997) stated that risks factors for psychopathology in children could be divided into three categories: 1) risks in the socioeconomic context, 2) risks in the family, and 3) risks related to the child. Protective factors in these areas are likely to buffer against risks.

Research suggests that most children are exposed to a single physical or psychosocial risk factor in early life, without significant consequence (Ogg et al., 2010). Exposure to multiple risk factors is much more predictive of psychopathology and the cumulative risk approach has been helpful in explaining psychopathology in children (Evans & King, 2013). There may also be a complex interplay between risk factors and moderating and mediating variables, which may play important toles in developmental trajectories to psychopathology (Breitborde, Srihari, Pollard, Addington & Woods, 2010).

A recent systematic review investigated the risk factors for psychopathology in young children (Carneiro, Dias & Soares, 2016). The review was rigorous and included empirical studies (>*n* 100) that used the Child Behaviour Checklist (Achenbach, 1983; CBCL) – a reliable measure of child psychopathology, that focused on TD children. In terms of socioeconomic context risks, the authors concluded that lower socioeconomic status was highly associated with increased psychopathology symptoms (Ronan, Canoy & Burke, 2009). In terms of family risks, lack of parental education (Velders et al., 2011), maternal psychopathology (Batenburg-Eddes et al., 2013), and parental domestic violence (Goodman, Slobodskaya & Knyazev, 2005) were all associated with higher levels of child psychopathology.

In terms of risks related to the child, males have been found to be at higher risk of externalising symptoms (e.g. Twomey et al., 2013) and females have been found to be at higher risk of internalising symptoms (Pathak et al., 2011). Low emotional control (e.g. Ezpeleta, Granero, De la Osa, Penelo & Domenech, 2012), lower IQ, and poorer academic ability (Goodman et al., 2005; Sameroff, Seifer, Barocas, Zax & Greenspan, 1987) are also risk factors. Conversely, psychological attributes of higher self-efficacy, self-esteem, and optimism have been found to be protective factors (Young, Hanson, Craig, Clapham & Williamson, 2017).

Neurodevelopmental disorders such as Autism Spectrum Disorder (ASD) are also highly associated with psychopathology (Bora & Berk, 2016). ASD is characterized by impairments in social (social interaction and communication) and non-social (restricted and repetitive behaviours) domains (DSM-V; ICD-10). Poor social skills, which are frequently associated with neurodevelopmental disorders, are also associated with psychopathology (Ratcliffe, Wong, Dossetor & Hayes, 2015).

* + 1. Psychopathology in Children With VI

Psychological development may be impacted by both the functional and physical limitations of VI and by atypical treatment by other members of society (Wallander & Varni, 1998). A diathesis-stress model postulates that distress arises when there is an imbalance between perceived competency and environmental demands. The risk of an imbalance between these two factors may be higher in children with VI (Ammerman, Van Hasselt, Hersen & Moore, 1989). Higher levels of dependency upon others, increased parental control, negative thoughts about physical appearance, discrimination, developmental delays, and the impacts of comorbid neurological impairments, may interact and contribute to psychological distress (Hurre & Aro, 2000; Lifshitz, Hen & Weisse, 2007).

Qualitative research has provided insights into the mental health of children with VI. In one study, 16 children with VI aged 9 to 13 years, participated in two focus group interviews. Findings indicated that children with VI were particularly anxious and worried about being excluded from activities, being teased by peers and about coming to physical harm as a result of their VI (Visagie, Loxton, Stallard & Silverman, 2017). Tadic, Cooper, Lewando-Hundt, Keeley and Rahi (2014) conducted semi-structured interviews with 32 children with VI aged 10-15 years. Children and adolescents with VI discussed the importance of family and peer support, difficulties balancing independence, support and safety, the emotional burden of living with a disability, concerns about education and job prospects in the future and functional restrictions and limitations. A common theme in qualitative research has been the difficulty balancing independence, support and safety (Khadka, Ryan, Margrain, Woodhouse & Davies, 2012; Tadic et al., 2014; Rainey et al., 2016). Rainey et al., 2016 found that the views of professionals and parents do not always correspond to the perspective of children and adolescents with VI. This indicates the need to include the perspective of the child in research.

The findings of empirical research into psychopathology in children with VI is mixed, with consensus between certain studies and contradictory results in others. Bakhla, Sinha, Verma and Sarkhel (2011) found that children with VI in specialist schools in India were at lower risk of psychiatric disorder compared to TD peers. Harris and Lord (2016) found the opposite in children with VI in the UK, who attended a range of educational provisions. It may be that better support systems or more realistic environmental demands are operating for children with VI attending specialist schools, compared to mainstream schools. This may vary between countries due to different service and cultural contexts.

Augestad (2017) conducted a systematic review of mental health in children with VI. Seventeen studies were included in the review, which represented 13 countries. The sample size ranged from 22 to 189 and studies included children aged six to 22 years. Eleven studies examined general psychopathology, and of these, seven found that children with VI had more emotional problems than TD peers (Brunes, Flanders & Augestad, 2015; Harris & Lord, 2016; Huurre & Aro, 1998, 2000; Pinquart & Pfeiffer, 2012, 2014; Visagie, Loxton & Ollendick, 2013) and four did not (Huurre & Aro, 1998; Huurre, Komulainen & Aro, 2001; Konarska, 2005; Yoshida, Ichikawa, Ishikawa & Hori, 1998). There does not appear to be any clear methodological differences to explain the mixed findings.

Seven studies, out of the 17 included the systematic review, investigated depression. Four studies found increased rates of depression in children with VI compared to TD peers (Garaigordobil & Bernaras, 2009; Huurre & Aro, 2000; Koenes & Karshmer, 2000; Konaeska, 2007) and three studies found no differences between groups (Bolat et al., 2011; Huurre & Aro, 1988; Yoshida et al., 1998). Finally, five studies investigated anxiety, three of which found increased rates in children with VI compared to TD peers (Bolat et al., 2011; Garaigordobil & Bernaras, 2009; Konarska, 2007) and two studies found no differences between groups (Halder & Datta, 2012; Yoshida et al., 1998). Again, there did not appear to be any clear methodological differences to explain these mixed findings.

It may be that the mixed findings could be explained by sample differences, which are not always clearly outlined. Childhood VI is extremely heterogeneous: there is a high level of comorbidity with other conditions and differing severity of VI, which may lead to different levels of functional impact. It is therefore important that studies define the sample sufficiently, so that generalisability can be assessed. This has often not been the case in research to date. For instance, within Augestad’s (2017) review, some studies include samples of children who have significant comorbidities (e.g. ID) and the severity of VI is often poorly defined and differs between studies. This lack of consistency and clarity across samples limits the opportunity for cross-review of studies and makes it increasingly challenging to reliably identify critical factors.

The systematic review concluded that despite mixed findings, children and young people who have VI are at higher risk of mental health disorder than TD peers (Augestad, 2017). Most of the included studies referred to adolescents only. The overall quality of studies included in the systematic review were coded as ‘good’, though the quality assessment tool was not adapted to measure issues specific to VI research. This may have led to an overestimation of quality within the review. Further research is needed to explore the roles of risk and protective factors associated with psychopathology in children with VI. Increased knowledge about risk and protective factors may help to understand mixed findings and to tailor appropriate intervention.

* + 1. Risk and Protective Factors in Children and Adolescents Who Have VI
       1. Risks in the socioeconomic context.

Socioeconomic disadvantage is associated with psychopathology in the general population (Green, McGinnity & Meltzer, 2005). One study included in Augestad’s (2017) systematic review considered the role of socioeconomic status and found that there was no association with psychiatric disorder in adolescents with VI (Bakhla et al., 2011). Whilst socioeconomic status is sometimes reported in VI studies, it is rarely investigated in relation to psychopathology symptoms. Little is known about how the socioeconomic context relates to psychological distress in this population.

* + - 1. Risks in the family.

Negative parenting and family issues may be risk factors for children with VI as some parents may have greater difficulties interacting with and adapting to their child in the absence of vision mediated interaction and communication, in the early years (Howe, 2006). A recent study found that 35% of mothers of very young children with VI scored in the clinical range for parenting stress, compared with the known norm of 15% in the broader community, at a similar age (Sakkalou, Sakki, O’Reilly, Salt & Dale, 2017). As parental mental health is associated with psychopathology in the general population (Batenburg-Eddes et al., 2013), parenting issues are likely to be influential in the psychological development of children with VI. Broader family functioning is associated with psychopathology in the TD population (Bogels & Brechman-Toussaint, 2006) but this has not yet been investigated within the childhood VI population.

* + - 1. Risks related to the child.

Social difficulties are common in children with VI (e.g. Aro & Huurre, 2000; Pinquart & Pfeiffer, 2011). Functional difficulties (e.g. a lack of visual information about facial expressions) in predicting the social intentions of others (Pinquart & Pfeiffer, 2013), may lead to greater difficulties in interacting. Coupled with higher levels of dependency on adults (Sacks, Kekilis & Garlord-Ross, 1992), there may be fewer opportunities for socialisation and increased levels of loneliness (Hadidi & Al Khateeb, 2013).

Autism Spectrum Disorder (ASD).

Poor social skills are associated with ASD; the most commonly reported developmental disorder co-occurring in children with VI (Ek, 2010). The incidence of ASD in children with VI is high, with variable estimates to date but some studies reporting up to 40% of children with VI receiving a clinical diagnosis by school age (Mukaddes, Kilincaslan, Kucukyazici, Sevketoglu & Tuncer, 2007; Parr, Dale, Schaffer & Salt, 2010) and an even higher number showing subthreshold traits (Tadic, Pring & Dale, 2010).

The estimated childhood prevalence of ASD is substantially higher in the VI population than in the general population; estimated at 1% by age of ten years, in the UK (Baird et al., 2006). Social communication difficulties are found in children with VI and ASD and this may stem from multiple vulnerabilities in social cognition, social skills, emotional control, and relationship understanding. Up to 70% of sighted children who have ASD have at least one comorbid psychiatric disorder (Simonoff et al., 2008) and ASD traits may therefore be a particularly important risk factor in the context of VI.

Intellectual Disability (ID).

ID is a risk factor for psychopathology in the general population, with 35-40% of children with ID having a diagnosable psychiatric disorder, in a UK study (Einfield & Tonge, 1996; Emerson, 2003). This may be related to reduced coping skills (Luthar, 2003) and increased exposure to lower socioeconomic status, which are risk factors for psychopathology (Green et al., 2005; Emerson, Graham & Hatton, 2006). There may also be a shared neural or biological basis for a predisposition to ID and some forms of psychopathology (Einfield, Ellis & Emerson, 2011).

Prevalence of ID in children with congenital VI is up to ten times higher than in the general population (Nielsen, Skov & Jensen, 2007). VI originating in the main cerebral region (cortical VI) and very severe VI are particularly associated with ID. VI originating in congenital disorders of the peripheral visual system (CDPVS) with no additional known brain deficits, are associated with lower rates of ID (Dale and Sonksen, 2002). In a recent study, Harris and Lord (2016) found that eleven-year-old children who had VI and comorbid ID were at increased risk of psychopathology (29%) compared to children with VI only (18%). ID may therefore be an important risk factor for psychopathology in children with VI.

Importantly, ID and ASD are highly comorbid (Bourke, Klerk, Smith & Leonard, 2016). The complex shared prevalence of and possible interactions between VI, ID and ASD may help to explain elevated levels of psychopathology reported in some studies of children with VI. Previous research has not always specified whether children with ID and ASD have been included in VI samples, making findings difficult to interpret.

Other risk factors

Augestad (2017) highlighted that two studies in her review had found that more severe VI was associated with increased psychopathology symptoms (Huurre & Aro, 1998; Visagie et al., 2013), though further investigation is required. One study found that adolescents with earlier onset VI showed more emotional difficulties over time (Pinquart & Pfeiffer, 2014), though this has not been investigated in other studies. Seven studies found that girls were at higher risk of psychopathology than boys, but the potential role of confounds that may have biased the gender findings was not investigated.

* + 1. Current Study

Psychopathology is a known precursor to psychiatric disorder (Stein, Blum & Barbaresi, 2011), and children and adolescents who have VI may be at elevated risk of psychopathology. Further research is therefore needed to understand which variables increase risk, act as protective factors, or mediate the effects of vision loss on the development of psychopathology in these children (Pinquart & Pfeiffer, 2011). This may lead to appropriate preventative or interventionist care.

The aim of this systematic review was therefore to investigate risk and protective factors associated with psychopathology, in children and adolescents with VI. To the authors’ knowledge, this was the first review to systematically investigate this topic. The systematic review sought to include all studies meeting eligibility criteria, which includes samples of children with VI aged 4-19 years. All included studies also needed to conduct at least one within-group investigation of a possible risk or protective factor that may be associated with a measure of psychopathology.

* 1. Methods

Evidence for the potential risk and protective factors associated with psychopathology in 4-19-year olds who have VI was assessed by conducting a systematic review of published research evidence. The review adhered to published guidance for undertaking reviews in health care (Centre for Reviews and Dissemination, 2009).

* + 1. Identification of Studies

Studies were identified by searching two major electronic psychology and clinical databases: PSYCHINFO and MEDLINE. Bibliographies of previous reviews and retrieved articles were searched. Further attempts to identify studies were made by contacting clinical experts and by examining the reference lists of retrieved articles. The search took place in January 2018.

* + - 1. Inclusion and exclusion criteria.

In terms of the participants of each study, the following inclusion criteria were formulated based on previous research (Augestad, 2017): (1) having low vision and (2) being a child (4-11 years) or adolescent (12-19 years). Most studies defined ‘low vision’ according to WHO criteria, or in relation to the definition used in the country where the research took place. A minority of studies defined ‘low vision’ in relation to parent-report. Most studies investigated psychopathology in adolescents, but the broad age range was selected to also allow consideration of younger children. In terms of the study design, the following inclusion criteria were formulated (3) a measure of internalising (e.g. depression, anxiety) or externalising symptoms (e.g. aggression, behaviour), (4) a research question related to possible risk or protective factors associated with psychopathology, with a related statistical analysis and (5) a quantitative design.

The subject query consisted of the combination of two subjects: (1) vision disorder and (2) psychopathology. Medical Subject Headings (MeSH) were used to search synonyms and related terms for the subjects (see appendix A for research protocol and all accompanying search terms). Boolean operators were used (‘AND’ between the two subjects and ‘OR’ within subjects). Limits were set to include children (age 4-12) or adolescents (age 12-19). Information on studies in progress, unpublished research, research reported in grey literature, editorials, and letters were excluded from the search to identify higher quality research. Studies where the full-text was not available and studies not available in English were excluded. Qualitative designs and case studies were also excluded to target studies powered to run statistical analyses.  Studies investigating a total clinical population of children with VI and additional disabilities (e.g. the whole sample had known comorbid ID or hearing impairment) were excluded due to the decision to focus on ‘primary’ VI. The search strategy was applied.

* + 1. Selection of Articles.

Two reviewers (the first author and a research assistant) independently screened all unique titles and abstracts. Full-text papers of any titles and abstracts that were considered relevant by either reviewer were obtained where possible. Cohen’s Kappa was run to determine if there was agreement between the two raters. There was good agreement between the two raters, *k* = .83, *p* < .001. Any remaining discrepancies were subsequently resolved by consensus.

* + 1. Data Extraction Process.

When the articles for inclusion were agreed, a standardised protocol and reporting form was used to abstract the following data from each publication: names of authors, research design, year of publication, country in which the study was conducted, age and number of children in the study population, recruitment sites, definition of VI used, any comorbidities, the research question relevant to the review, the measure of psychopathology used, risk and protective factors investigated, and any relevant findings.

A data extraction form was used with each study and the information was used to create a data extraction table. A second researcher checked the accuracy of the data extraction.

* + 1. Quality Assessment

A quality assessment tool was developed to assess the studies included in the systematic review. Quality refers to the degree to which a study employs measures to minimise bias and error in its design, conduct, and analysis (Khan et al., 2003). The quality assessment tool was based on items included in The Mixed Methods Appraisal Tool (MMAT) (Pace et al., 2012). The tool has been established to have good validity and reliability (Souto et al., 2015). Only items relevant to quantitative designs were incorporated, as this was a criterion for inclusion in the systematic review. These included items about: clear research questions, representativeness, valid measures and acceptable response and completion rates (>80%). Additional specific items were added in relation to power and well-defined sample (e.g. if the vision characteristics, location, and period of recruitment were provided).

Three additional items were added after being identified as important predictors of psychopathology for typically sighted children in previous studies; gender, socioeconomic status and family factors. Whether information pertaining to these variables were included in the study contributed to the quality assessment score. Five additional items were added as they may be important for quality assessing studies using VI samples. These referred to whether studies outlined the severity of VI, the aetiological origin of the VI and the age of VI onset. They also referred to whether children with ID or other medical comorbidities were excluded or included as a separate comparison group.

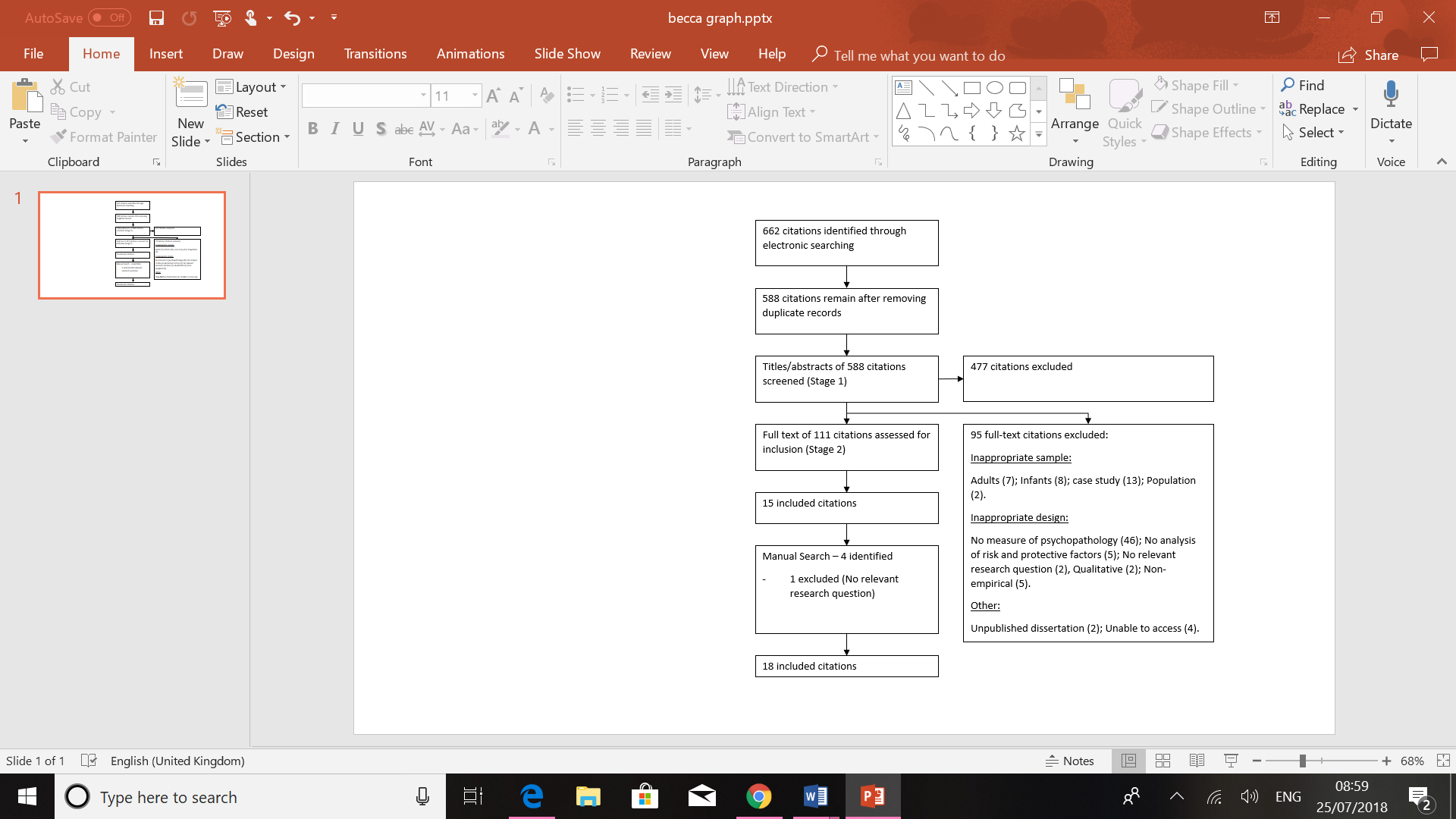
Each study was quality assessed in relation to each item and scored as fulfilling or not fulfilling the quality criteria. The score was presented as a percentage and calculated by dividing the actual score by the maximum total score (17), which was adjusted when specific items were not relevant to the study. To describe the quality rating for each study, the total percentage score was divided into quintiles and each quintile was assigned a descriptive rating. Studies were scored as ‘very high quality’ (>81%), ‘high quality’ (61-80%), ‘moderate quality’ (41-60%), ‘poor quality’ (21-40%), or ‘very poor quality (<20%).

* + 1. Data Synthesis

A narrative data synthesis was subsequently undertaken. Studies included in the review used a wide range of measures of psychopathology and had differences in sample characteristics and research methodology. This heterogeneity meant that a mathematical combination of effect sizes would be invalid and thus a meta-analysis was not conducted as part of the review.

* 1. Results
     1. Quantity of Research Available

The initial search generated a total of 662 abstracts. Once duplications were removed, 588 unique citations were left to be screened for inclusion. Their titles and abstracts were assessed for relevance to the review (Stage 1 screening) resulting in 111 being identified as potentially relevant. The full texts of these citations were obtained, and after applying the inclusion criteria (Stage 2 selection), 95 citations were excluded leaving 15 studies. Further manual searches identified four additional papers, three of which met the systematic review inclusion criteria. Eighteen citations were included in the systematic review (Figure 1). Table 1 lists the 18 included articles.

*Figure 1.* PRISMA Diagram

* + 1. Study Characteristics

All but three of the evaluated articles (Brunes et al., 2015; Dursun et al., 2015; Pinquart and Pfeiffer, 2014a) reported observational studies with a cross-sectional design. These three studies used a longitudinal design. The 18 articles represent eleven countries, including one UK study. The sample size of children with VI ranged from *n*=20 to *n*=226 (mean size: 97). Seven studies had more than 100 participants with VI (Pinquart & Pfeiffer, 2012; Pinquart & Pfeiffer, 2012b; Erol & Ergun, 2013; Pinquart & Pfeiffer, 2014a; Pinquart & Pfeiffer, 2014b; Heyl & Hintermair, 2015; Harris & Lord, 2016). The age of the sample of children with VI ranged from four to nineteen years (mean age: 14 years). Most of the studies had an adolescent sample (*n*=10), some included both children and adolescents (*n*=5) and three studies investigated children only (Alimovic, 2013; Runjic, Prcic & Alimovic, 2015; Harris & Lord, 2016). Most of the studies recruited from schools for children with VI only (*n*=10), some recruited from both schools for children with VI and mainstream schools (*n*=4), one recruited from mainstream schools only, one recruited via VI centres, and two used community sampling.

Multiple measures of psychopathology were used in studies included in the review. These included The Child Behaviour Checklist (Achenbach & Edelbrock, 1983) (*n*=3), The Revised Symptom Checklist (Derogatis, 1983) (*n*=1), Hopkins Symptom Checklist (Derogatis, Lipman, Rickels, Uhlenhuth & Cori, 1974) (*n*=1), Beck’s Depression Inventory (Beck, Steer & Brown, 1996) (*n*=2), Depression subscale of Adolescent Psychopathology Scale (Reynold, 1998) (*n*=1); Children’s Depression Inventory (Steinsmeier-Pelster, Schurmann & Duda, 2000) (*n*=1), Strengths and Difficulties Questionnaire (Goodman, 1997) (*n*=5), South African Fear Survey Schedule for Children (Burkhardt, Loxton, Kagee & Ollendick, 2012) (*n*=1), Penn State Worry Questionnaire for Children (Chorpita, Tracey, Brown, Collica & Barlow 1997) (*n*=1), Social Skills Rating System (Gresham & Elliott 1990) (*n*=1) and The University Personality Inventory Survey (Isoda, 1988) (*n*=1). Most questionnaire measures were self-report (*n*=11), some were parent only (*n*=3), one was teacher report only (Heyl & Hintermair, 2015), one was self and teacher report (Pinquart & Pfeiffer, 2012b), one was parent and teacher report (Harris & Lord, 2016). Only one study incorporated a combination of teacher, parent and self-report (Van Hasselt et al., 1986).

Risk factors studied in relation to psychopathology were varied and included gender (*n*=10), severity of VI (*n*=7), age (*n*=5); cognition (*n*=5), type of school (*n*=2) type of VI (*n*=2); sociodemographic factors (*n*=1), physical activity (*n*=2), relationship factors (*n*=2), social factors (*n*=2), communication factors (*n*=2), personality traits (*n*=1), and executive functioning (*n*=1). There were few studies investigating protective factors of psychological resilience (*n*=4),

Table 1

*Data Extraction of risk and protective factors associated with psychopathology in children with VI*

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| Source, Year | VI Sample | Nature of Sample | Psychopathology Measures | Relevant Research Questions  (\* = primary RQ) | Factors potentially associated with psychopathology | Results |
| Teare, 1984 | *N* = 23  Mean age = 10.7 years (S.D = 3.5) | PS (47.8%): no further definition and B (52.2%) = logMAR 1.4 or worse  Onset before age 2 (95.7%) or 3.  USA - Midwest  Residential blind school (1)  Comorbidities: Not mentioned, but mean IQ in normal range.  Vision conditions: Not listed | Child Behaviour Checklist (Achenbach & Rescorla, 1984).  (Dorm or natural parents). | \*What are the contributions of intellectual and demographic variables to VI children’s adjustment?  Cross-Sectional | Cognitive ability  VI severity  Gender  Age  Socioeconomic status | \*\*No significant differences between children classified as B or PS.  \*\*No significant differences between males and females.  No significant impact of age (*r* = -.14, *n* = 18, *p* > .05), socioeconomic status (*r* = -.23, *n* = 18, *p* > .05), or number of years in school (*r* = .11, *n* = 18, *p* > .05).  Higher cognitive ability predicted fewer difficulties measured by the CBCL (R squared = .45, *F* = 14.03, *p* < .01). **The magnitude of the effect was large (cohen’s *d* = 1.0).** |
| Van Hasselt et al., 1986 | *N* = 35  Mean Age = 17.2 years  Range: 13-19 years | PS <20/200 B (28.6%)  1 VI school and mainstream schools  Pittsburgh, Pennsylvania  Mean IQ 99.5 (range 87-122)  Comorbidities: Excluded  Vision conditions: Listed | Child Behaviour Checklist (Achenbach & Rescorla, 1984).  (Parent, teacher and self-report) | What are the behavioural problems and social and school adjustment needs of children with VI?  Cross-Sectional | School Type | Children in residential schools had more difficulties on schizoid, hostile, obsessive-compulsive, uncommunicative, internalising and total problems (*F* (2, 41) = 3.76, *p* < .03) than children in mainstream schools (parent rating). **The magnitude of the effect was medium (cohen’s *d* = .52).**  Children in residential schools had more internalising difficulties than children in mainstream schools (teacher report and self-report) (*F* (2, 41) = 4.99, *p* < .01). **The magnitude of the effect was large (cohen’s *d* = 1.5).** |
| Yoshida et al., 1998 | *N* = 52  Age 18-19 | Degree 1 (total blindness: 5.8%) Degree 2 (severe amblyopia: 17.3%) Degree 3 (mild amblyopia: 59.6%) Degree 4 (VA > .3: 17.3%)  Tsukuba College of Technology (for VI and HI)  Japan  Comorbidities: Not mentioned  Vision conditions: Not mentioned | University Personality Inventory Survey (Isoda, 1988)  (Self -report) | What are the mental health characteristics of students with VI?  Cross-Sectional | Gender | \*\*No statistical difference in scores between males and females. |
| Garaigordobil and Bernaras, 2009 | *N* = 29  Mean age = 14.99 years SD = 2.02).  Range: 12-17 years | Degree of vision as a function of the loss of acuity: 24.1% had a vision level between 0.4 and 0.3. 27.6% had a level between 0.25 and 0.12. 34.5% had a vision level of 0.1 or less.  Spain  VI Resource Centres  Vision conditions: Listed  Comorbidities: Excluded | The Revised Symptom Checklist (Derogatis, 1983).  (Self-report) | Are there gender differences across variables?  What are the relations between self-concept, self-esteem with other personality traits with and without psychopathological symptoms.  Cross-Sectional | Gender  Self-Concept  Self-Esteem | Females showed significantly more difficulties with: obsessive-compulsions, depression, anxiety, hostility, melancholy depression, total symptoms (*F* (1, 27) = *5*.18, p < .05) and severity of symptoms, than males. **The magnitude of the effect was large (cohen’s *d* = .76 to 1).**  There were significant negative correlations between self-concept (*R* = -.57, *n* =29, *p* < .01) and self-esteem (*R* = -.80, *n* =29, *p* < .01) with total psychopathological symptoms. **The magnitude of the effect was medium (cohen’s *d* = .45 to .70).** |
| Pinquart and Pfeiffer, 2011 | *N* = 89  Mean age = 15.57 years (S.D = 2.05) | PS, VA logMAR 0.5 or worse (75.3%). B, VA logMAR 1.4 or worse (24.7%)  Germany  VI schools (6)  Vision conditions: Not listed  Comorbidities: not mentioned | Emotional Symptoms Scale: Strengths and Difficulties Questionnaire (Goodman, 1997).  (Self-Report) | \*Do higher levels of VI associate with a higher risk of being bullied or bullying other students?  Is victimization associated with lower levels of psychological adjustment and does this association differ by vision status and availability of social support.  Cross-Sectional | VI severity  Support from peers  Victimisation | Blind students were less likely to report victimization by peers than students with low vision (*F* = *5*.22, p < .01). **The magnitude of the effect was small (cohen’s *d* = -.29).**  Blind students were less likely to report that they had bullied other students than students with low vision (*F* = *4.67*, p < .01). **The magnitude of the effect was small (cohen’s *d* = -.32).**  Bullying (beta = .20, *p* < .01) and lack of peer support (beta = 1.39, *p* < .05) were risk factors for higher emotional symptoms. Peer support acted as a protective factor for emotional distress. **The magnitude of the effect was medium (cohen’s *d* = .65).** |
| Pinquart and Pfeiffer, 2012a | *N* = 177  Mean age = 15.9 years (S.D = 2.2). | PS VA logMAR 0.5 or worse (64.4%). B logMAR 2.0 or worse (35.6%).  Germany  3 VI schools (some boarders)  Vision Conditions: Not listed  Comorbidities: (13%). | Emotional Symptoms scale: Strengths and Difficulties Questionnaire (Goodman, 1997).  (Self-Report) | Does the strength of the connection between body image and levels of life satisfaction and emotional problems vary between young people with and without VI?  Cross-Sectional | Gender  Body image | Females had higher levels of emotional problems than males (*F* (1, 693) = 6.47, *p* <.02). **The magnitude of the effect was medium (cohen’s *d* = -.59).**  Poor body image was associated with more emotional symptoms (*r* = -.47, *p* < .001, particularly in females. **The magnitude of the effect was medium (cohen’s *d* = .50).** |
| Pinquart and Pfeiffer, 2012b | *N* = 158  6th-11th Grade  Range: 12 to 19 years  Mean age = 16.01 (S.D. = 1.92) | B(36.5%) logMAR 1.4 or worse PS (64.5%) logMAR 0.5 or worse  Germany  3 VI schools (some boarders)  Vision conditions: Listed  Comorbidities: Included | Strengths and Difficulties Questionnaire (Goodman, 1997)  Child Behaviour Checklist (Muris, Meesters & van den Berg, 2003).  (Self and teacher reports). | Does the psychological adjustment of students with VI vary by the magnitude of the vision loss?  Cross-Sectional | VI severity | No differences in psychopathology between B and PS (*F* (1, 153) = 1.15, *p* > .05) |
| Alimovic, 2013 | *N* = 80  (M: 7.65 years).  Range: 4 to 11 years | VI (defined according to Croatian regulations).  Mild LD (50%)  Zagreb  Mainstream and special schools (Live at home).  Vision conditions: Not listed  Comorbidities: Not mentioned (LD included) | Child Behaviour Checklist (Achenbach & Rescorla, 2001).  (Parent Report) | \*Do children with mild ID and VI have a higher prevalence and different type of difficulties than peers with mild ID or single VI?  Cross-Sectional | Mild ID | Mild ID was a significant risk factor for total emotional and behavioural problems (*F* (39) = 11.7, *p* = .007) and specifically for attention problems, withdrawal and social problems. **The magnitude of the effect was large (cohen’s *d* = .80).** |
| Emam (2013) | *N* = 91  Age 12-17 years | VA not reported  Residential VI School (1).  Egypt  Vision conditions: Not listed  Comorbidity: Not mentioned | Beck Depression Inventory for Youth (Beck, Beck, Jolly & Steer, 2005).  Depression subscale of Adolescent Psychopathology Scale (Reynold, 1998).  (Self-Report) | \*What is the association between cognitive variables (Attributional Style and Problem-Solving Orientation) and depressive symptoms in adolescents with VI?  Cross-Sectional | Gender  Problem-Solving Skills  Attributional Style | Females were more at risk of depression than males (R squared = .23, *F* (7, 90) = 3.56, *p* < .001). **The magnitude of the effect was large (cohen’s *d* = 1.1).**  Negative Attributional Style (beta = .23, p < .001) and Negative Problem-Solving Orientation (beta = .23, p < .001) were also risk factors for depression. **The magnitude of the effect was large (cohen’s *d* = 1.1).** |
| Erol & Ergun, 2013 | *N* = 130  Mean Age = 13.67 years (S.D = 1.47).  Range: 12-18 years. | VA logMAR 1.0 or worse  VI school (1) (75.8% boarders).  Istanbul  Vision conditions: Not listed  Comorbidity: Excluded | Beck Hopelessness Scale (Beck, 1974).  (Self-Report) | What are the relationships between hopelessness and social comparison in adolescents with VI?  Cross-Sectional | Gender  Age  Type of school  Social comparison  Negative feelings towards fathers, mothers, teachers & siblings. | \*\*Gender, age, school status (e.g. boarder), feelings towards mothers and feelings towards siblings were not found to associate with hopelessness.  Social comparison was significantly negatively correlated with hopelessness (*r* = - 0.46; *p* < 0.001). **The magnitude of the effects was large (cohen’s *d* = 1.0).**  \*\*\*Negative feelings towards Fathers and Teachers were associated with hopelessness (*p* < .001) |
| Visagie, et al., 2013 | *N* = 67  Mean age = 10.36 years (S.D = 1.46).  Range = 8-13 years. | PS: logMAR 0.5 or worse (70.1%). B: logMAR 1.4 or worse (29.9%).  South Africa  VI schools (2)  Vision conditions listed.  Comorbidities: excluded. | South African Fear Survey Schedule for Children (Burkhardt, Loxton, Kagee & Ollendick, 2012).  (Self-Report). | How do different fears manifest when variables related to gender, age, and degree of visual impairment were taken into account?  Cross-Sectional | Gender  VI severity  Age | More severe VI (blindness) was associated with more significantly more fears (*F* (2, 126) = 9.04, *p* < .001). **The magnitude of the effects was large (cohen’s *d* = .98).**  Females had more fears than males. (*F* (1, 125) = 23.87, *p* < .001). **The magnitude of the effects was medium (cohen’s *d* = .78).**  No difference between younger (aged 8-10) and older (aged 11-13) groups. (*F* (1, 125) = .10, *p* = .76). |
| Pinquart and Pfeiffer, 2014a | *N* = 182  Time 1: (M = 15.61 years, S.D. 2.04). | PS, VA logMAR 0.5 or worse (63.2%). B (36.8%).  Congenital (75.4%)  Acquired (24.4%).  Germany  VI schools (3) (12% boarders)  Comorbidity: included (13%). | Strengths and Difficulties Questionnaire (Goodman, 1997).  (Self-Report) | Do change of psychological problems vary between adolescents who are blind and their peers with low vision, as well as by age and onset of visual loss?  Longitudinal | Gender  Age  VI Severity  VI onset | Later onset of vision loss associated with stronger improvements of total difficulties (*Z* = -2.2, *p* < .05), conduct problems, peer problems and prosocial behaviour. **The magnitude of the effects was small (cohen’s *d* = .28).**  VI severity had no significant impact on initial SDQ scores (*Z* = -.48, *p* > .05) or level of change (*Z* = 1.09, *p* > .05)  \*\*\*Females had more overall difficulties and emotional problems than males (*Z* = 2.01, *p* <.05), but better prosocial behaviour (*Z* = 4.32, *p* < .01).  \*\*\*Average SDQ scores declined over time (*Z* = -4.93, *p* < .001). |
| Pinquart and Pfeiffer, 2014b | *N* = 162  Mean age = 16.9 years (S.D 2.6). | PS (73%) B (27%) (according to their certificate of disability)  Congenital (82%) Acquired (18%).  2 VI schools  Germany  Vision conditions: Not listed  Comorbidities: Not mentioned | Penn State Worry Questionnaire for Children (Chorpita et al, 1997).  Children’s Depression Inventory (Steinsmeier-Pelster, Schurmann & Duda, 2000).  (Self-Report) | What are the correlates of worrying in adolescents with VI (age, gender, VI severity, grades, psychological resources, depression)?  Cross-Sectional | VI Severity  Gender  Visual Decline  Grades  Psychological resilience | Children with more severe VI had higher levels of worry (beta = .65, *p* < .001). **The magnitude of the effects was large (cohen’s *d* = 1.7).**  No gender differences (beta = .07, *p* > .05)  Perceived visual decline was not associated with worry (beta = -.04, *p* > .05).  Worse grades associated with more worries (beta = .15, *p* > .05). **The magnitude of the effects was small (cohen’s *d* = .30).**  Depression associated with worry. Optimism inversely related to worry and depression (*r* = .61, *p* < .001). **The magnitude of the effects was large (cohen’s *d* = 1.5).** |
| Brunes et al., 2015 | *N* = 46  Age range: 12-17 | Self-report ‘severe VI’  Norway  State Schools  Population Sample  Vision conditions Not listed.  Comorbidities: Not mentioned. | Hopkins Symptom Checklist (Derogatis, Lipman, Rickels, Uhlenhuth & Cori, 1974).  (Self-Report) | \*Examine the effect of leisure-time Physical Activity on symptoms of mental health problems and subjective well-being among adolescent with no impairment and VI.  Longitudinal | Emotionally Unstable personality  Introverted personality  Physical Activity | \*\*/\*\*\*Being either more emotionally unstable or more introverted and scoring non-weekly physical activity was significantly associated with more mental health difficulties, than those scoring weekly physical activity. |
| Dursun et al., 2015 | *N* = 20  Mean age = 12 years.  Range = 8-16 years. | VA < logMAR 1.0  Turkey  VI School (1). Boarders.  Vision conditions Not listed.  Co-morbidities: Excluded | Strengths and Difficulties Questionnaire (Goodman, 1997).  (Self-Report) | \*Evaluate the effects of an ice skating programme on self-concept, behavioural and emotional problem domains and sleep quality of children and adolescents with VI or HI.  Longitudinal | One-hour, twice weekly sessions that lasted for 3 months. Each student has their own trainer. | The ice skating programme was associated with a decrease in emotional problems (SDQ) (*t* = (19) = .83, *p* < .001). **The magnitude of the effects was small (cohen’s *d* = .38).** It also associated with an increase in hyperactivity-inattention (*t* = (19) = 3.24, *p* < .01). **The magnitude of the effects was large (cohen’s *d* = 1.4).** It also associated with an increase in peer problems (*t* = (19) = 2.14, *p* < .05). **The magnitude of the effects was large (cohen’s *d* = .98).** |
| Heyl & Hintermair, 2015 | *N* = 226  Mean Age = 12.0 years (S.D = 3.5).  Range = 5-18 years. | PS (logMAR 0.5 or worse)(60%), PVI (logMAR 1.3 or worse )(21.4%) and B (logMAR 1.7 or worse)(18.6%).  Germany  Mainstream and VI schools  Additional disability: mainstream (75.6%), special (30.1%).  Vision conditions Not listed.  Comorbidities: 50%. Excluded in second analysis. | Strengths and Difficulties Questionnaire (Goodman, 1997).  (Teacher Report) | Do VI students at mainstream schools differ in executive function from VI students at special schools?  Are there any relationships between executive function, communicative competencies and behavioural problems in VI students?  Cross-Sectional | Executive function  Communicative Competence.  Age  Gender  Type of School  ID  Immigration background  VI severity | Increased executive function problems correlated with more behaviour problems (*r* = .72, *p* <.001). **The magnitude of the effects was large (*d* = 2.1).** Increased executive function problems associated with lower levels of communicative competence (*r* = -.67, *p* < .001). **The magnitude of the effects was medium (cohen’s *d* = .66).**  Gender (female gender was protective) associated with SDQ (beta = .18, *p* <.001). **The magnitude of the effects was small (cohen’s *d* = .36).**  Age (older age was protective) associated with SDQ (beta = .19, *p* < .001). **The magnitude of the effects was small (cohen’s *d* = .39).**  Executive function (particularly behaviour regulation) associated with SDQ (beta = .63, *p* < .001). **The magnitude of the effects was large (cohen’s *d* = 1.6).**  Communicative competence (non-significant trend) (beta = -.09, *p* > .05) associated with SDQ score.  \*\*Immigration background, severity of VI, additional disability and type of school did not associate with SDQ score. |
| Runjic et al., 2015 | *N* = 39  Age 4 – 11 | PS (76.9%) B (23.1%)  VI defined according to Croatian regulations  Croatia  Mainstream schools and VI school (1)  Vision conditions Not listed.  Comorbidities: excluded | Social Skills Rating System | \*What are the relationships between social skill performance, cooperation, responsibility and behavioural problems in students with V?  Cross-Sectional | Cooperation  Social Skills  Ability to follow instructions (responsibility) | There is a significant correlation between social skills performance and behavioural problems. More cooperative and responsible children have fewer problems in behaviour (*r* = .57, *p* < .001). **The magnitude of the effects was large (cohen’s *d* = 1.3).** |
| Harris & Lord, 2016 | *N* = 189  Age 11 | Parent-Report VI  UK  Millennium Cohort  Vision conditions Not listed.  Comorbidities: included | Strengths and Difficulties Questionnaire (Goodman, 1997).  (Parent and Teacher report). | Does the presence of special educational needs and disability act as an additional risk factor?  Cross-Sectional | ID | \*\*/\*\*\*ID was a significant risk factor for total score, emotional symptoms, conduct problems, hyperactivity and prosocial behaviour. It did not impact on peer problems. |

Cohen’s d descriptive ranges: .2 (small effect), .5 (medium effect), .8 (large effect)

\*Primary research question is about risk and protective factors. PS (partially sighted), B(Blind), VA(Visual Acuity), ID (Intellectual Disability), IQ (intelligence quotient)

\*\*p value not given

\*\*\* Mean and SD not given and so effect size cannot be calculated.

Numerous factors were investigated as potential risk factors and a small number of studies investigated potential protective factors (Table 2). P values and effect sizes are given where available.

Table 2

*Findings of risk and protective factors associated to psychopathology in children with VI*

|  |  |  |
| --- | --- | --- |
| Risk or Protective Factor: | Number of Studies considering this: |  |
| **Demographic Factors** | 10 | *Females at greater risk of psychopathology than males:*  Garaigorobil and Bernaras (2009) (*p* < .05, large effect size), Pinquart and Pfeiffer (2012a) (*p* < .02, medium effect size); Pinquart and Pfeiffer (2014a) (*p* < .05); Emam (2013) (*p* < .001, large effect size); Visagie et al., 2013 (*p* < .001, medium effect size).  *Males at greater risk of psychopathology than females:*  Heyl and Hintermair (2015) (*p* < .001, small effect size)  *No gender differences*  Teare (1984); Yoshida et al., (1998); Erol & Ergun (2013); Pinquart and Pfeiffer (2014b) (p *>* .05). |
| Gender |
| Type of School | 3 | *Residential school pupils have more difficulties*  Van Hasselt et al., (1986) (personality difficulties: *p* < .03, medium effect size, internalising difficulties: *p* < .01, large effect size)  *Type of school does not have an effect*  Erol and Ergun (2013); Heyl and Hintermair (2015). |
| Age | 4 | *Younger age is associated with increased psychopathology symptoms*  Pinquart and Pfeiffer (2014a) (*p* < .001); Heyl and Hintermair (2015) (*p* < .001, small effect size)  *No impact of age*  Teare (1984) (*p* > .05) |
| Sociodemographic Factors | 1 | *No impact of socioeconomic status*  Teare (1984) (*p* > .05) |
| **Vision Factors** | 7 | *More severe VI is associated with increased psychopathology symptoms* |
| Vision Level | Visagie et al., (2013) (*p* < .001, large effect size); Pinquart and Pfeiffer (2014b) ) (*p* < .001, large effect size).  *More severe VI is associated decreased psychopathology symptoms*  Pinquart and Pfeiffer (2011) (*p* < .01, small effect size)  *No difference in psychopathology between different severity of VI*  Teare (1984); Pinquart and Pfeiffer (2012b) (*p* > .05); Pinquart and Pfeiffer (2014a) (*p* > .05); Heyl and Hintermair (2015). |
| Age of VI onset | 1 | *Later onset of VI is associated with greater improvement in externalising symptoms*  Pinquart and Pfeiffer (2014a) (*p* < .05, small effect size). |
| Vision Decline | 1 | *Perceived vision decline is not associated with worry*  Pinquart and Pfeiffer (2014b) |
| **Cognitive factors** | 2 |  |
| Intelligence | *Stronger intelligence associated with fewer symptoms of psychopathology*  Teare (1984) (*p* < .01, large effect size); Pinquart and Pfeiffer (2014b) (*p* < .05, small effect size); |
| Intellectual Disability | 3 | *Intellectual Disability associated with more symptoms of psychopathology*  Alimovic (2013) (*p* = .007, large effect size); Harris and Lord (2016) |
|  |  | *Additional disability is not related to psychopathology*  Heyl and Hintermair (2015) |
| Executive functioning | 1 | *Executive functioning associated with psychopathology*  Heyl and Hintermair (2015) (*p* < .001, large effect size). |
| **Personality Factors** | 1 | *Personality traits associated with psychopathology*  Brunes et al., (2015) |
| **Social Factors**  (Support from peers, victimisation, social comparison) | 2 | *Social factors associated with psychopathology*  Pinquart and Pfeiffer (2011) (*p* < .05, medium effect size); Erol and Ergun (2013) (*p* < .001, large effect size). |
| **Communication Factors**  (Communicative Competence, Cooperation, Social Skills, Responsibility) | 2 | *Communication factors associated with psychopathology*  Heyl and Hintermair (2015) (*p* < .001, medium effect size); Runjic et al., (2015) (*p* < .001, large effect size). |
| **Family Factors**  Quality of family interactions | 1 | *Family factors associated with psychopathology*  Erol and Ergun (2013) (*p* < .001). |
| **Physical Factors**  (Physical Activity, Ice Skating Programme) | 2 | *Physical factors associated with more symptoms of psychopathology*  Dursun et al., (2015) (*p* < .001, small effect size).  *Physical factors associated with fewer symptoms of internalising and more symptoms of externalising*  Brunes et al., (2015); Dursun et al., (2015) (*p* < .05, large effect size). |
| **Protective factors**  (self-concept, self-esteem, body image, psychological resilience, attributional style, optimism) | 4 | *Psychological factors associated with psychopathology*  Garaigorobil and Bernaras (2009) (*p* < .01, medium effect size); Pinquart and Pfeiffer (2012a) (*p* < .001, medium effect size); Emam (2013) (*p* < .001, large effect size); Pinquart and Pfeiffer (2014b) (*p* < .001, large effect size); |

* + - 1. Risks in the Socioeconomic Context

Sociodemographic factors.

Teare’s research (1984) was rated as ‘high’ in the quality assessment and found no effects of socioeconomic status. Children and adolescents with lower socioeconomic status are at increased risk of psychopathology in the TD population. This review has not found evidence to support this pattern within the VI population, though it has been under-researched.

Type of school.

Three studies investigated the type of school attended in relation to psychopathology. Of these studies, one found that the type of school was associated with psychopathology (Van Hasselt et al., 1986) and two did not (Erol & Ergun, 2013; Heyl & Hintermair, 2015). Van Hasselt et al., (1986) found that children in residential schools had more internalising and externalising problems than children in mainstream schools, whereas Erol and Ergun (2013) found no differences in levels of hopelessness between residential and day students. Heyl and Hintermair (2015) found no differences in behaviour difficulties between students attending mainstream and specialist VI schools. Given the different educational inclusion policies between countries, it is difficult to draw conclusions regarding the impact of distinct types of schooling. For example, in some countries all children who have VI may be educated in specialist schools and in others, it may only be the children who have co-occurring disorders such as ID or ASD who are educated in specialist schools.

* + - 1. Risks in the Family

Family factors related to psychopathology.

One study investigated the role of family relationships in psychopathology. Erol and Ergun (2013) found that feelings towards fathers and teachers correlated with feelings of hopelessness. There has been a distinct lack of research investigating family factors in relation to psychopathology.

* + - 1. Risks Related to the Child

Gender.

Ten studies investigated gender differences in relation to psychopathology. Of these, five studies concluded that females were at higher risk of psychopathology than males (Garaigorobil & Bernaras, 2009; Pinquart & Pfeiffer, 2012a; Emam, 2013 & Visagie et al., 2013; Pinquart & Pfeiffer, 2014b).  One study concluded that males were at higher risk of psychopathology than females (Heyl & Hintermair, 2015) and four studies did not report any gender differences in relation to psychopathology (Teare, 1984; Yoshida et al., 1998; Erol & Ergun, 2013 & Pinquart & Pfeiffer, 2014b).

Of these studies, five were rated as ‘high’ or ‘very high’ quality. Three found that females were at greater risk of psychopathology than males and two found no gender effects. Two of the ten studies excluded children with ID: one found gender differences (Garaigorobil & Bernaras, 2009) and one did not (Erol & Ergun, 2013). It therefore cannot be concluded with confidence that female gender is a risk factor for psychopathology in children with VI.

Age.

Four studies investigated the relationship between age and psychopathology (Teare, 1984; Erol & Ergun, 2013; Pinquart & Pfeiffer, 2014a; Heyl & Hintermair, 2015). Two of the studies found no association between age and behaviour (Teare, 1984; Erol & Ergun, 2013). Heyl and Hintermair (2015) found that younger children had more behavioural difficulties than adolescents. In a longitudinal study, Pinquart & Pfeiffer (2014a) found that internalising and externalising symptoms improved over a period of two years. Mixed findings may be related to differences in methodology (cross-sectional compared to longitudinal) and differences in age groups investigated. Most of these studies did not exclude children with comorbid ID, and so the developmental level of the samples may differ from the chronological age, which may confound findings.

Vision factors.

Seven studies investigated the relationship between severity of VI and psychopathology. Two studies compared ‘blind’ and ‘partially sighted’ children and adolescents and found that the blind groups were more at risk of psychopathology than the sighted groups (Visagie et al., 2013; Pinquart & Pfeifer, 2014b). One study found that adolescents who were ‘partially sighted’ had more behavioural difficulties than adolescents who were ‘blind’, though this was associated with bullying which may be less of a risk for blind adolescents who are likely to have a high level of adult support (Pinquart & Pfeiffer, 2011). Four studies did not find differences in psychopathology between children with different severity of VI (Teare, 1984; Pinquart & Pfeiffer, 2012b; Pinquart & Pfeiffer, 2014a; Hely & Hintermair, 2015).

Of the three studies which were rated as ‘high’ or ‘very high’ quality, two studies found no effect of VI severity (Teare, 1984; Pinquart & Pfeiffer, 2012b) and one found that ‘blindness’ was a risk factor for psychopathology (Pinquart & Pfeiffer, 2014a). Both studies conducted by Pinquart and Pfeiffer included children with additional comorbidities and it is not clear whether children with medical comorbidities were excluded from Teare’s (1984) study. As children who are blind are more likely to have comorbidities than children who are partially sighted, this could be a significant confound in analyses investigating the relationship between severity of VI and psychopathology. One study found that later onset VI was associated with a greater improvement in externalising symptoms compared to earlier onset VI (Pinquart & Pfeiffer, 2014a) and one found that perceived visual decline was not a risk factor for psychopathology (Pinquart & Pfeiffer, 2014b), though these findings require replication.

Cognitive factors.

Seven studies investigated cognitive factors in relation to psychopathology. Two studies found that ID was associated with higher levels of psychopathology (Alimovic, 2013; Harris & Lord, 2016) and one did not (Heyl & Hintermair, 2015). The former two studies used samples of children, whereas the latter used a broad age range of five to 18 years. It may be that ID particularly relates to psychopathology in younger children who have VI. Two studies found that stronger cognitive ability was associated with lower levels of psychopathology (Teare, 1984; Pinquart & Pfeiffer, 2014). Two studies found that various aspects of cognitive processing were associated with psychopathology (Heyl & Hintermair, 2015; Emam, 2013). Executive functioning problems (Heyl & Hintermair, 2015) and negative problem-solving orientation (Emam, 2013) were also associated with more behaviour problems and more symptoms of depression.

Personality factors related to psychopathology.

One study investigated the relationship between personality traits and psychopathology. Brunes et al., (2015) found that emotional instability and introversion are associated with increased symptoms of psychopathology.

Social factors related to psychopathology.

Two studies investigated the relationship between social factors and psychopathology. Bullying and lack of peer support was found to associate with higher levels of emotional difficulties (Pinquart & Pfeiffer, 2011) and social comparison was found to negatively associate with hopelessness (Erol & Ergun, 2013).

Communication factors related to psychopathology.

Two studies investigated the relationship between communication factors and psychopathology. Heyl and Hintermair (2015) found a non-significant trend between communicative competence and behaviour problems. Runjic et al., (2015) found a significant correlation between poorer social skills and behaviour difficulties and found that children who were less cooperative and less responsible have increased behaviour problems and anxiety symptoms than children with less difficulties in these areas.

Physical factors related to psychopathology.

Two studies investigated the role of physical activity in relation to psychopathology. Brunes et al., (2015) found that weekly physical activity was associated with fewer symptoms of psychopathology than less than weekly physical activity. Dustun et al., (2015) found that whilst a physical activity in the form of an ice-skating programme was associated with a decrease in emotional difficulties, it was also associated with an increase in hyperactivity-attention and peer problems.

Protective factors.

Just four studies investigated how potential protective factors associated with psychopathology (Teare, 1984; Garaigorobil & Bernaras, 2009; Pinquart & Pfeiffer, 2012; Emam, 2013; Pinquart & Pfeiffer, 2014). Garaigorobil and Bernaras (2009) found that higher self-concept and self-esteem associated with lower levels of psychopathology. Positive body image (Pinquart & Pfeiffer, 2012) and positive attributional style (Emam, 2013) were also found to associate with lower levels of psychopathology. Pinquart and Pfeiffer (2014) found that optimism was inversely related to worry.

Table 3

*Quality Assessment*

|  |  |  |  |  |  |  |  |  |  |  |  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
|  | External Validity - Sampling | | | | Internal Validity | | | | Are psychopathology confounders measured? | | | Percentage Score | | Are VI confounders measured? | | | | | Percentage Score | Overall Score |
|  | Representativeness | Participation rate | Defined Study Population | Power | Research Question | Were variables reliable and clearly defined | Withdrawal/Drop Out Report | Percentage of participants included | Gender | Family Factors | Socioeconomic Factors | |  | VI Severity | VI origin | VI Onset | LD Excluded? | Other excluded? |  |  |
| Teare (1984) | 🗶 | ✓ | ✓ | ✓ | ✓ | ✓ | ✓ | ✓ | ✓ | 🗶 | ✓ | | 75% | ✓ | 🗶 | ✓ | 🗶 | 🗶 | 40% | 65% |
| Van Hasselt et al., 1986 | ✓ | NS | ✓ | ✓ | ✓ | ✓ | 🗶 | NS | 🗶 | 🗶 | 🗶 | | 42% | ✓ | ✓ | ✓ | ✓ | ✓ | 100% | 59% |
| Yoshida et al., 1998 | 🗶 | NS | ✓ | ✓ | ✓ | 🗶 | 🗶 | NS | ✓ | 🗶 | 🗶 | | 33% | ✓ | 🗶 | 🗶 | 🗶 | 🗶 | 20% | 29% |
| Garaigorobil & Bernaras (2009) | ✓ | 🗶 | ✓ | ✓ | ✓ | ✓ | NA | ✓ | ✓ | 🗶 | 🗶 | | 64% | ✓ | ✓ | ✓ | ✓ | ✓ | 100% | 75% |
| Pinquart & Pfeiffer (2011) | 🗶 | ✓ | ✓ | ✓ | ✓ | ✓ | NA | ✓ | ✓ | 🗶 | 🗶 | | 64% | ✓ | 🗶 | 🗶 | 🗶 | 🗶 | 20% | 50% |
| Pinquart & Pfeiffer (2012a) | 🗶 | ✓ | ✓ | ✓ | ✓ | ✓ | ✓ | ✓ | ✓ | 🗶 | ✓ | | 75% | ✓ | 🗶 | ✓ | 🗶 | 🗶 | 40% | 65% |
| Pinquart & Pfeiffer, 2012b | 🗶 | ✓ | ✓ | ✓ | ✓ | ✓ | ✓ | ✓ | ✓ | 🗶 | ✓ | | 75% | ✓ | ✓ | 🗶 | 🗶 | 🗶 | 40% | 65% |
| Alimovic (2013) | ✓ | NS | ✓ | ✓ | ✓ | ✓ | NS | NS | ✓ | 🗶 | 🗶 | | 50% | 🗶 | 🗶 | 🗶 | NA | NA | 0% | 40% |
| Emam (2013) | 🗶 | NS | 🗶 | ✓ | ✓ | ✓ | 🗶 | ✓ | ✓ | 🗶 | ✓ | | 50% | 🗶 | 🗶 | 🗶 | 🗶 | 🗶 | 0% | 35% |
| Erol & Ergun (2013) | 🗶 | ✓ | ✓✓ | ✓ | ✓ | ✓ | NA | ✓ | ✓ | ✓ | ✓ | | 91% | ✓ | 🗶 | 🗶 | ✓ | ✓ | 60% | 81% |
| Pinquart & Pfeiffer (2014a) | ✓ | ✓ | ✓ | ✓ | ✓ | ✓ | ✓ | 🗶 | ✓ | 🗶 | ✓ | | 82% | ✓ | 🗶 | ✓ | 🗶 | 🗶 | 40% | 65% |
| Visagie, et al., (2013) | 🗶 | NS | ✓ | ✓ | ✓ | ✓ | 🗶 | NS | ✓ | 🗶 | ✓ | | 60% | 🗶 | ✓ | 🗶 | 🗶 | ✓ | 40% | 47% |
| Pinquart & Pfeiffer (2014b) | 🗶 | ✓ | ✓ | ✓ | ✓ | ✓ | 🗶 | ✓ | ✓ | 🗶 | 🗶 | | 58% | ✓ | 🗶 | ✓ | 🗶 | 🗶 | 40% | 53% |
| Brunes et al., (2015) | 🗶 | NS | ✓✓ | ✓ | ✓ | ✓ | 🗶 | 🗶 | ✓ | 🗶 | 🗶 | | 50% | 🗶 | 🗶 | 🗶 | 🗶 | 🗶 | 0% | 35% |
| Dursun et al., 2015 | 🗶 | ✓ | ✓ | ✓ | ✓ | ✓ | NA | ✓ | ✓ | 🗶 | 🗶 | | 58% | ✓ | 🗶 | 🗶 | ✓ | ✓ | 60% | 67% |
| Heyl & Hintermair, 2015 | ✓ | NS | ✓ | ✓ | ✓ | ✓ | 🗶 | NS | ✓ | 🗶 | ✓ | | 58% | ✓ | 🗶 | 🗶 | 🗶 | 🗶 | 20% | 47% |
| Runjic et al., 2015 | ✓ | NS | ✓ | ✓ | ✓ | ✓ | 🗶 | NS | ✓ | 🗶 | 🗶 | | 50% | 🗶 | 🗶 | 🗶 | ✓ | ✓ | 40% | 47% |
| Harris & Lord, 2016 | ✓ | NS | ✓ | ✓ | ✓ | ✓ | 🗶 | NS | 🗶 | 🗶 | 🗶 | | 42% | 🗶 | 🗶 | 🗶 | NA | NA | 0% | 33% |
| Overall Score | 37% | 47% | 55% | 100% | 100% | 95% | 29% | 53% | 90% | 10% | 47% | | **60%** | 68% | 26% | 32% | 35% | 41% | **39%** | **53%** |

✓✓ yes (defined study population item – defines location, vision characteristics and time period of recruitment) ✓yes (item adequately addressed) (defined study population item – defines at least two of the measured aspects); 🗶 no (item not adequately addressed) (score 0); NS not stated (score 0); NA not applicable (adjust total score).

1st-20th percentile = very poor, 21st-40th percentile = poor, 41st-60th percentile = moderate, 61st-80th percentile = high, 81st-100th percentile = very hig

* + 1. Quality of Included Studies

The average overall score for total methodological quality was 53%, which is in the ‘moderate’ range. One study scored in the ‘very high’ range (Erol & Ergun, 2013). Six studies scored in the ‘high’ range (Teare, 1984; Garaigorobil & Bernaras, 2009; Pinquart & Pfeiffer, 2012; Pinquart & Pfeiffer, 2012b; Pinquart & Pfeiffer, 2014a; Dursen et al., 2015). Six studies scored in the ‘moderate’ range (Heyl & Hintermair, 2015; Pinquart & Pfeiffer, 2011; Pinquart & Pfeiffer, 2014b; Runjic et al., 2015; Van Hasselt et al., 1986; Visagie et al., 2013). Five studies scored in the ‘poor’ range (Almovic, 2013; Brunes et al., 2015; Emam, 2013; Harris & Lord, 2016; Yoshida et al., 1998). Overall, studies used valid research questions and measures, and analyses were appropriately powered. Participation and withdrawal rate was mixed, as was the adequacy of sampling. Representativeness was an issue in most studies and reasons for participant withdrawal were rarely given. Most studies considered gender and VI severity, some considered socioeconomic factors, and few considered vision diagnosis, onset of VI, ID, or family factors.

* 1. Discussion

This systematic review aimed to investigate risk and protective factors associated with psychopathology, in children and adolescents with VI.  The systematic review found that several possible risk factors were identified in the relevant literature. The results of the studies in the reviewed articles were often inconsistent, which may be due to the different research purposes, study designs, samples, measurements, and analyses of psychopathology and risk factors. The most frequently researched variables were gender, VI severity, and cognitive ability. There was a lack of research investigating potential protective factors.

There was some evidence that females with VI were at greater risk of psychopathology than males, with medium to large effect sizes, though findings between studies were mixed and so a clear conclusion could not be drawn in relation to gender. There was some evidence that children with more severe VI were at greater risk of psychopathology compared to those with less severe VI, with large effect sizes, though findings were mixed, and most studies did not demonstrate significant effects. The only clear-cut finding was therefore that lower cognitive ability was a risk factor for psychopathology in children with VI. Of the six studies investigating this, five found a significant relationship between cognition and psychopathology symptoms, with small to large effect sizes. One study did not find an effect but did not report statistics in relation to this.

There was a lack of research investigating other potential VI-related risk factors in relation to psychopathology such as onset and course of the VI, which may have an important impact. Conclusions cannot be drawn in relation to age effects due to mixed findings. Individual studies have found evidence to suggest that protective factors (e.g. self-esteem, body image, optimism), social and communication skills, family relationships, executive functioning and physical activity may associate with lower levels of psychopathology, but these findings need to be replicated in future studies. The only clear-cut finding is therefore that ID is a risk factor for psychopathology within VI samples. There has been a lack of multivariate analysis, making it difficult to draw conclusions regarding the relative importance of risk factors associated with psychopathology in children who have VI. There has also been a distinct lack of research investigating protective factors.

* + 1. Strengths and Limitations of the Included Studies

The overall methodological quality of included studies was rated as ‘moderate’. The heterogeneity of childhood VI means that it is difficult for studies to be representative and valid. Overall, studies had valid research questions and measures and appropriate sample sizes. Participant rate, withdrawal rate, and samples representativeness and definition were weaker. Most studies reported on gender and VI severity, but few considered socioeconomic, family, ID, health condition, or specific vision disorder factors as possible confounders. Few studies explained whether the vision disorders were cortical or peripheral ophthalmological in origin, and few stated whether the sample included children who had acquired or later onset VI. Studies often did not describe the vision disorders in the sample, though specific phenotypes may be associated with the underlying genotypes. There was a lack of information regarding comorbid disorders, which are common in childhood VI. Studies included children with differing levels of visual acuity and the severity of VI was not always defined, particularly in community studies. Without this information, it was often not possible to assess the generalisability of findings or to explain mixed findings between studies.

A further consideration is that most studies recruited from specialist schools for children who have VI only. In the UK, children who have VI who attended specialist schools usually had a co-occurring disability such as ID or additional sensory impairment, though inclusion policies in educational provisions vary between countries. It may therefore be that children included in previous studies frequently have had comorbid conditions, which may have had a cumulative impact on the child with VI. Children who have congenital VI, cortical VI, and total blindness are thought to be at higher risk of neurological impairment such as ID (Sonksen & Dale, 2002), which is predictive of psychopathology.

The age range of children included within studies was often large and spanned multiple developmental periods, which might have impacted on the validity of findings. Longitudinal studies have found that externalising disorders generally decrease with age (Hicks et al., 2007), whereas internalising disorders may increase with age (Sagatum, Lien, Sogaard, Bjertness & Heyerdahl, 2008). Another limitation is that most studies did not consider socioeconomic factors. This is potentially problematic as children with disabilities are more at risk of social disadvantage than children without disabilities, and this is a known risk factor for psychopathology (Pinquart & Sorensen, 2000). This may therefore also be relevant when considering psychopathology in children with VI. Studies were also limited to specific geographical areas, which may have impacted on the generalisability to other regions or countries, where socioeconomic context, service provision, and culture differences may have an impact on experience and reporting of psychopathology.

Most studies had a cross-sectional design and used correlational data, which does not allow for consideration of causality. A wide range of measures of psychopathology have been used, making it difficult to make reliable comparisons between studies. Related to this, it was not possible to synthesize the results into a meta-analysis. Measures of psychopathology have not been validated for use with VI samples. Moreover, the question of whether items were adapted, or needed to be adapted for use with this population was often not reported in studies. Most studies included self-report measures only, whereas a multiple-informant perspective may be more effective in considering child psychopathology, particularly in relation to externalising symptoms. As is true for a plethora of research topics, publication bias may have also had an impact. Some studies may thus have remained unpublished due to non-significant findings, potentially skewing the conclusions of the systematic review.

* + 1. Strengths and Limitations of the Systematic Review

The search strategy used in the systematic review was comprehensive. The search terms were expansive and able to capture most of the relevant studies. The age range (4 to 19 years) was broad but necessary given the paucity of research in this specific area. The focus was on psychopathology, rather than broader constructs of wellbeing, which allowed consideration of more robust measures of symptoms associated with long term difficulties. The decision was made to limit the search to empirical studies and so case studies and studies with qualitative designs were excluded. Whilst this allowed for more robust findings and conclusions, the review may also have missed some potentially relevant findings. The review may be slightly less representative due to the purposeful exclusion of unpublished and grey literature, qualitative designs, and texts that were not available in English, however this allowed for the rigor of quality control to be maximised.

There were more eligible studies than was anticipated, though notably most of the studies were published within the past five years. The search strategy was piloted by supplementing the results of the electronic search with a search of reference lists of included papers. Good reliability between two independent raters, ensured that there was strong confidence in the conclusion that all relevant research was included in the systematic review and that conclusions from this review can be reliably drawn on synthesis of all the available evidence. Another strength of the review was that all included studies were quality assessed.

* + 1. Comparisons with Other Research on Psychopathology

Most studies focused on the role of within-child risk factors. Findings that children and adolescents who have VI and comorbid ID are at higher risk of psychopathology resemble those findings from the hearing-impaired community (Theunissen, 2014). This is also in line with findings in the general population (Nielsen et al., 2007). It is likely that higher cognitive ability is a protective factor against psychopathology for children with VI. Better cognitive reserves may enable the child to overcome the disability challenges arising from VI, such as difficulties navigating the physical environment. Further research is needed to understand variables that mediate the relationship between ID and psychopathology in children with VI.

Although some studies found that females who have VI were at increased risk of psychopathology compared to males with VI, the results were too inconsistent to form a conclusion. This contradicts findings in the sighted literature, where there is clear evidence that females are at higher risk of internalising disorder and males are at higher risk of externalising disorder. Mixed findings regarding the role of the severity of VI were surprising and not as anticipated from previous reviews, which indicated that more severe VI is associated with higher levels of maladjustment (Ammerman et al., 1986) and lower levels of self-concept and self-esteem (Augestad, 2017) in children with VI.

Findings that social communication difficulties are associated with increased psychopathology symptoms in children with VI are comparable with findings in the sighted literature (Humphrey et al., 2007). Further research is needed to understand variables that mediate the relationship between social communication and psychopathology in this group. For example, it is not known whether vision loss leads to lower mood, which leads to social withdrawal (Burlingham, 1979) or whether VI leads to social skill deficits, which leads to loneliness and low mood (Pinquart & Pfeiffer, 2013). No studies investigated ASD as a risk factor, despite findings that up to 40% of children with VI meet criteria for a diagnosis of ASD (Mukkaddes et al., 2007). Findings that increased self-concept and self-esteem and positive body image and attributional style and optimism were potential ‘protective factors’ against psychopathology is in line with findings in the general population though replication is required (Ezpleta et al., 2012; Young et al., 2017).

There has been a lack of research investigating the role of the social context (e.g. socioeconomic variables) with only one study investigating the role of socioeconomic status. There has also been a lack of research investigating the role of family factors. One study found that children’s feelings towards their fathers and teachers correlated with feelings of hopelessness (Erol & Ergun, 2013). No other studies addressed any family factors, such as parenting stress. This is a critical area for future research, as parental mental health is associated with psychopathology in the general population (Batenburg-Eddes et al., 2013) and parents of children with VI have been found to present above average levels of parenting stress (e.g. Jan et al., 1977; Sakkalou et al., 2017).

Although children and adolescents who have VI are likely to be at higher risk of psychopathology than TD peers, this systematic review has not found any risk or protective factors that are not found in the TD population. In line with findings in the sighted population, ID is an important risk factor for psychopathology in children who have VI. It is likely that variables related to vision (such as severity of VI, onset of VI, and specific disorders) as well as comorbidities play a role in the development of psychological distress in children with VI and high-quality studies are needed to investigate which children in this group are at the highest risk of psychopathology. It may be that a cumulative risk approach is most helpful in explaining risk in this group. The presence of VI may be the most important risk factor and quantity of subsequent risk factors contribute to the overall level of psychopathology. Multivariate research is needed to investigate the relative importance of different variables in predicting psychopathology within this group.

* + 1. Implications for Practitioners

The findings of the systematic review have mixed implications for practice and provision to support the mental health needs of children with VI. Children who have VI may be at high risk of psychopathology, but a lack of consensus regarding risk and protective factors mean that it is difficult to know where to target early support. Like in the sighted population, children with ID, are at a higher risk of psychopathology and this should be considered in service planning. It is important that teachers, parents and health professionals are aware that children who have VI may be at ‘high risk’ of psychopathology and should be prepared to identify difficulties as they present and early on. Early identification and appropriate clinical interventions may lead to improved outcomes in psychological wellbeing, adjustment and mental health (Department for Education, 2017). More research is needed to establish the risk and particularly protective factors of psychopathology and the patterns of psychopathology in children with VI. This may help to identify resource needs and appropriate interventions that can be effective with this population.

* + 1. Implications for Future Research

Longitudinal studies are needed to understand more about the developmental trajectory of psychopathology in children who have VI. Future research is required to address the outlined limitations, to understand the impact of VI on development, and how this may relate to psychopathology in children with VI. A research recommendation is that studies recruit clearly defined samples of children who have VI. For instance, including samples limited to children who have a congenital disorder of the peripheral visual system (CDPVS), where the origin of VI is in the globe, retina, or anterior optic nerve who do not have a comorbid neurological condition, would enable focus on the impact of the VI as cortical confounds and ID are reduced (Sonksen & Dale, 2002). Other studies could investigate psychopathology in samples of children who have Cerebral VI, additional diagnosis of ID, or later onset VI. The key recommendation is that samples are not mixed as each group may be discrete and face different challenges, unless the sample is extremely large and participant characteristics are well defined and used within a multivariate analysis. Clarity and consistency of sampling methodology and measurement tools will be needed to improve reliability and consensus across research in this field.

An increased understanding of the specific nature of psychopathology and related risk factors in children with VI is required to inform appropriate intervention planning and related service provision needs. VI specific service delivery that can respond to earlier risks rather than full blown psychopathology may be indicated, but this could be more difficult to deliver in countries that provide more generic child and adolescent mental health provision or that lack resources for any provision. Systematic and multivariate research into earlier preventative intervention in this area is required across the developmental span. Research with children who have mild-to-moderate VI, where functional impairment is less severe but where other factors (e.g. facial disfigurement) may lead to similar or different mental health difficulties, is also needed. The specific level of VI should be measured and described to investigate differences between different bandings of VI severity. A focus on primary school aged children with VI is also required, as most research in this area has focused on adolescents and middle childhood is thought to be a critical period of psychosocial development which is necessary for successful transition to adolescence (Zembar & Blume, 2009).

Exploration is warranted in relation to the role of sociodemographic variables, parental mental health, family factors, type of vision disorder, autism traits, and adaptive functioning, which may play a significant role in the development of psychopathology in children with VI. Zeanah et al., (1997) highlighted the importance of socioeconomic and family risks as well as risks related to the child, and there has been a lack of research in these areas in children with VI.

The findings of the systematic review were used to inform the design of a new empirical study. The new study aimed to extend knowledge of patterns of psychopathology in children with VI, by using a sample of children who had VI categorised as CDPVS and ‘low vision’ or ‘blindness’ according to ICD-10 criteria, with no known comorbid ID. This provided a more valid and less confounded sample than used in previous research. The new study also aimed to extend knowledge of risk factors relating to psychopathology in children with VI. It builds on previous research by incorporating a multivariate design and by investigating risk factors which have not yet been investigated in relation to psychopathology in children with VI, such as autism traits and adaptive functioning skills. Children with moderate to profound VI who were aged 8 to 11 were included, as there has been a lack of research with children with VI in this range. There has been a lack of research investigating parent and VI child agreement on standardised measures of mental health and so this was also investigated in the new study.

* + 1. Conclusions

The challenge of research on psychopathology in children with VI is that it often has different research aims, designs, samples, measurement, and analyses and that it is undertaken in many different countries, which may have contributed to the difficulty of comparing different studies and to the inconsistent findings. Children who have VI and ID may be at the highest risk of psychopathology and this is important to consider in the context of service planning. Future research should aim to further understand risk and protective factors in this population, using well-defined samples of children for whom VI is the ‘primary’ disability. Future research into the role of protective factors will serve to highlight areas for psychological intervention. Further research into the association of specific vision factors with psychopathology will help to identify whether children who have specific characteristics of VI are particularly at risk of psychopathology. As children with VI are at ‘high risk’ of ASD, this may be a potentially important risk factor in this group which requires further investigation. Research investigating effective interventions for children with VI who present with symptoms of psychopathology is also required.

1. Psychopathology profiles in children with congenital visual impairment
   1. Abstract

Children with visual impairment (VI) may be at increased risk of psychopathology, compared to typically developing peers (Harris & Lord, 2016). This study investigates patterns of psychopathology within children with VI, and potentially associated risk factors.

Thirty-four children (aged 8-11) with congenital disorders of the peripheral visual system, and their parents, were included in the study. Children had ‘low vision’ or ‘blindness’ (ICD-10), with no known neurological or physical comorbidities. Participants were asked to complete a questionnaire battery, designed to understand psychopathology profiles. Parents completed questionnaires which assessed internalising and externalising symptoms, social communication, adaptive functioning and quality of life. Children completed self-report questionnaires which assessed internalising symptoms and quality of life. Medical and demographic information were obtained, from parent report and medical records. A cross-sectional within-group design was used. Between-group analyses were used to compare sample and normative population scores.

The results indicated that children in the sample were at increased risk of psychopathology, compared to typically developing peers. Eleven children (33%) were at ‘high risk’ of an internalising disorder and 7 children (21.2%) were at ‘high risk’ of an externalising disorder, compared to 10% of the typically developing population. Children in the sample were particularly ‘at risk’ of separation anxiety. Risk factors associated with psychopathology were social communication difficulties, blindness, and adaptive functioning weaknesses. Gender, socioeconomic status and age were not associated with psychopathology symptoms. In the multivariate analysis, social communication difficulties was the only factor associated with increased psychopathology symptoms.

Findings add to the growing evidence base that children with VI are at ‘high risk’ of psychopathology. The recommendation is that a preventative approach is taken to support the emotional needs of children with VI. This may include supporting the development of a secure attachment style and social communication skills.

* 1. Introduction

Improving mental health in children is an international (World Health Organisation) and UK governmental priority (Future in Mind, Department of Health, 2015; Department for Education, 2017). Children with paediatric disorders are at increased risk of psychopathology (Royal College of Paediatrics and Child Health; RSPCH, 2018). An informed understanding of the impact of specific paediatric disorders on psychopathology is required, to plan appropriate support and intervention. It is estimated that 2 per 1,000 children and young people in the UK, are affected by visual impairment (VI) (Rahi & Cable, 2003). Congenital VI has a major impact on development, learning and autonomy. International research suggests that children and adolescents with VI may be at increased risk of psychopathology, compared to typically developing (TD) peers (Augestad, 2017).

Internalising and externalising disorders are the two main presentations of psychopathology in childhood (Patalay et al., 2015). Internalising symptoms consist of excessive control over one’s behaviours, emotions and thoughts, whereas externalising symptoms involve a lack of control over these (Nunes, Faraco, Vieira & Rubin, 2013). Internalising psychopathology may be expressed in the form of depression or anxiety, whereas externalising psychopathology may be expressed by aggressive, impulsive and challenging behaviours. Both forms of psychopathology may lead to limited social experience, creating obstacles for psychological development and predicting later psychiatric disorder (Blanken et al., 2017). Understanding psychopathology profiles in children with VI is important in developing effective interventions and achieving greater understanding of this forms the basis of this thesis.

* + 1. Visual Impairment (VI)

VI is commonly understood as an absence or lack of vision, which is chronic and cannot be medically restored or corrected. Previous research investigating psychopathology in children with VI has used differing criteria to define and recruit samples. This has included the use of country-specific disability criteria and parent reported VI, which makes it difficult to synthesise findings across studies. This study defined VI in relation to the International Classification of Diseases 2010 (ICD-10).

VI can be congenital (early or later onset) or acquired. It can be caused by conditions that affect the anatomy and function of the eye, the neural connections between the eye and the brain, or parts of the brain involved with vision processing. Congenital disorders of the peripheral visual system (CDPVS) originate in the eye (globe, retina, or anterior optic nerve) (Sonksen and Dale 2002). These are distinct from congenital cortical visual disorders (CVI), which originate in the brain substrate. VI is therefore heterogeneous in origin. It is often associated with comorbidities such as Intellectual Disability (ID), particularly in the context of CVI, due to the increased neurological involvement (Sonksen & Dale, 2002). In this study we included children with VI who have the lowest incidence of associated neurological impairment and ID and therefore excluded children exposed to potentially brain damaging events and those with ‘potentially complicated’ CDPVS[[1]](#footnote-1). This study included only children with ‘simple’ CDPVS who had no known additional neurological impairment. This reduced confounds and enabled investigation of psychopathology within a ‘primary’ VI sample (Sonksen & Dale, 2002).

* + 1. Visual Impairment and Psychological Distress

Psychological development and distress may be impacted by the functional limitations of VI and by atypical responding from others (Wallander & Varni, 1998). A diathesis-stress model postulates that distress arises when there is an imbalance between perceived competency and environmental demands. The risk of imbalance may be increased in children with VI, due to intra-psychological (e.g. negative thoughts about physical appearance, developmental delays and learning difficulties, impacts of comorbid neurological impairment) and interpersonal (e.g. higher levels of dependency upon others) factors, as well as by atypical responding from others (e.g. increased parental control, discrimination). These factors may contribute as sources of psychological distress (Hurre & Aro, 2000; Lifshitz, Hen & Weisse, 2007).

* + 1. Visual Impairment and Psychopathology

Children with VI may therefore be at increased risk of psychological distress, compared to TD peers. Some cross-sectional studies have concluded that children with VI have higher rates of depression and anxiety than TD peers (Huurre & Aro, 2000; Bolat, Dogangun, Yavuz, Demir & Kayaalp, 2011), whilst others have not (e.g. Kovacs, 1992; Yoshida, Ichikawa, Ichikawa & Hori, 1998; Halder & Datta, 2012). Mixed findings may be related to the sensitivity of measures used, as well as sample differences. Studies using the Strengths and Difficulties Questionnaire (SDQ) have produced more consistent findings. For example, Pinquart and Pfeiffer (2012) found significant differences between 16-year-olds with VI (*n*=158) and sighted controls: 13.5% were ‘at risk of psychiatric disorder’ compared to 5.1% of controls. Measures of broader constructs of psychopathology (e.g. SDQ) may be more sensitive than more specific measures (such as measurement of mood) in VI samples. Mixed findings may also be attributable to sample differences.

Pinquart and Pfeiffer (2011) concluded in a meta-analysis that adolescents with VI had lower emotional wellbeing than TD peers, with small effect sizes. No differences were found between children with VI and TD peers, though there were fewer studies. A more recent systematic review concluded that children and adolescents with VI were at increased risk of mental health difficulties (Augestad, 2017), though findings were mixed, and there was still a lack of studies investigating younger children. More research is needed to understand psychopathology profiles in children with VI who are in middle childhood. This is a critical period of social, emotional-behavioural, cognitive and physical skill development, which is necessary for successful transition to adolescence (Zembar & Blume, 2009). Many of the studies did not outline sufficient information about samples, which makes it difficult to assess generalisability or to explain the contradictory nature of the findings.

A UK study of 11-year-olds (*n*=189) (Harris & Lord, 2016) found that 18-29% of children with VI were at ‘high risk’ of psychiatric disorder, as measured by the SDQ, compared to 7-10% of TD children. This provides evidence that younger children with VI may also be at increased risk of psychopathology. Children were identified as having VI based on parent-report only and the authors highlighted that the sample was likely to have included children who had very mild VI. There is no information about the type of vision disorders, or age of onset of VI in the sample, so the representativeness is unclear.

Another confound of previous research is the inclusion of children who have ID. A substantial proportion of children with VI have associated non-ophthalmic disorders (Sonksen & Dale, 2002) and are up to ten times more likely to have an ID, than TD children (Nielsen, Skov & Jensen, 2007). ID is highly comorbid with psychopathology in the sighted literature (Emerson, 2003) and Harris and Lord (2016) found that children with VI who had ‘additional disability or special educational needs’, were at the highest risk of psychopathology. These findings provide rationale to exclude children with comorbid ID in future studies in order to focus on ‘primary’ VI. The current study therefore aimed to investigate patterns of psychopathology in a sample of school-aged children with ‘potentially simple’ CDPVS and ‘low vision’ or ‘blindness’ according to ICD-10 criteria, with no known comorbid ID.

As this was one of the first studies to investigate psychopathology profiles in school-aged children who have VI, it was important to explore the comparability of parent and child perspectives of child mental health to inform clinical decision-making, and future research studies. Self-report is thought to be important for adolescents, as they report higher levels of internalising symptoms compared to parent-report (Jensen et al., 1999). Parent-report is thought to be particularly useful to assess externalising disorders and to report on preadolescent children (Klein et al., 2005). A recent study found ‘moderate’ agreement between parent and child scores on the Revised Childhood Anxiety and Depression Scale (RCADS) (Ebesutani et al., 2011) and a different study found ‘moderate’ to ‘good’ agreement in Pediatric Quality of Life Inventory (PedsQL) scores (Varni, Limbers & Burnwinkle, 2007). Both studies included children aged 8 to 18 years and their parents. Little is known regarding comparability in the context of VI. This study therefore aimed to investigate agreement on these measures.

* + 1. Risk Factors Which May Be Associated with Psychopathology in Children Who Have VI

This study also set out to investigate possible risk factors associated with psychopathology in children with VI. This study aimed to investigate possible risks related to the child, in line with previous research. Previous research has demonstrated mixed findings regarding how the severity of VI associates with psychopathology. Some studies have found that more severe VI is associated with higher levels of internalising difficulties (Yoshida et al., 1998; Visagie, Loxton, Ollendick & Steel, 2013; Pinquart & Pfeiffer, 2014) though children with the most severe VI are at the highest risk of ID (Sonksen & Dale, 2002) which may have confounded findings. Some studies have not found that severity of VI makes a difference (Heyl & Hintermair, 2015; Pinqaurt & Pfeiffer, 2013). Mixed findings may be related to varying criteria used between studies to define VI.

Another risk factor of interest is gender. In line with findings in the sighted literature, some studies investigating psychopathology in children and adolescents with VI have concluded that boys are at higher risk of externalising difficulties (e.g. Heyl & Hintermair, 2015) and girls are at higher risk of internalising difficulties (e.g. Emam, 2013; Visagie et al., 2013; Garaigorobil & Bernaras, 2009). Other studies have not found gender differences (e.g. Teare, 1984; Erol & Ergun, 2013) which may be related to sample confounds. Most of the existing research examining gender differences in children with VI have used adolescent samples. Teare (1984) found no gender differences in psychopathology symptoms in school aged children with VI.

Another risk factor of interest is lower socioeconomic status. Children with disabilities are more at risk of social disadvantage than children without disabilities, and this is a known risk factor for psychopathology (Pinquart & Sorensen, 2000). Two studies have found no association between socioeconomic status and psychopathology symptoms in children with VI (Bakhla et al., 2011; Teare, 1984). This is not in line with findings in the TD population and this study aimed to investigate this further.

Another potentially important risk factor for psychopathology in children with VI is social communication difficulties. Children with VI tend to have poorer social skills than TD peers (Sacks, Kekilis & Garlord-Ross, 1992; Huurre & Aro, 1988). Functional difficulties in predicting the social intentions of others, due to a lack of visual input about facial expressions (Pinquart & Pfeiffer, 2013), may interact with higher levels of dependency on adults (Sacks et al., 1992) and fewer opportunities for socialisation, leading to increased levels of loneliness (Hadidi & Al Khateeb, 2013). Few studies have investigated the role of social communication in relation to psychopathology in children with VI, though lack of peer support (Pinquart & Pfeiffer, 2011), social comparison (Erol & Ergun, 2013), and reduced social skills (Runjic, Prcic & Alimovic, 2015) have been found to associate with increased psychopathology symptoms.

Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder characterized by impairments in social (social interaction and communication) and non-social (restricted and repetitive behaviours) domains (DSM-V, APA, 2013; ICD-1O, WHO, 1993). ASD is the most commonly reported developmental disorder co-occurring in children with VI (Ek, 2010), with an estimated prevalence rate of up to 40% (Mukaddes et al., 2007) compared to 1% of the sighted population. Social communication deficits are associated with psychopathology symptoms (Bora & Berk, 2016). Up to 73% of individuals with ASD have at least one comorbid mental health disorder (Leyfer et al., 2006). Children who have both VI and ASD traits may be at a greater combined risk of psychopathology that children who have VI alone.

Adaptive behaviours are skills that enable independence. Previous research has indicated that adaptive behaviours are reduced in children with VI (Papadopoulos, Metsiou & Agaliotis, 2011; Greenaway, Pring, Schepers, Isaacs & Dale, 2016). Reduced adaptive behaviours may be related to parents concerns about the safety of their children in engaging in independent activities (Papadopoulos et al., 2011) leading to overprotectiveness or underestimation of the child’s potential (Greenaway et al., 2016). A recent study found a correlation between adaptive behaviour and quality of life in children with VI (Bathelt, De Haan, Salt & Dale, 2018). To date, no studies have investigated the relationship between adaptive functioning and psychopathology in this group.

This study therefore aimed to understand whether more severe VI (‘blindness’), female gender, lower socioeconomic status, social communication difficulties, and weaker adaptive functioning was associated with psychopathology in children with VI. Knowledge of risk and protective factors specific to this group will be important in planning appropriate screening and intervention.

* + 1. Summary of Limitations of Previous Research

There are significant limitations in previous research investigating VI and psychopathology. This VI population is extremely heterogeneous and studies comparing children with and without VI do not always control for sample differences, other than presence of VI. Most studies are limited to specific geographical areas and have been recruited from specialist VI schools only. The vision characteristics of the samples (e.g. aetiological origin of VI, specific vision disorders, severity and onset of VI) are often insufficiently described, despite their potential differential developmental impacts. Due to the rarity of VI, the age range used is often broad and spanning multiple developmental periods, which may impact the validity of findings. There has been a lack of consideration of socioeconomic factors, despite findings that children with disabilities are at increased risk of social disadvantage (Pinquart & Sorensen, 2000). Additionally, a wide variety of measures of psychopathology are reported in previous studies, which may account for mixed findings.

* + 1. The Current Study

The current study aimed to address the limitations of previous research and to explore the psychopathology profiles of 8-11-year olds with VI, due to a lack of research in this age range. To understand the relationship between psychopathology and VI, specific criteria were used to select children with VI who have the lowest incidence of associated neurological impairment. Therefore, in this study, we included only children who had a ‘potentially simple’ CDPVS. Children who had a known comorbid ID, motor impairment or hearing impairment were excluded. This reduced neurological confounds and enabled a valid investigation of patterns of psychopathology in children with a primary VI (Sonksen & Dale, 2002). All children had ’low vision’ or ‘blindness’, according to ICD-10 criteria, enabling comparison between different level of severity of VI. Participants were recruited through two national/tertiary paediatric ophthalmological departments which drew on a patient population from a wide region, particularly Southern England and Midlands.

A questionnaire battery, designed to investigate psychopathology profiles in children, was used in a cross-sectional design. The aim of the study was to increase knowledge of the pattern of psychopathology and associated risk factors in children with VI. The analyses were predominantly within-group with some between-group analyses to compare scores in the sample with population normative means. The following research questions were developed and investigated to address the study aim:

1. Are school aged children with VI at increased risk of psychopathology compared to typically developing (TD) children and what patterns of psychopathology are found in this group?
2. How do parent report and child self-report of internalising symptoms and quality of life, compare in children with VI?
3. Are children who are ‘blind’ at higher risk of psychopathology than children who are less visually impaired and have ‘low vision’?
4. How do the potential risk factors of more severe VI, gender, lower socioeconomic status, social communication difficulties and lower adaptive functioning relate to psychopathology patterns in children with VI?

Findings informed the need for appropriate screening, timely diagnosis, and effective support and intervention, in children with VI.

* 1. Method
     1. Participants

Participants were recruited through two national tertiary paediatric ophthalmological services in London. Recruitment took place from September 2017 to February 2018. A priori power analysis indicated that a sample size of *n*=50 would required to detect between medium and large effect sizes at the normal power of .80, alpha of .05 (2-tailed) which was appropriate given the size of effects found in similar studies. This was initially determined to be feasible but ultimately a sample of *n*=34 was achieved.

* + - 1. Inclusion and exclusion criteria.

Children aged 8 to 11.11 years with congenital VI originating in ophthalmological disorders, referred here as ‘potentially simple’ CDPVS (disorders originating in the globe, retina or anterior optic nerve) were included in the study. To meet inclusion criteria for the study, the child also needed to have ‘low vision’ or ‘blindness’ according to ICD-10 criteria[[2]](#footnote-2). Details of the vision condition and severity of VI (binocular distance visual acuity and after refraction correction) were taken from the most recent ophthalmic report. Exclusion criteria for the study were: ‘potentially complicated’ CDPVS[[3]](#footnote-3), cortical VI, acquired VI, later onset congenital VI, Intellectual Disability (ID), hearing impairment (HI), and neurological motor impairment.

* + 1. Measures

Two measures of psychopathology were used to investigate internalising and externalising symptoms in the sample. Measures of quality of life, social communication, adaptive functioning, and demographic and medical variables were used to describe other child characteristics or possible risk factors for psychopathology in the sample. The self-report questionnaires were enlarged to the child’s preferred font size for children with ‘low vision’ and was administered orally by parents for ‘blind’ children.

* + - 1. Revised Children’s Anxiety and Depression Scale (RCADS).

The RCADS (Chorpita et al., 2000) is a 47-item measure of child internalising symptoms, which maps onto diagnostic nosology and indexes the features of the most prevalent anxiety disorders: separation anxiety disorder, social phobia, generalized anxiety disorder, obsessive-compulsive disorder and panic disorders, as well as depression (Ebesutani et al., 2011) (Appendix B). It is widely used as a screening and outcome measure in child and adolescent mental health services (CAMHS). The parent-report version is validated for use with children aged 6 to 18 years and the self-report version is validated for use from 8 years (Appendix C). Both versions were used in this study. Items are rated on a four-point Likert Scale and raw scores are converted into *t*-scores (in relation to age and gender) to determine whether there is a clinical concern (a *t*-score of 65 or more).

The RCADS has adequate internal consistency (all alphas in the .70 to .80 range) and test-retest stability (correlation coefficients .68 to .85) (Chorpita et al., 2000; Muris, Meesters & Schouten, 2002). The RCADS has not been validated for use with children with VI, though no items are directly dependent on vision for scoring. It has not been used previously with this child population in the reported literature.

* + - 1. Strengths and Difficulties Questionnaire – Parent Report (SDQ-P).

The SDQ-P is a 25-item screening tool used to identify children who are likely to meet criteria for a diagnosis of psychiatric disorder (Goodman, 1997) (Appendix D). Items are divided into five sub-scales which cover internalising symptoms (emotional symptoms and peer problems) and externalising symptoms (conduct problems and hyperactivity). Items are rated on a three-point Likert Scale. Each subscale generates a score which is assigned to one of three bands that indicate the probability that the child has a psychiatric disorder: normal/low risk; borderline/moderate risk, and abnormal/high risk. Cut-offs were calculated so that approximately 10% of cases are abnormal (Goodman, 1997). The SDQ-P has been found to have satisfactory properties in relation to internal consistency, test-retest reliability, inter-rater agreement, construct validity, and concurrent validity (Stone, Otten, Engels, Vermulst & Janssens, 2010). The SDQ-P is not validated for use with children with VI, though no items are directly dependent on vision for scoring. The SDQ has been used previously in VI studies (e.g. Pinquart & Pfeiffer 2012; 2014; Harris & Lord, 2016).

* + - 1. Paediatric Quality of Life Inventory (PedsQL).

The Paediatric Quality of Life Inventory (PedsQL) is a measure of health-related quality of life (Varni, Seid & Kurtin, 2001). Both parent-report (Appendix E) and child self-report (Appendix F) versions were used in this study. The PedsQL is comprised of 23 items which are summed to provide summary scores for physical health, psychosocial health, and overall health score. Scores are converted into percentages. The population mean score is 81.3% (*S.D*. 15.92) on parent-report and 82.9% (*S.D.* 13.16) on child report. Scores one standard deviation below the mean indicate poor quality of life (Varni, Burwinkle, Seid & Skarr, 2003), which is 69.7% for self-report and 65.4% for parent report. Internal consistency reliability is good (alpha .8 to .9). Validity has been demonstrated in that the PedsQL distinguishes healthy children and paediatric patients (Varni et al., 2001). The measure has not been validated for use with children with VI, though items are not vision dependent and it has previously been used in VI samples (e.g. Dahlmann et al., 2017; Wong, Machin, Tan, Wong & Saw, 2009).

* + - 1. Adaptive Behaviour Assessment System, Third Edition, Parent Report (ABAS-III).

The ABAS-III (Harrison & Oakland 2015) measures functional skills that are required for daily living (Appendix G). Each item is rated on a four-point Likert Scale. In the version for children aged 5-21 years, items are organised into the subscales which are combined into three composite scores: Conceptual, Social, and Practical. These are combined to calculate a General Adaptive Composite. Composites have a mean of 100 and a standard deviation of 15. Reliability is good (alpha .97 to .99) in terms of internal consistency and inter-rater reliability (.82 to .81). Concurrent validity is also good (0.70) (Harrison & Oakland, 2015). To our knowledge, the ABAS-III has not previously been used with samples of children with VI, though the previous version (ABAS-II) has been used with this population (e.g. Greenaway, Pring, Schepers, Isaacs & Dale, 2017).

The ABAS has not been validated for use with children with VI. There were three items requiring vision which were removed and prorated[[4]](#footnote-4). Scores were prorated by calculating the mean item score of the subscale for each participant and replacing the missing item with the mean score. Some items asked about reading and writing skills and parents were asked to score these in relation to braille skills where appropriate[[5]](#footnote-5).

* + - 1. Social Responsiveness Scale 2, School Age Children Parent Report (SRS-2).

The SRS-2 (Constantino, 2005) is a 65-item parent-report questionnaire (Appendix H). It measures symptoms indicative of ASD in children aged 4 to 18. Each item is scored on a four-point Likert Scale, generating a total score ranging from 0 to 195. Scores can also be generated for five symptom domains. Results can be converted into *t*-scores to determine severity of social communication difficulties in relation to age and gender norms (Constantino & Gruber, 2012). A total raw score cut point value of 70 is associated with a sensitivity value of .78 and a specificity value of .94 for ASD diagnosis (Constantino, 2005), suggesting that the SRS-2 is a robust instrument for discriminating between individuals with and without ASD. A *t*-score of 60 or more is indicative of clinical concerns.

To our knowledge, the SRS-2 has not been used previously in research studies with children who have VI. It was felt that it could be used validly within this sample, as there were less vision-dependent items (e.g. eye contact, non-verbal gestural communication) than found in other measures of social communication. Two items that are vision dependent were removed and prorated[[6]](#footnote-6). Scores were prorated by calculating the mean item score of the subscale for each participant and replacing the missing item with the mean score.

* + - 1. Demographics and Medical Information Questionnaire.

A questionnaire was constructed to collect parent-reported detailed demographic (Appendix I) and medical information (Appendix J). Parents gave consent for medical records to be accessed where there was missing data.

* + 1. Procedure

Initial identification was done by clinicians of potential participants via patient databases and medical records. This involved reviewing information relating to age, visual diagnosis and visual abilities, as well as relevant paediatric information, to determine whether potential participants met inclusion criteria. Potential participants were cross-referenced with upcoming clinical appointments. Eligible participants who were not due to attend an appointment during the data collection period were sent an information pack about the study in the post, which included information about the study for parents (Appendix K), information for children (Appendix L), an expression of interest form (EOI) (Appendix M), and a return stamped envelope. Eligible participants who attended routine clinical appointments during the data collection period were given the information pack during the appointment.

Families interested in taking part were asked to send the EOI to the research team. A member of the research team then phoned parents to answer any questions and to take verbal consent. Parents were asked to provide an address to have the questionnaire pack posted to. Parents were also asked about accessibility for the child questionnaires. The questionnaire pack which included a consent form (Appendix N) and questionnaires was then posted to the child and parent’s home. Participants were asked to return the questionnaires within two weeks. Follow-up phone calls were made if the questionnaire had not been returned by two weeks.

* + 1. Statistical Methods

Preliminary analysis confirmed that most of the data was normally distributed. The SRS variable was not normally distributed and was transformed into normal distribution, using a square root algorithm. Descriptive statistics were used to describe psychopathology profiles in the whole sample. Cronbach Alpha’s were calculated to assess the reliability of the scales in this population, as indicated by their internal consistency. Missing data was excluded pairwise in the analyses. For missing questionnaire items, a research assistant contacted participants where possible by telephone to obtain any missing data. For any other remaining missing data, instructions in the scoring manual of each questionnaire were followed, which normally permitted pro-rata scoring when a minority of items were missing. Scores were prorated by calculating the mean item score of the subscale for each participant and replacing the missing item with the mean score. Most parent questionnaires were completed and included in the analyses. One parent did not complete the SDQ or SRS 2. One child was unwilling to complete the RCADS-C and one child was unwilling to complete the RCADS-C or PedsQL-C.

One sample *z*-tests were then used to compare study population means to normative data means. Data means based on raw scores were taken from published papers for the RCADS (US sample, aged 8-12; Ebesutani et al., 2011), SDQ (UK sample, aged 5-15, Goodman, 2001), PedsQL (US sample, aged 5-16, Varni et al., 2003), and SRS 2 (UK sample, aged 5-8, Wigham et al., 2012). Data using the closest age range possible was taken. Normative data means based on standardised scores were taken from the test manual for the ABAS III (Harrison & Oakland, 2013) as raw normative data was not available. Cohen’s *d* was calculated to describe the effect size[[7]](#footnote-7) Chi-square goodness of fit tests were used to compare the proportions of children who were ‘at risk’ or ‘not at risk’ of psychopathology in the study sample, compared to proportions in the TD population.

To examine the agreement between parent report and child self-report on the RCADS and PedsQL, Intraclass Correlations (ICC)[[8]](#footnote-8) were used. In addition, mean differences between parent report and child self-reports were calculated by using dependent sample *t*-tests to explore differences in scores. Independent-samples *t*-tests were then used to investigate whether there was a difference in level of psychopathology between children with ‘low vision’ and ‘blindness’. Preliminary analyses confirmed normal distribution and homogeneity of variance. A Bonferonni correction was applied to account for multiple comparisons, setting a new p-value of .016. There were no differences in gender distribution or age between groups. Missing data was excluded pairwise.

Partial correlations (controlling for age) were conducted to investigate the possible associations between the possible risk variables and dependent/outcome measures of psychopathology[[9]](#footnote-9). Assumptions for normality linearity and homoscedasticity were met. Where there were associations, variables were taken forward into the multiple regression analyses. Preliminary analyses were conducted to ensure no violation of the assumptions of normality, linearity, multicollinearity homoscedasticity, and independence of residuals were met. Missing data was excluded pairwise. The independent variables were: gender, vision level, socioeconomic status, social communication ability (measured by the SRS) and adaptive functioning skills (measured by the ABAS). The dependent variable was psychopathology symptoms (as measured by the SDQ).

The project received full Ethical Approval (Appendix O) including full Health Research Authority approval (Appendix P), and site-specific research and design department approvals from the recruitment sites (Appendix Q and R).

* 1. Results
     1. Participant Characteristics

Thirty-four parents and 32 children took part in the study (21 male, 13 female). The mean age of the child was 9.68 years (*S.D.* 1.14, age range 8 to 11.75 years). Most parents in the study were married (*n*=30, 88.2%). Most parents (*n*=33, 97.1%) reported that their child received support from a VI specialist in school. A minority of parents reported that their child received support from a VI specialist at home (*n*=7, 20.6%) with most reporting none (*n*=27, 79.4%).

Information about parent-reported socioeconomic status (Table 1), parent-reported sample characteristics (Table 2), vision disorders (Table 3) and vision level characteristics (Table 4) according to a combination of parent report and medical records is presented. Thirty-two parents (94%) provided information about the child’s birth weight and weeks’ gestation. The average birth weight in the sample was 3231 grams (S.D. 510) and the average weeks’ gestation was 39.5 weeks (*S.D.* 2.1). This is in line with findings in the general UK population (Norris et al., 2017).

Table 1

*NS-SEC Analytic Class (Prevalin & Rose, 2002)[[10]](#footnote-10)*

|  |  |
| --- | --- |
| Analytic Class | Frequency (*n* = 34) |
| 1: Higher managerial/professional occupations | 10 (29.4%) |
| 2: Lower managerial/professional occupations | 12 (35.3%) |
| 3: Intermediate occupations | 9 (26.5%) |
| 4: Small employers and own account workers | 3 (8.8%) |
| 5: Lower supervisory and technical occupations | 0 |
| 6: Semi-routine occupations | 0 |
| 7: Routine occupations | 0 |
| 8: Unemployed | 0 |

Table 2

*Sample Characteristics*

|  |  |
| --- | --- |
| Type of School | Frequency (*n* = 34) |
| Mainstream school (no additional support) | 4 (12%) |
| Mainstream school (with additional support) | 21 (62%) |
| Mainstream school with VI unit | 5 (14%) |
| VI school | 2 (6%) |
| Other specialist school | 1 (3%) |
| Missing | 1 (3%) |
| Parent Reported Social Communication/Language Difficulties | |
| Yes | 9 (27.3%) (6, 18.2%, reported diagnosed ASD) |
| No | 24 (72.7%) |
| Missing | 1 (2.9%) |
| Parent Reported Sleep Difficulties | |
| Severe | 4 (11.8%) |
| Mild | 10 (32.4%) |
| None | 18 (52.9%) |
| Missing | 2 (5.9%) |
| Has the Child Ever Accessed Mental Health Support | |
| Yes | 15 (44.1%) |
| No | 18 (52.9%) |
| Missing | 1 (2.9%) |
| Type of Support | |
| Charity | 4 |
| School | 8 |
| CAMHS | 3 |
| Specialist VI psychologist | 3 |
| Ethnicity | |
| White or White British | 25 (73.5%) |
| Asian or Asian British | 5 (14.7%) |
| Mixed | 3 (8.8%) |
| Black African/Caribbean | 1 (2.9%) |

Of the children receiving additional support in mainstream schools, parents reported that their child received between 10 and 40 hours of TA support per week (mean: 27.5 hours).

Table 3

*Vision Disorders of the sample[[11]](#footnote-11)*

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
| Origin |  |  | Vision Disorder | Frequency |
| Retinal |  |  | Stargardt Disease  Oculocutaneous albinism  Cone-Rod Dystrophy  Achromatopsia  Leber’s congenital amaurosis  Retinal Dystrophy  Other retinal | 3  6  1  4  4  3  4 |
| Total |  |  |  | 25 |
| Globe |  |  | Peter’s anomaly  Glaucoma  Anophthalmia/Microphthalmia  Enpothelial dystrophy  Aniridia  Pseudophakia | 2  1  2  1  1  1 |
| Total |  |  |  | 8 |
| Optic nerve |  |  | Optic atrophy | 1 |
| Total |  |  |  | 1 |

Table 4

*Vision level characteristics of the sample*

|  |  |
| --- | --- |
| Has sight reduce over time | Frequency |
| Yes | 8 (23.5%) |
| No | 23 (67.6%) |
| Missing | 3 (8.8%) |
| Visual acuity categories (ICD-10) | Frequency |
| Moderate VI (logMAR 0.5-1.0) | 18 (52.9%) |
| Severe VI (logMAR 1.1-1.3) | 7 (20.6%) |
| Blindness (logMAR 1.4 or worse) | 9 (26.5%) |

Table 5

*Recruitment and Participation Rate*

|  |  |  |  |
| --- | --- | --- | --- |
|  | Recruitment site 1a | Recruitment site 1b | Recruitment site 2 |
| Patient database (postal recruitment) | Packs sent: 40  EOI received: 17  No. participated: 13 | Packs sent: 50  EOI received: 6  No. participated: 5 |  |
| Routine clinic appointment (face to face recruitment) | Patients approached: 0  EOI received: 0  No. participated: 0 | Patients approached: 10  EOI received: 9  No. participated: 8 | Patients approached: 28  EOI received: 24  No. participated: 8 |

Details of recruitment and participant rates are presented in Table 5. One hundred and twenty-eight parents were ascertained and approached about the study. Fifty-six parents (43.8%) returned an EOI. Of these, three were excluded as they did not meet eligibility criteria for the project[[12]](#footnote-12). Thirty-four (64.2%) completed the project and 19 (35.8%) did not. The recruitment and participation rates were 43.8% and 64.72% respectively. The overall response rate was therefore 27.2%.

* + 1. Are children with VI at increased risk of psychopathology compared to typically developing (TD) children and what patterns of psychopathology are found in this group?
       1. Revised Children’s Anxiety and Depression Scale (RCADS).

Thirty-four (100%) parents and 32 (94.1%) children completed the RCADS (Table 6). According to parent-report, eleven children (33.4%) scored in the clinical range for internalising symptoms.

Table 6

*Number of participants scoring in the clinical range on each subscale of the RCADS*

|  |  |  |
| --- | --- | --- |
|  | Parent Report (*n* = 34) *n* (%) | Child Self-Report (*n* = 32) *n* (%) |
| Separation Anxiety  Social Phobia  Generalised Anxiety  Panic  Obsessive Compulsive  Depression | 10 (29.4%)  1 (2.9%)  0  0  0  0 | 4 (12.5%)  2 (6.3%)  2 (6.3%)  2 (6.3%)  0  4 (12.5%) |

* + - 1. Strengths and Difficulties Questionnaire (SDQ).

Thirty-three (97.1%) parents completed the SDQ (Table 7) shows the numbers of children scored in the ‘high risk of psychiatric disorder’ range).

Table 7

*Number of participants scoring in the ‘high risk of psychiatric disorder’ range on the SDQ*

|  |  |
| --- | --- |
|  | Parent Report (*n* = 33) *n (%)* |
| Any  Internalising Disorder  Externalising Disorder | 8 (24.2%)  11 (33.3%)  7 (21.2%) |

* + - 1. Pediatric Quality of Life Scale (PedsQL).

Thirty-four (100%) parents and 33 (97.1%) children completed the PedsQL (Table 8).

Table 8

*Number of participants scoring below the cut-off for good quality of life on the PedsQL*

|  |  |  |
| --- | --- | --- |
|  | Parent Report | Child Self-Report |
| *n* (%) | 16 (47.1%) | 19 (57.6%) |

* + - 1. Social Responsiveness Scale (SRS).

Thirty-three (97.1%) parents completed the SRS (Table 9). Overall, 15 children (45.5%) scored in the clinical concern range, from mild to severe.

Table 9

*Number of participants scoring in each score category of the SRS*

|  |  |
| --- | --- |
|  | Parent Report |
| ‘normal’ range | 18 (54.5%) |
| ‘mild’ range | 5 (15.2%) |
| ‘moderate’ range | 6 (4 (18.2%) |
| ‘severe’ range | 4 (12.1%) |

* + - 1. Adaptive Behaviour Assessment System (ABAS III).

Thirty-four (100%) parents completed the ABAS III (Table 10).

Table 10

*Number of participants scoring below the average range for adaptive functioning on the ABAS III.*

|  |  |
| --- | --- |
|  | Parent Report |
| *n* (%) | 17 (50%) |

* + - 1. Reliability

To check the reliability of the questionnaires used in this study, the Cronbach’s alpha coefficient was calculated. A minimum standard of 0.70 was assumed for the Cronbach’s alpha coefficients for adequate internal reliability. Reliability was found to be adequate across all measures (Table 11).

Table 11

*Cronbach’s Alpha Coefficients*

|  |  |
| --- | --- |
|  | Cronbach’s Alpha |
| RCADS-P | .94 |
| RCADS-C | .96 |
| SDQ | .76 |
| PedsQL-P  PedsQL-C  SRS | .92  .92  .94 |
| ABAS III | .98 |

* + 1. Comparisons of Questionnaire Scores and Population Norms.

The questionnaire scores of the psychopathology measures are shown in Table 12 and the questionnaire scores of independent or explanatory (or ‘risk’) variables shown in Table 13. One sample *z* tests were used to compare the parent-reported scores of the study population with normative data. The children with VI showed significantly more symptoms of psychopathology on the RCADS-P and SDQ compared to normative data, with medium effect sizes. The children with VI had lower quality of life, more social communication difficulties, and weaker adaptive functioning skills, compared to normative data, with large effect sizes. These differences were considered potential risk factors and later entered into the regression analyses.

Table 12

*Comparisons of mean psychopathology scores of children who have VI and population norms, using one sample z tests*

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
|  |  | Children with VI | Normative Data | One sample Z test |
| RCADS-P | *N*  Mean Score  Standard Deviation  Cohen’s *d* | 34  28.79  17.65 | 378[[13]](#footnote-13)  22.49  12.9 | *z* = 2.85, *p* = .004,  .50 |
| SDQ | *N*  Mean Score  Standard Deviation  Cohen’s d | 33  12.66  6.8 | 10298[[14]](#footnote-14)  8.4  5.8 | *z* = 4.22, *p* < .001,  .74 |

Table 13

*Comparisons of mean quality of life, adaptive functioning and social communication scores of children who have VI and population norms, using one sample z tests*

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
|  |  | Children with VI | Normative Data | One sample Z test |
| PedsQL-P | *N*  Mean Score  Standard Deviation  Cohen’s *d* | 34  66.31  18.2 | 10070[[15]](#footnote-15)  81.34  15.92 | *z* = -5.50, *p* < .001,  .94 |
| SRS 2 | *N*  Mean Score  Standard Deviation  Cohen’s *d* | 33  55.97  29.2 | 500[[16]](#footnote-16)  29.82  16.64 | *z* = 9.03, *p* < .001,  1.57 |
| ABAS III | *N*  Mean Score  Standard Deviation  Cohen’s *d* | 34  85.47  14.47 | ?[[17]](#footnote-17)  100  15 | *z* = -5.64, *p*< .001,  .99 |

* + - 1. RCADS.

A chi-square goodness-of-fit test indicated that there was a significant difference in the proportion of children who were in the clinical range for an internalising disorder (separation anxiety) based on parent-report identified in the current sample (*n*=10, 29.4%) as compared with the value of 7% specified in the normative data: χ2 (1, *n*=34) = 26.23, *p* < .0001 (Figure 1).

*Figure 1.* Proportion of children scoring in the clinical range for separation anxiety on the RCADS in the VI group) compared to the normal population

* + - 1. SDQ.

A chi-square goodness-of-fit test indicated that there was a significant difference in the proportion of child ‘at high risk of psychiatric disorder’ according to the SDQ identified in the current sample (*n* = 8, 24.2%) and ‘at medium risk of psychiatric disorder’ (*n* = 8, 24.2%) as compared with the value of 10% ‘at high risk’ and 10% ‘at medium risk’ specified in the normative data (Goodman, 2001), χ2 (1, *n*=33) = 7.44, *p* = .006 (Figure 2).

*Figure 2.* Proportion of children ‘at risk’ of psychopathology according to SDQ score in the VI group) compared to the normal population

* + 1. How Do Parent Report and Child Self-Report of Internalising Symptoms and Quality of Life, Compare in Children with VI?

The average level of agreement between parent report and child self-report was moderate for the RCADS (ICC= 0.18-0.87, M=.48, S.D .18) and for the PedsQL (ICC= 0.02-0.93, M=.53, S.D .27). Paired-samples t-tests were conducted to compare parent report and chid self-report. There was a significant difference between parent-report (*M* = 43.7, *S.D.* = 10.7) and self-report (*M* = 32.77), *t* (29 = 2.67, *p* = .012) on the RCADS, with parents reporting more difficulties than the child. The mean decrease in scores from parent to child was 10.93 with a 95% confidence interval ranging from 2.54 to 19.33. The eta square statistic (.2) indicated a small effect size. There was no statistically significant difference between parent-report (*M* = 65.4, *S.D.* = 18.02) and self-report (*M* = 66.38), *t* (29 = -.31, p = .76) on the PedsQL.

* + 1. Are Children Who Are ‘Blind’ at Higher Risk of Psychopathology than Children who are Less Visually Impaired and Have ‘Low Vision’?

Independent samples *t*-tests were conducted to compare total psychopathology questionnaire scores for children with ‘low vision’ (logMAR 0.5 to 1.3) and ‘blindness’ (logMAR 1.4 or worse). (Table 14). There was no significant difference in age between the ‘low vision’ (*M* = 115.9, *SD* = 14.2) and ‘blind’ (*M* = 116.8, *SD* = 12.5) groups: *t* (32) = -.17, *p* = .65. There was no significant difference in gender distribution between the ‘low vision’ (*M* = 1.3, *SD* = .5) and ‘blind’ (*M* = 1.4, *SD* = .5) groups: *t* (32) = -.43, *p* = .49. There was no significant difference in socioeconomic status between the ‘low vision’ (*M* = 3.1, *SD* = 1.0) and ‘blind’ (*M* = 3.1, *SD* = 1.0) groups: *t* (32) = .022, *p* = .95.

Table 14

*Independent Sample t-tests between children with ‘low vision’ and ‘blindness’*

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
|  |  | ‘Low Vision’ | ‘Blindness’ | Independent Sample *t*-test |
| RCADS-P | *N*  Mean Score  Standard Deviation  Cohen’s d | 25  27.16  17.67 | 9  33.33  16.88 | *t* (32) = -.89,  *p* = .38  .36 |
| RCADS-C | *N*  Mean Score  Standard Deviation  Cohen’s d | 24  31.21  24.66 | 7  37.29  14.49 | *t* (29) = -.62,  *p* = .54  .49 |
| SDQ | *N*  Mean Score  Standard Deviation  Cohen’s d | 24  10.96  6.59 | 9  17.22  5.4 | *t* (31) = -2.54,  *p* = .016  .98 |

Children who were ‘blind’ had higher mean scores on the SDQ, indicative of more difficulties, than children with ‘low vision’ across all subscales. A Bonferonni correction was applied to account for multiple comparisons, setting a new p-value of .016. The difference was not statistically significant on the RCADS. There was a significant difference in scores for ‘blind’ and ‘low vision’ groups on the SDQ. The effect size of the differences in the means (mean difference = 15.94, 95% CI: -11.09 to -.81) was large (cohen’s *d* = .98). A post hoc power analysis indicated a power of .80, alpha of .05 (one-tailed), indicating sufficient power to perform the analysis. For the RCADS, post hoc power analyses indicated a power of .23-.30, alpha of 0.05 (one-tailed), indicating that the analyses were not powered to detect the smaller effects.

* + 1. How do the potential risk factors of more severe VI, gender, lower socioeconomic status, social communication difficulties and lower adaptive functioning relate to the psychopathology patterns in children with VI?

The relationships between the explanatory (potential risk) variables of adaptive functioning (ABAS), social communication (SRS), VI severity (‘low vision’ and ‘blindness’), socioeconomic status (analytical classes), and gender and the SDQ-Total and Internalising symptoms and total Internalising symptoms (RCADS-P) with were investigated using a partial Pearson product-moment correlation coefficient controlling for age (Table 15)

|  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- |
|  | VI severity | Gender | Demographics | ABAS | SRS |
| SDQ | -.36\* | -.23 | -.20 | -.39\* | .75\*\* |
| RCADS-P | -.20 | -.19 | .10 | -.15 | .42\* |

*Table 15*

*Partial correlations between explanatory risk factors and dependent psychopathology measures*

\*\*significant at p <.001, \*significant at p < .05

There was a strong positive correlation between SRS and SDQ (*r*=.75, *n*=33, *p* < .001). There were moderate negative correlations between VI severity and SDQ score (*r*=-.37, *n*=33, *p* = .04) and ABAS and SDQ score (*r*=-.39, *n*=34, *p* = .03). There was a weak correlation between gender and SDQ score (*r*=-.23, *n*=34, *p* = .21) and a weak positive correlation between socioeconomic status and SDQ (*r*=.20, *n*=34, *p* = .28). There was a moderate positive correlation between SRS and RCADS (*r*=.42, *n*=33, *p* = .02). There were weak nonsignificant negative correlations between VI severity and RCADS (*r*=1.20, *n*=33, *p* = .28) and ABAS and RCADS (*r*=-.15, *n*=34, *p* = .41). There was a weak correlation between gender and SDQ (*r* = -.19, *n*=34, *p* = .28) and a weak nonsignificant positive correlation between socioeconomic status and SDQ score (*r*=.10, *n*=34, *p* = .56).

* + - 1. Hierarchical multiple regressions.

As the explanatory risk factors were more strongly associated with SDQ than RCADS, the SDQ only was taken forward into the next analysis. A hierarchical multiple regression was used to investigate whether the explanatory factors of SRS, ABAS, VI severity, gender, and socioeconomic status predicted the outcome variable of SDQ, controlling for age. Age was entered in step one, explaining 0.1% of the variance in the total SDQ score. After the additional variables were added at Step 2, the total variance explained by the model as a whole was 59.8%, *F* (6, 25) = 6.20, *p* < .001. In the final model, only one variable was statistically significant and explained most of the variance in the model (SRS, beta = .71, *p* < .001). A post hoc power analysis indicated a sufficient power of .99, alpha of .05 (2-tailed). Entered on its own, SRS explained 56% of the variance in the SDQ score.

Taking this finding further, secondary hierarchical multiple regressions were performed to investigate the ability of SRS score to separately predict internalising and externalising symptoms (SDQ) after controlling for age. When the internalising scale of the SDQ was the dependent variable, the total variance explained by SRS was 25.8%, *F* (2, 30) = 5.2, *p* = 0.01 (beta .50, *p* = .003). A post hoc power analysis indicated a power of 0.67, alpha of .05 (2-tailed), indicating that the analysis was underpowered. When the externalising scale of the SDQ was the dependent variable, the total variance explained by SRS was 60.6%, *F* (2, 30) = 23.1, *p* < .001. (beta .76, *p* < .001). A post hoc power analysis indicated a power of 0.99, alpha of .05 (2-tailed), indicating sufficient power to perform the analysis.

* 1. Discussion

This study provided a novel cross-sectional investigation into the pattern of psychopathology and the potential risk factors associating with the pattern, within a sample of children who had ‘low vision’ and ‘blindness’ (ICD-10). The children were 8-11 years of age, with VI deriving from the paediatric disorders of ‘potentially simple’ CDVPS. They had no known additional comorbid impairment and were estimated to be within the normal intellectual range.

* + 1. Psychopathology Profiles in the Sample

The results showed that the children with VI in this study were at increased risk of psychopathology, compared to TD peers. One third of the sample scored in the clinical range for internalising symptoms on the RCADS, according to parent report. A quarter of children in the sample scored in the ‘high risk of psychiatric disorder’ category on the SDQ, compared to a lower proportion (10%) of the TD population. The findings are broadly in line with or slightly higher than other studies using the SDQ, in adolescent VI samples (Pinquart & Pfeiffer, 2012; Pinquart & Pfeiffer, 2014) and in a subsample of 11-year-olds with VI (*n*=87) without additional special educational needs, where 18% were at ‘high risk’ (Harris & Lord, 2016). Findings of moderate agreement between parent report and child self-report scores, indicate the value of using both parent and child reporting for preadolescent children who have VI, who may have a unique perspective and insight into their internal world. Importantly, the majority of children in the sample did not score in the clinical range on either measure of psychopathology, suggesting that protective factors may be operating in this age range.

On the SDQ, children were found to be ‘at risk’ of both internalising (*n*=11, 33.3%) and externalising disorders (*n*=7, 21.2%). The cognitive theory of anxiety disorders (Beck, 1985) suggests that anxiety is associated with danger appraisal and beliefs that one cannot cope with threat, leading to anxiety. Anxiety leads to the attempt to reduce danger through avoidance or safety behaviours, which prevents disconfirmation of danger beliefs, reinforcing fears. It is feasible that the world is interpreted as particularly threatening by a child with VI (King, Josephs, Gullone, Madden & Ollendick, 1994) due to the functional limitations of VI. The child is at increased risk of physical danger and may have severe difficulty in discerning the environment visually to assess and respond to external threats, leading to increased anxiety. Whilst this may suggest a raised predisposition for anxiety in children with VI, the majority of children in this study did not present with clinically significant difficulties with anxiety and protective factors may be playing a buffering role.

A clinically relevant finding is that almost a third of the parents scored their child to be in the clinical range for separation anxiety on the RCADS. This is in line with findings of elevated rates of separation anxiety in children with paediatric disorders (Campo et al., 2004). Bowlby (1969) postulated that the attachment formation is delayed in children with VI; supported by findings of delayed separation anxiety in this group (Troster & Brambring, 1992). Attachment theory describes the innate tendency of an infant to seek proximity to an attachment figure (Bowlby, 1969). Functions that may be important for healthy development of attachment formation may be jeopardised or more limited; for instance, early eye contact is important in the development of joint attention (Tadic, Pring & Dale, 2009) and vision is needed for infants to communicate with the primary caregiver. Attachment formation may be further affected in children with VI (Hindley & Salt, 2007) due to the lack of visual scanning to maintain proximity and assess safe distance from the caregiver. Reduced nonverbal communication, less responsiveness, and fewer initiations with the primary caregiver may also impact attachment formation. It is also possible that separation anxiety is caused by the physical risks for the child with VI when having to navigate independently.

Parental factors may also be relevant, though this was not explored in this study. Howe (2006) argues that the mother-VI child interaction is negatively impacted by a VI related ‘communication barrier’ which can lead to maternal distress and an increased risk of insecure attachment, as the mother feels unable to understand the child’s communication cues and respond appropriately to her child’s needs and desires. Anxious parental cognitions and behaviours may be related to the development and maintenance of childhood anxiety (Apetroaia, Hill & Creswll, 2015). Parents of children with VI may have negative expectations of coping skills, which could lead to behaviours (e.g. parental overcontrol, modelling of anxiety and anxious rearing) that impede the development of autonomy. This may interact with separation anxiety between the parent and the child (McLeod, Wood, & Weisz, 2007; Creswell et al., 2011).

* + 1. Risk Factors Associated with Psychopathology

Children who were ‘blind’ (*n*=9) were at a significantly higher risk of psychopathology, and particularly of externalising symptoms, than children who had ‘low vision’ (*n*=25)*.* Previous research has demonstrated mixed findings regarding the impact of VI severity on psychopathology symptoms, which may be explained by differences in VI definition between studies. For example, Pinquart and Pfeiffer (2014) found no difference in psychopathology symptoms between ‘blind’ and ‘low vision’ adolescents but did not define ‘blindness’ and the ‘blind’ group may have included adolescents with more functional vision than the current study.

Only the ‘risk’ factor of social communication difficulties (as measured by the SRS 2), as an independent or explanatory factor in the analyses, was found to significantly explain the variance in psychopathology symptoms in the multivariate analysis and it was particularly predictive of externalising symptoms. The findings that social communication difficulties were associated with increased psychopathology is in line with reported findings in the VI literature (Pinquart & Pfeiffer, 2011; Runjic et al., 2015). Social communication difficulties may conceivably arise from functional difficulties in predicting the social intentions of others, due to a lack of visual input about facial expressions and intention behaviour of the other, (Pinquart & Pfeiffer, 2013). This could interact with higher levels of dependency on adults (Sacks et al., 1992), fewer opportunities for socialisation (Halen, 2004), and increased levels of loneliness (Hadidi & Al Khateeb, 2013).

This is potentially an important finding, as up to 40% of children with VI are diagnosed with ASD by school-age (Mukkades et al., 2007) and ASD is highly comorbid with psychopathology in the sighted population (Simonoff et al., 2008). Children with ASD are substantially less likely to be securely attached to their caregivers than TD children (Rutgers, Bakermans-Kranenburg, Ljzendoorn & Berckelaer-Onnes, 2004) and this may increase risks for children with VI and ASD traits. There may be additional relational factors as lack of social cues exhibited by the autistic child arouses less insight and reflections among parents into the narratives of their child, which may hinder further the parent-child interactions (Hutman, Siller & Sigman, 2009; McKenzie & Dallos, 2017). As VI may also increase the risk of attachment difficulties, it is conceivable that VI, ASD traits and insecure attachment styles, including increased parental anxiety, interact or compound together to increase the risk and severity of aspects of psychopathology in this subgroup of the population.

There was some evidence that adaptive functioning (as measured by the ABAS-III) was associated with psychopathology, and particularly externalising symptoms, in the group. Gender as a possible risk factor did not show significant associations with psychopathology symptoms. The findings of no gender differences are not in line with some other studies which have found girls with VI to be at higher risk of internalising difficulties (e.g. Huurre & Aro, 1988; Emam, 2013; Pinquart & Pfeiffer, 2013). Most of the existing research used adolescent samples and Teare (1984) found no gender differences in psychopathology symptoms between school-aged girls and boys with VI.

Taken together, the results indicate that the children in the sample, whatever their level of VI, could be at elevated risk of psychopathology and particularly of separation anxiety. It is likely that there were also protective factors operating as there were within sample individual differences and the majority of children did not reach clinical levels of psychopathology. In this sample potential risk factors of gender (i.e. females) do not appear to associate with increased risk of internalising difficulties compared to males and socioeconomic status (i.e. lower socioeconomic status) does not appear to associate with increased risk, though the total sample is skewed towards higher socioeconomic status, so the dataset may not be representative. Social communication difficulties was found to be the only risk factor that explained a significant and large amount of variance in psychopathology symptoms, and particularly externalising symptoms. Being ‘blind’ (ICD-10) associated with increased symptoms of psychopathology and increased externalising symptoms in particular, compared to having ‘low vision’. Adaptive functioning difficulties (as measured by the ABAS-III) was associated with increased symptoms of psychopathology.

* + 1. Strengths and Limitations

A strength of this study was the use of a stringent and clearly defined inclusion and exclusion criteria, to increase homogeneity of the sample and also potential replicability of the study. Restricting the sample to ‘potentially simple’ CDPVS, enabled a specific focus on children with a ‘primary’ VI and no other known comorbid impairments, such as ID, which could also influence psychopathology. Using both parent report and child self-report enabled a richer picture of the psychopathology profiles in the sample and enabled investigation of inter-rater reliability.

Recruitment from two paediatric ophthalmological departments and the use of detailed ophthalmic information from current medical records were also a strength. Further, the design permitted analysis of the main literature driven and selected risk factors as ‘predictive’ variables, which builds upon the correlation technique employed in previous studies though still did not permit consideration of causality. Although the sample was small and the ‘blind’ group was particularly small, the analyses were shown retrospectively to be mainly appropriately powered to detect large effects.

A limitation of the study is that a control group was not incorporated. Population normative data was used to compare mean scores on questionnaires within the VI sample with mean scores on questionnaires in the general population. There are several limitations with this approach. First, the age range of the normative data was typically broader than the age range of the VI sample. As age is associated with differences in psychopathology symptoms in the general population, this may have impacted the validity of the comparison between the mean scores. Second, the normative data was not always collected in the UK which may also impact validity. Third, a lack of control group meant that participants could not be matched in relation to non-vision factors, such as socioeconomic data and ethnicity. As there was a positive skew in socioeconomic status in the VI sample, it may have been particularly important to control this variable to investigate mean differences. The decision not to include a control group was made for feasibility reasons and is a limitation of the study.

Another limitation of the study is the risk of Type 1 (falsely positive) errors, given the use of multiple comparisons, though Bonferroni corrections were applied to reduce this risk. Another limitation is that the questionnaire measures used in the study were not validated for use with children with VI, though they appeared to be appropriate and reliable. Most items were not vision dependent, internal reliability of scales was generally acceptable, and levels of agreement between parent and child raters was moderate (in line with findings in the TD population). Measurement of psychopathology symptoms did not include any independent clinician judgement, which may have impacted on reliability. Further, the self-report measures of children who were ‘blind’ had to fill in their questionnaires orally with parents, in comparison to children who had ‘low vision’ who could independently fill in questionnaires with enlarged font, and this may have affected responding due to social desirability effects. A more valid approach may have been for the researcher to administer the self-report questionnaires orally via skype to all children, in order to control questionnaire conditions. ID was excluded based on parent report and medical records rather than cognitive assessments, due to feasibility restrictions. The limitations of the study may influence generalisability to the wider VI population. Other limitations in the sample representativeness may also influence generalisability, such as positive skewed socioeconomic status, over representation of retinal conditions and a low response rate.

* + 1. Clinical Implications

First, the evidence shows that children with VI who are of relatively normal intelligence may be at ‘high risk’ of psychopathology, compared to TD children. This may indicate the need for a preventative approach to meet the mental health needs of children with VI. Parents and professionals may benefit from increased psychoeducation about the psychological needs of children with VI and the best ways to promote positive mental health. Children with VI should be regularly and routinely screened for psychopathology symptoms. Where there are concerns, the child should have timely access to appropriate intervention (RSPCH, 2018). More priority may be needed at policy level to meet the needs of children who have long term paediatric disorders, including VI, whose needs have not been outlined in the recent Green Paper for Mental Health (Department for Education, 2017).

Second, children who are ‘blind’ may be particularly vulnerable to psychopathology and it may be important for symptoms to be monitored from an early age. Third, the high reported incidence of separation anxiety in the sample highlights possible attachment vulnerability in the group. Children with VI who have raised anxiety about navigating the physical environment may be particularly at risk of separation anxiety. It is important to foster ‘independence’ in a way that is psychologically manageable for the child and, notably, the majority of parents in this sample reported a lack of input from specialist VI professionals in the home environment. Habilitation training may be important to promote independence skills in children with VI, which may lead to decreased anxiety in the domestic and external environment. It will be important to consider how best to support the development of secure attachment styles from the early years and through middle childhood.

Fourth, social communication was the only variable that explained a significant amount of the variance in psychopathology symptoms (as measured by the SDQ) in the multivariate analysis. Children who had social communication difficulties were particularly ‘at risk’ of externalising symptoms. Conversely, behavioural difficulties in children with VI may be indicative of ASD risk. As children with VI are at ‘high risk’ of ASD, social communication and behaviour may therefore be important targets for screening and intervention. It may be that this combined presentation is a specific form of social communication disorder within the VI population, which may require tailored diagnostic criteria and interventions.

Fifth, the questionnaire battery, including child report, was valuable in picking up the symptom profiles of children with VI and could be useful for use in the clinic or educational environment. Integrated clinical formulation may be needed to reach an accurate assessment of the individual child’s psychological status and possible psychopathology. This may include a comprehensive developmental history, clinician-scored behavioural measures and teacher informants. This may be particularly important in the context of children with VI, who may show reduced nonverbal communication (e.g. lack of range of facial expressions) which may present as flat affect (Dale & Edwards, 2015).

Overall, children with VI may present with a range of difficulties which together with their primary disability of VI have a substantial collective impact on functioning. Of concern from anecdotal clinical report, their presentations may not meet the severity threshold and criteria for local CAMHS input. Standardised measures of psychopathology may be less effective in picking up psychopathology in a VI population than in the TD population. A National Blind CAMHS, similar to the national Deaf CAMHS, may therefore be able to better serve the needs of this population, with formulations and interventions designed and delivered by clinicians who have expertise in the unique developmental and psychological experience of children who have VI.

* + 1. Future research

Future research should aim to replicate this study with representative samples of children who have CVI, ‘potentially complicated’ CDPVS, and comorbid ID, across the age range, to establish the needs of the total VI population and to inform service delivery. It is anticipated that the profile and pattern of psychopathology will be affected by differing origins of VI, brain involvement and developmental and chronological age. Longitudinal studies are needed to understand the developmental trajectories of possible risk and protective factors to psychopathology, and variables which may moderate and mediate this relationship. This may help to inform theoretical models of psychopathology in children with VI, to be tested in future research and of future clinical relevance. Future research could also consider the development of a specific measure of psychopathology symptoms for children with VI, which captures psychological distress specific to the vision loss and issues of stigma and discrimination which may be pertinent in this group.

The current study focussed on within-child risk factors for psychopathology and future studies are needed to evaluate the impact of parenting factors. Mothers of young children with VI are at ‘high risk’ of parenting stress (Sakkalou, Sakki, O’Reilly, Salt & Dale, 2017) and maternal mental health is associated with child psychopathology in the general population (Batenburg-Eddes et al., 2013). Further research is required to explore whether secure attachments can be supported by promoting ‘sensitive parenting’ using video feedback techniques, as in the sighted population (Bakermans-Kranenburg, 2003). Investigation is required to understand what ‘sensitive parenting’ looks like for children with VI. A recent systematic review indicated that interaction, inter-subjectivity and joint attention needed to be addressed in adapting intervention programmes for children with VI (van den Broek et al., 2017).

Future research should examine tailoring preventative and intervention approaches of psychopathology, to the needs of children with VI. A growing body of research demonstrates the efficacy of interventions including cognitive behavioural therapy, acceptance and commitment therapy, and family therapy for children with a range of paediatric disorders (Fonagy et al., 2014; Sansom-Daly, Peate, Wakefield, Bryant & Cohn, 2012). Interventions may need to be adapted for use with children with VI and research is needed to determine the efficacy of different interventions with this group. A recent study found that an adapted CBT for anxiety intervention for children with VI was not effective in reducing internalising symptoms (Visagie, 2016). Other therapeutic approaches, such as narrative therapy, which focusses on instilling a sense of agency and self-acceptance and positive self-attributions to the child, may be suitable for use with children who have VI. Programmes to support the development of social communication and social skills in children with VI should also be trialled, particularly to see if this reduces the risk of externalising symptoms.

* + 1. Conclusion

Children with VI are at significant risk of psychopathology and may therefore require targeted support, which may include preventative interventions. Social communication difficulties (as measured by the SRS 2) explained over half of the variance in psychopathology (as measured by the SDQ) and was particularly associated with externalising symptoms. This may therefore be an appropriate target for intervention. Children with VI are at increased risk of ASD and early identification and diagnosis is important for interventionist reasons. Programmes to increase adaptive functioning skills may be protective against psychopathology. Children who are ‘blind’ may be particularly vulnerable and need to be monitored carefully. Given the limited availability of CAMHS in the UK (House of Commons Health Committee, 2014) it is important to consider how best to meet the mental health needs of the VI population and new forms of service delivery may be appropriate.

1. Integration, Impact and Dissemination
   1. Systematic Review: Methodological Reflections
      1. Strengths and rationale for decision making process.

A recent systematic review concluded that children and adolescents with VI were at increased risk of psychopathology compared to sighted peers, though findings between included studies were contradictory (Augestad, 2017). To extend current knowledge in this area, I set out to investigate which risk and protective factors were associated with psychopathology in children with VI, which had not yet been systematically investigated. Systematic reviews are designed to locate, appraise and synthesise the best available evidence relating to a specific research question and this methodology was therefore felt to be the appropriate (Dickson, Cherry & Boland, 2014).

I decided to focus on studies that included a measure of psychopathology (e.g. internalising or externalising symptoms), rather than broader constructs of mental health (e.g. psychological wellbeing or quality of life), to enable a more robust consideration of symptoms associated with long term difficulties. I also decided to exclude studies where the whole VI sample had additional needs (e.g. Intellectual Disability; ID, or hearing impairment). Whilst this may have led to reduced representativeness, it enabled a more targeted focus on the impact of VI without other potentially confounding factors. I piloted the search strategy and supplemented the results of the electronic search with hand searching, to ensure inclusivity. This led to an additional three included studies. A research assistant and I independently screened all papers using the inclusion criteria and agreement was found to be good.

A strength of the review was that all included studies were quality assessed. Quality analysis can be used to assist in study selection and/or data analysis and synthesis. I decided not to exclude studies based on the quality analysis and instead to use this to contextualise findings. This is because many studies lacked sufficient information about samples and excluding based on this would have led to an extremely small data set. Instead I aimed to use the quality assessment within the data analysis and synthesis to investigate whether quality differences provided an explanation for differences in findings.

I developed a quality assessment tool, based on items included in The Mixed Methods Appraisal Tool (Pace et al., 2012). Items relevant to the quality assessment of quantitative studies were incorporated into the quality assessment tool. A strength of the quality assessment was that additional items were added to ascertain whether variables important in the context of psychopathology and VI were considered in the research design. I decided not to undertake a meta-analysis as part of the data synthesis, as the assumption of homogeneity was not met (Dickson et al., 2014). The differences across study designs, sample characteristics, and diversity of findings, precluded a statistical synthesis of findings and so a narrative synthesis was undertaken.

* + 1. Limitations.

The representativeness of the review may have been impacted by the decision to exclude case studies and qualitative designs. I made this decision so that studies powered to run statistical analyses could be targeted. Related to the decision not to use quality analysis to inform study selection, a limitation of the review is that included studies used different inclusion criteria, so the sample (e.g. VI severity, comorbid impairment) varied substantially, which may have impacted on validity. This reflects the limitations of previous research and informed my decisions about sampling in the empirical study. Another possible limitation of the review is that the included age range was broad, though I felt that this was necessary given the scarcity of research in this specific area. Differences in age range of samples between studies did not seem to explain the mixed findings, though there were not enough studies of children (compared to adolescents with VI) to draw conclusions about this.

* 1. Empirical Study: Methodological Reflections
     1. Strengths and rationale for decision making process.

The findings of my systematic review highlighed that previous research investigating psychopathology and associated risk and protective factors in children with VI have produced mixed findings, which may be related to methodological differences between studies. Importantly, many previous studies have not excluded children who have known comorbid impairment (e.g. ID), and many have not sufficiently defined VI characteristics, which impacts the validity of findings. My study therefore aimed to overcome these limitations by using a sample of children who had ‘potentially simple’ Congenital Disorders of the Peripheral Visual System[[18]](#footnote-18) (CDPVS) with ‘low vision’[[19]](#footnote-19) or ‘blindness’[[20]](#footnote-20) (ICD-10). Children who had known comorbidities (e.g. ID) were excluded. This reduced possible neurological confounds and enabled a more valid and specific investigation of the impact of VI on psychological development (Sonksen & Dale, 2002), though leads to reduced generalisability of findings as ID is so frequently comorbid with VI.

I decided that the age range should be 8-11 years, given the limited research in this area identified in the systematic review and knowledge that middle childhood represents a critical period for establishing strong psychosocial foundations which may protect against future psychopathology (Laurens et al., 2017). During this period, children are increasingly exposed to influences beyond the home and may encounter new challenges, particularly at school (Poulton, Moffitt & Silva, 2015). This may be a particularly challenging time for children who have a disability, such as VI which is likely to impact school functioning. Although this age range was narrow compared to many previous studies of children with VI, it was possible that age effects impacted findings. Age was controlled for within the analyses, but it was found to have a minimal effect. Recruitment from two paediatric ophthalmological departments and the use of detailed ophthalmic information from current medical records were also a strength. This method led to improved reliability in considering the relative impact of VI severity, as I was able to accurately group participants into VI severity bands.

The measures that I used were found to be reliable and valid in previous research. All measures appeared to be reliable within the study sample, with sufficient Cronbach Alpha values, indicating good internal consistency. None of the measures had been validated for use with children who have VI, though most did not include vision-dependent items and when they did, the items or scoring were adapted appropriately, and the measures were therefore felt to be valid for use with this sample. The use of both parent report and child self-report measures enabled a richer picture of the psychopathology profiles in the sample and enabled investigation of inter-rater reliability, which has not been investigated previously in children with VI of this age. I used parent report only in analysis of risk factors, though it may have been more reliable to use statistical methods to aggregate data from parent and child self-report (Bird, Gould & Staghezza, 1992). I conducted appropriate preliminary and statistical analyses and although the sample was small, the analyses were mostly appropriately powered to detect large effects and Bonferroni corrections were applied to reduce Type 1 (false positive) errors.

* + - 1. Service user involvement.

There was a workshop, developed using co-production methods, in June 2017 at Great Ormond Street Hospital called: “What more can we do to support children and young people who have a vision impairment?” This was attended by parents, young people with VI, and professionals and was a forum for discussion about the needs of children with VI. I liaised with parents and young people with VI during the workshop. I also liaised with a parent of a child with VI during the design and recruitment stage. Service user involvement informed the study rationale and design. Parents highlighted that mental health was an area of high concern and they asked for further research about what types of mental health difficulties children with VI are at risk of and why. Young people felt that it was important to include child self-report measures, given the unique developmental experience of children with VI and so this was included in the design. Parents and young people were concerned about accessibility for child reporting and asked for this to be considered. Unfortunately, there was not enough funding to provide braille self-report measures, but we did ensure that questionnaires could be given in enlarged font and electronically where required. A further plan for service user involvement within the project is to involve parents who participated in plans for dissemination and follow up action by the clinician co-investigators.

* + 1. Limitations and difficulties encountered in the research process.

Recruitment difficulties impacted on the final sample size. A priori power analyses indicated that a sample size of *n*=50 would be sufficient for the planned multiple regression, which was discussed with consultant clinicians at participant identification sites and agreed to be feasible within the data collection period. It became apparent that clinicians had overestimated the number of eligible children attending routine clinical appointments. Some clinicians suggested the inclusion of participants who had comorbid neurological impairment, to reach the target sample size. I decided not to pursue this, as this would have introduced a significant confound to my research which aimed to investigate psychopathology profiles in children with VI who did not have comorbid impairment. It was also suggested that the age range be extended upwards into adolescence. I decided against this as children and adolescents are likely to present differently in relation to psychopathology. For example, longitudinal studies of TD children and adolescents have found that externalising disorders generally decrease with age (Hicks et al., 2007), whereas internalising disorders may increase with age (Sagatum, Lien, Sogaard, Bjertness & Heyerdahl, 2008). The impact of these decisions was a more valid but smaller sample. To maximize sample size, I extended the recruitment period by two months.

Another challenge to recruitment was that it was difficult for clinicians, whom often had busy schedules, to identify and recruit participants for the project. I therefore decided that I would attend clinics to remind clinicians to identify participants and to introduce the project to eligible families. This led to the identification of 128 eligible participants. Despite extending the recruitment period and providing recruitment support, I was not able to reach target numbers. The current sample size is 34, which is smaller than the majority of studies in the field but possibly more valid due to the sampling strategy. Due to very large effect sizes, the multiple regression was still sufficiently powered. Bonferroni corrections were applied to reduce the risk of Type 1 errors.

Ascertainment rate was 43.8% (*n* = 53) and of this, the participation rate was 64.2% (*n* = 53). The overall response rate was 27.2%, which is low (Hamilton, 2003). Face-to-face recruitment may have yielded higher recruitment rates, but this was not always feasible and was dependent upon whether potential participants had a routine clinical appointment during the data collection period. Face-to-face participation may have also yielded higher participation rates. Clinicians did not feel that they had time to liaise with families prior to appointments to arrange this and it was not appropriate to ask eligible ascertained participants to participate on their clinic day, as it was important that they did not feel coerced to participate and that they had time to think about whether they would like to participate. All questionnaire packs were therefore completed at home. It is therefore likely that there was a sampling bias which affected the representativeness and generalisability of the study.

Another possible limitation is that all ‘blind’ children completed self-report questionnaires with their parents, whilst most of the partially sighted children completed the questionnaires independently, which may have impacted scores due to social desirability impacts. It was considered whether the questionnaires should be administered orally by the researcher to all children, but this was not feasible. A more valid approach may have been for the researcher to administer the self-report questionnaires orally via skype to all children, in order to control questionnaire conditions. ID was excluded based on parent report and medical records rather than cognitive assessments, due to feasibility restrictions. It is possible that some children had unknown milder cognitive impairments or specific learning difficulties which may have impacted on psychopathology. Measurement of psychopathology symptoms relied on parent and report only, without independent clinician or teacher judgements. These factors may have led to reduced reliability. Whilst the multivariate design enabled a comparative investigation of risk factors, it did not permit me to consider causality or to investigate the impact of moderating or mediating variables on risk factors, which may be a key area of future investigation.

The focus of the empirical study was primarily on within-child variables. I had originally planned to use the SCORE-15 (Index of Family Functioning and Change) to measure both risk and protective factors, but I decided to remove this questionnaire. There were concerns about the effects of multiple comparisons in a small data set, so potentially important parental and family factors were not investigated. The issue of participant burden of questionnaires was also raised by the NHS Ethics committee, which led to modification of the original design. Further, the study included a relatively broad range of VI. As children who were ‘blind’ appeared to have more difficulties than the rest of the sample, this decision to include a broad range may have led to diluted findings and reduced risk level of psychopathology severity. Conversely, evidence of psychopathology symptoms across the vision range highlighted that ‘low vision’ is also a risk factor for psychopathology. Finally, a considerable number of children scored in the clinical range for difficulties in the study and this was of clinical significance. This was anticipated, and appropriate steps were incorporated in the project design: all parents received a feedback report (Appendix S), and where clinically significant difficulties were indicated, parents were advised to contact their GP to access appropriate support.

* 1. Summary and Integration

Synergy was achieved between the systematic review and the empirical article. The systematic review identified the methodological limitations of previous research and some of these were addressed in the empirical study. The systematic review concluded that the only consistently identified risk factor for psychopathology in children who have VI, was poorer cognitive abilities. This informed my decision to exclude children with a comorbid ID from the empirical article, to investigate additional risk factors. The systematic review outlined a lack of valid and clearly defined samples in previous research, which may have explained contradictory findings between studies. I therefore decided to use a strict and valid ‘primary VI’ inclusion criteria in the empirical article. The systematic review identified that potentially important risk factors (e.g. lower socioeconomic status, autism traits, poor adaptive functioning) had not been sufficiently examined in relation to psychopathology and so these were investigated in the empirical study. The systematic review also indicated a lack of multivariate research, enabling examination of the relative importance of risk factors, and so the design of the empirical study addressed this.

In the empirical study, children in the sample were found to be at increased risk of psychopathology, and particularly separation anxiety, compared to sighted peers. This highlighted that children with VI without poorer cognitive abilities and ID were still significantly at risk of psychopathology. More severe VI (‘blindness’) and were associated with more symptoms of psychopathology. Gender, socioeconomic status, and age were not associated with psychopathology symptoms. Social communication was the only risk factor that significantly explained variance in psychopathology symptoms in the multivariate analysis, and particularly explained variance in externalising symptoms. The systematic review therefore informed the empirical article design which led to new findings.

* 1. Implications

The main clinical implication of the systematic review is that children with VI and comorbid ID may benefit from increased psychopathology symptom screening and support, given that ID was identified as a risk factor for psychopathology. In the absence of additional identified risk factors, despite findings that children with VI are at ‘high risk’ of psychopathology, ‘VI’ could therefore be considered as a predominant risk factor and all children with VI may therefore benefit from symptom screening.

The main clinical implication of the empirical article was that children in the sample, who were in middle childhood and were of relatively normal intelligence, were at ‘high risk’ of psychopathology compared to the TD population. ID is therefore not the only risk factor for psychopathology in children with VI and a preventative approach with more priority at policy level may be required (RSPCH, 2018). The questionnaire battery used in the empirical study could be used for screening purposes. This may be particularly important for children who are ‘blind’, who were found to be the most at risk of psychopathology symptoms, in the sample. Another clinical implication was that children in the sample were at particularly ‘high risk’ of separation anxiety, which could suggest a possible attachment vulnerability in children with VI and therefore a need for support in this area. Importantly, social communication difficulties were found to be a risk factor for psychopathology in the sample. There was also some evidence that adaptive functioning difficulties associated with increased symptoms of psychopathology. These may therefore be important areas for symptom monitoring, prevention and intervention.

* 1. Future Research

Future research should aim to replicate the empirical study with representative samples of children with VI, across the age range, to establish the needs of this clinical population and to inform service delivery. This may enable a future meta-analysis, which would lead to a more robust analysis of risk and protective factors for psychopathology in children with VI. Longitudinal multivariate studies are needed to understand the developmental trajectories of possible risk factors to psychopathology, and variables which may moderate and mediate this relationship. This should include protective factors such as parenting and family factors, which is currently an under-researched area in the context of VI.

Future research is also needed to examine how to tailor preventative and intervention approaches to psychopathology to the needs of children with VI. Findings from the empirical study suggest that interventions may need to target the parent-child relationship, as well as anxiety symptoms. Future research could also consider whether programmes to support the development of independence and social communication skills in children with VI leads to the reduction of psychological distress in children with VI.

* 1. Impact

The main impact is to assist the children with VI and their families who participated in the study by increasing the recognition of their mental health needs. This may lead to greater recognition of these needs, within the health and education services. It will be recommended that the national specialist recruitment sites involved in the study introduce routine screening for psychopathology symptoms and social communication difficulties, as this was found to be highly predictive of psychopathology in the sample. The hope is that wider dissemination will lead to increased use of screening tools for children who have VI in other specialist and local services. The recruitment sites may also use these findings to consider whether a provision for intervention can be offered within the service.

There can be a tendency to attribute symptoms of psychopathology to the primary disorder in children with paediatric disorders. This may lead to missed opportunities for early intervention. This has historically been the approach for explaining symptoms of autism in children with VI which are sometimes referred to as ‘blindisms’. The findings that not all children who have VI present apparently with symptoms of psychopathology (or autism traits), as measured by the questionnaires, suggest that symptoms are not just an inherent part of VI and so may therefore be both preventable and amenable to intervention. This concept may be important to consider in the context of other paediatric disorders. A cumulative risk approach may help to explain the overrepresentation of psychopathology in children who have paediatric disorders: the paediatric disorder may make the child more vulnerable to additional difficulties other than the primary disorder.

* + 1. Potential Beneficiaries

The scope of the impact is that professionals supporting children with VI will be informed by the research, which may be used to inform practice inputs. The findings provide evidence for the need for routine screening and greater attention in health, education, and social sectors, to the mental health needs of children with VI. The intention is that teachers, Special Educational Needs Coordinators and Qualified Teachers for The Visually Impaired will be beneficiaries of the research, as findings may help increase awareness of vulnerabilities of any child with VI. Health professionals may also be beneficiaries of the research, with findings suggesting that interventions for internalising symptoms, and particularly for separation anxiety may be effective in improving outcomes in this group. Speech and Language Therapists and Occupation Therapists/Habilitation Officers may also have a key role to take in supporting the development of adaptive and social communicative skills.

The practical yield of the research for families and practitioners could be maximised by using a consultation model. That is, health care professionals who have expertise in working with children who have VI could be involved in training local health, education, and social care professionals and parents in understanding and supporting the psychological needs of children with VI. This may include elements of psychoeducation about thoughts, feelings, and behaviours and basic strategies to manage anxieties and to improve mood. A self-help document could also be produced to be used by children with VI. The recommendation that professionals use screening tools for psychopathology with all children who have VI is feasible as the measures that have been used in this study are free to access. The benefits could be evidenced by clinical audits of the use of screening questionnaires in routine clinical appointments. There could also be an audit of parental or local professional understanding of child mental health needs before and after training. Clinicians could be asked to provide feedback about what they intend to do with the findings, including any policy or pathway changes.

* + 1. Originality

This research is important due to its originality. My systematic review indicated an array of methodological limitations with previous research. This may account for mixed findings between studies about whether children with VI are at increased risk for psychopathology than their sighted peers and what the associated risk and protective factors are. This was the first study to systematically investigate psychopathology and related risk and protective factors in children who have ‘low vision’ or ‘blindness’ (ICD-10) caused by ‘potentially simple’ CDPVS with no known comorbid ID. This enabled a more valid investigation into the specific impact of VI on psychological development. This study is also one of few studies to use a multivariate design, to assess the relative importance of possible risk and protective factors of psychopathology in this group. It is also the first study to investigate social communication and adaptive functioning as ‘predictive’ factors in this context.

* 1. Dissemination

The systematic review and the empirical article are being prepared for publication. The intent is for their submission to the Developmental Medicine and Child Neurology journal. This is considered to be an appropriate journal for submission due to the focus on paediatric neurology and neurodisability. It is one of the world’s leading journals in paediatrics and aims to disseminate information worldwide to improve the lives of children who have disabilities and that of their families. The impact factor of the journal is high (3.116), with a strong ranking in both paediatrics and clinical neurology. The Journal of Consulting and Clinical Psychology could also be an appropriate journal to consider for publication.

I will present my research at the Mary Kitzinger conference in July 2018 (paper accepted). This is a two-day international conference hosted by Great Ormond Street Hospital for Children, in collaboration with the Mary Kitzinger Trust and Royal Society for Blind Children. The title of the conference is: ‘Childhood visual impairment and mental health: Science into practice’ and will be attended by both professionals and service users with the aim of improving understanding within the field. I will be presenting an oral and poster presentation. I will also explore opportunities to present my research at other vision specific conferences. My primary recommendation will be that all children who have VI are screened for mental health difficulties within educational and health services.

I have also submitted an abstract to present my research at the Faculty for Children, Young People and their Families Annual Conference, Liverpool, in September 2018. The theme is: ‘The year of the child: Clinical psychology’s role in building futures’. Dissemination in VI specific conferences is likely to have a more direct impact, but it may also be important that findings are discussed in more general forums to raise awareness of the needs of children who have VI and possibly children who have paediatric disorders in general.

Findings will also be disseminated to clinicians who work with children who have VI in the National Specialist Services who were involved with the project, during a CPD session or team meeting. Findings will also be published on the affiliated research website. It is therefore hoped that dissemination here will have a wide reach. A written summary of findings will be produced to highlight implications and recommendations. Findings will also be disseminated to families who took part in the project. Parents will receive a summary of the overall project findings, in an end of project report, which will be adapted from the style of an academic paper and presented in an understandable form. Children who took part in the project will also receive a summary of the project findings, which will be presented in an age appropriate and accessible way.

Findings may also be disseminated to other families of children who have VI, via an online blog. These findings could be published on the South East Research Network for Schools (SERNS) website to reach school practitioners and teachers. Feedback about the project findings will be obtained at a later stage.

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1. Appendices

Appendix A

Systematic Review Research Protocol

**Review Question**

What are the risk and protective factors for psychopathology in visually impaired school-aged children and adolescents?

*This is an observational (relational) review question: investigating a relationship between 2 or more variables)*

**Inclusion Criteria**

|  |  |  |  |
| --- | --- | --- | --- |
| **Who** | **What** | **How** | **Where** |
| Children aged 4-19 with a visual impairment. | A measure of psychopathology. | Questionnaire measures – either parent report, teacher report or self-report. | Anywhere. |

|  |  |
| --- | --- |
| **Review Question** | What are the risk and protective factors for psychopathology in visually impaired children and adolescents? |
| **Population** | Children and adolescents aged 4-19 with low vision or blindness (must be the primary group of interest, i.e. not cerebral palsy, dual sensory impairment).  Sample Size N=20 or above |
| **Intervention** | N/A |
| **Comparator** | N/A |
| **Outcomes** | Includes at least one measure of psychopathology (internalising symptoms e.g. depressive/anxious feeling or somatization or externalising symptoms e.g. aggression, opposition, delinquency, impulsivity\*)  AND at least one measured risk or protective factor which is analysed in relation to psychopathology.  \*NB measures of quality of life, self-esteem, body concept, autism are not considered measures of psychopathology). |
| **Setting** | Any (e.g. schools, residential schools, hospitals, outpatient, community settings). |
| **Study Design** | Quantitative |
| **Other** | Published in a peer reviewed journal  Must be an empirical study  Full text must be available in English |

**Define terms**

**Risk Factor:** A characteristic at the biological, psychological, family and community or cultural level that precedes and is associated with a higher likelihood of problem outcomes.

**Protective Factor:** A characteristic cat the biological, psychological, family or community (including peers and culture) level that is associated with a lower likelihood of problem outcomes of that reduces the negative impact of a risk factor on problem outcomes.

**Psychopathology:**

Child psychopathology is characterized by two main categories referred to as internalising symptoms and externalising symptoms (Patalay et al., 2015). Internalising symptoms consist of excessive control over one’s behaviours, emotions and thoughts. Externalising symptoms consist of a lack of control over these (Nunes, Faraco, Vieira & Rubin, 2013). In particular, internalising psychopathology impacts the child directly as their excessive control is expressed through negative emotions such as depression, social withdrawal or anxiety. Externalising psychopathology, however, has an immediate impact on others where the effects of low control can lead to aggression, impulsive or antisocial behaviours (Nunes et al., 2013; Aunola & Nurmi, 2005).

**Visually Impaired**

The children in the sample are externally identified or self-identified as having a visual impairment. To the best of the reviewer’s knowledge, the visual impairment should be the specific point of interest in the study. For example, the sample should not be deaf-blind or be focussed on a specific medical condition that is not vision specific such as cerebral palsy.

**Children and Adolescents**

Age 4-19

**Search Terms**

vision disorder OR blind OR blindness OR visual impairment OR children with visual impairment OR vision disorders OR low vision OR partially sighted OR eye disorders OR vision impairment OR visually-handicapped

AND

Psychopathology OR Emotional and behavioural problems OR Optimism OR Worry OR Psychological well-being OR wellbeing OR well-being OR Psychological adaptation OR Emotional Adjustment OR Problem behaviors OR Behavior Problems OR Behaviour Problems OR Behaviour OR Behavior OR Emotional factors OR Executive Function OR Mental health problems OR Psychosocial OR Psychosocial development OR Conduct problems OR Emotional problems OR Prosocial behaviour OR Self-concept OR Self-esteem OR Psychological health OR Adjustment OR Adaptive behaviour OR Hopelessness OR Aggression OR Bullying OR Victimization OR Child Psychology OR Anxiety OR anxious OR Depression OR Mood OR Internalize OR Externalize OR Internalizing OR Externalizing OR Fear OR Major Depression OR mental health OR Difficult OR Behavioral OR Depressive OR Identity OR Problems OR Body Image

AND

Change settings – journals, English, school-age and adolescents

Appendix B

Revised Children’s Anxiety and Depression Scale (RCADS-P)

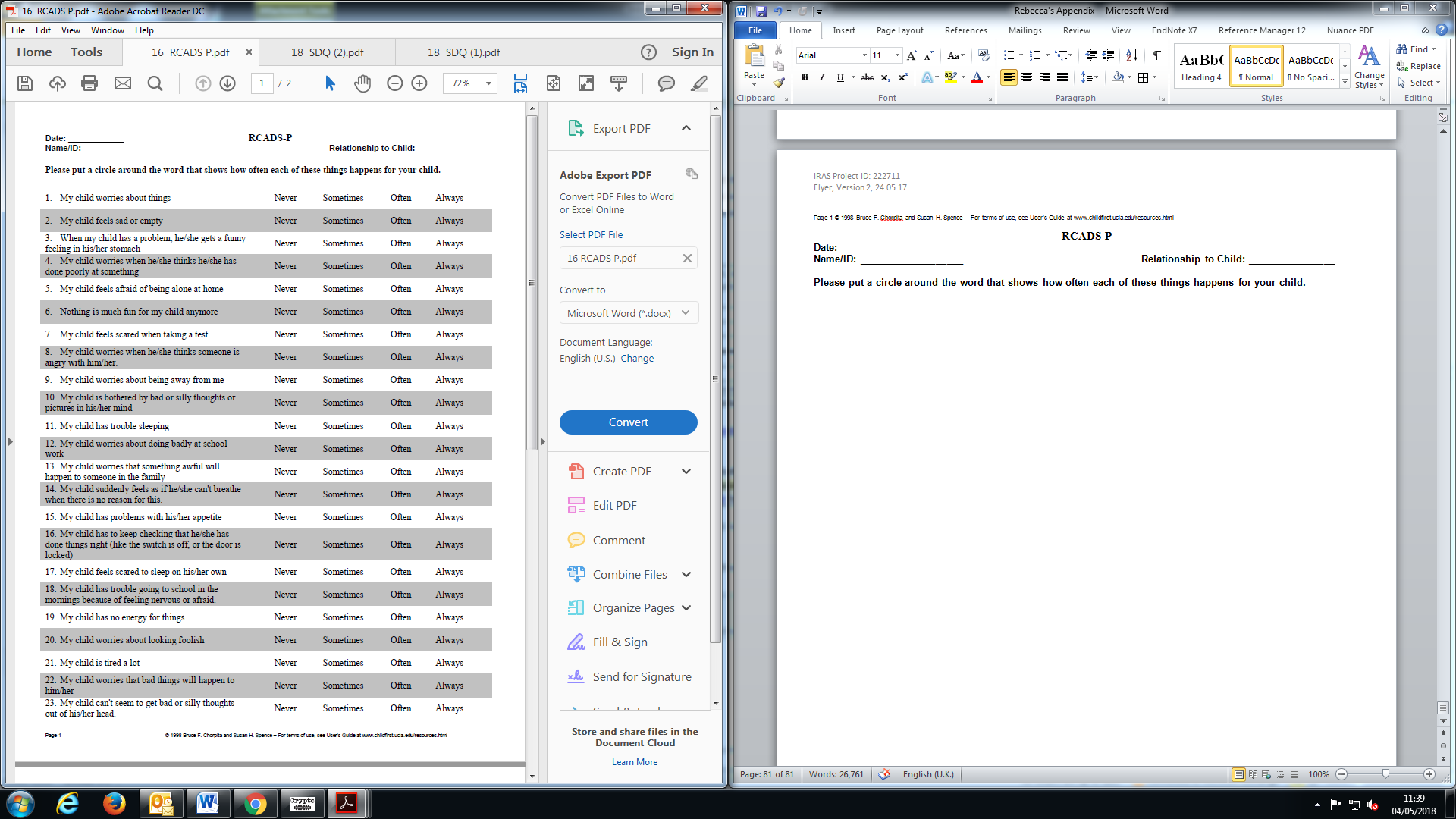
Page 1 © 1998 Bruce F. Chorpita and Susan H. Spence – For terms of use, see User’s Guide at www.childfirst.ucla.edu/resources.html

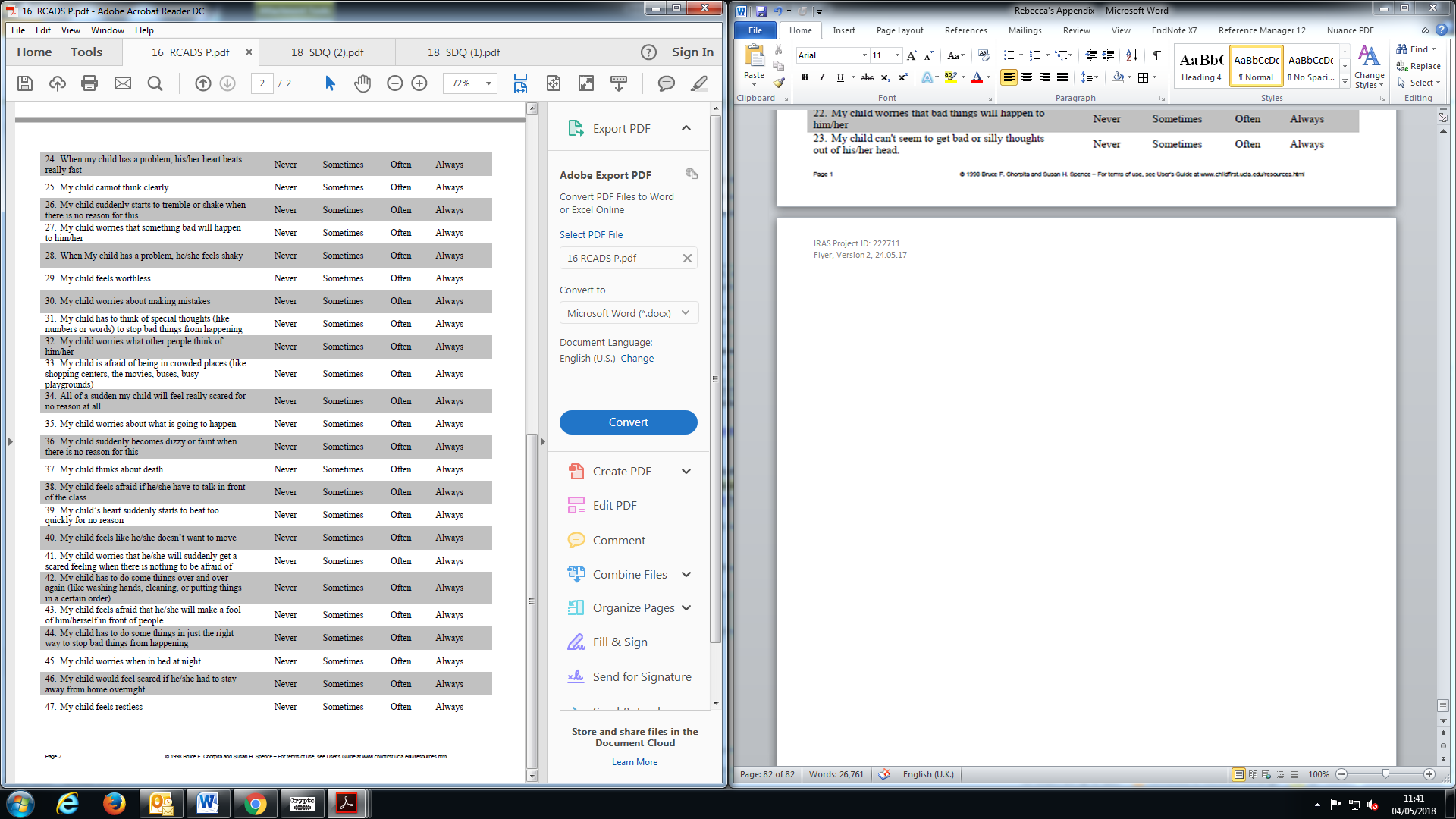
**RCADS-P**

**Date: \_\_\_\_\_\_\_\_\_\_\_\_**

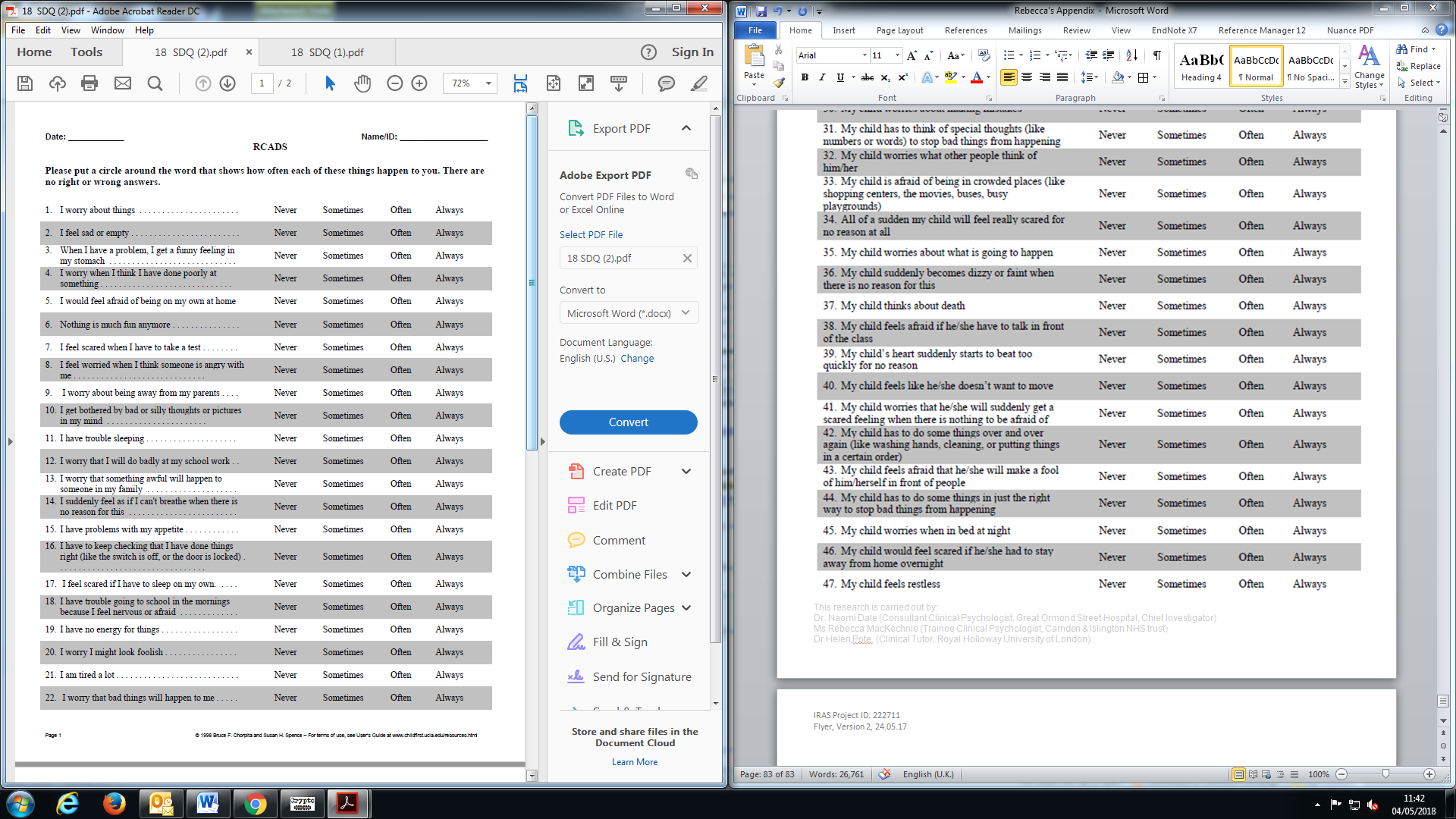
**Name/ID: \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_ Relationship to Child: \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_**

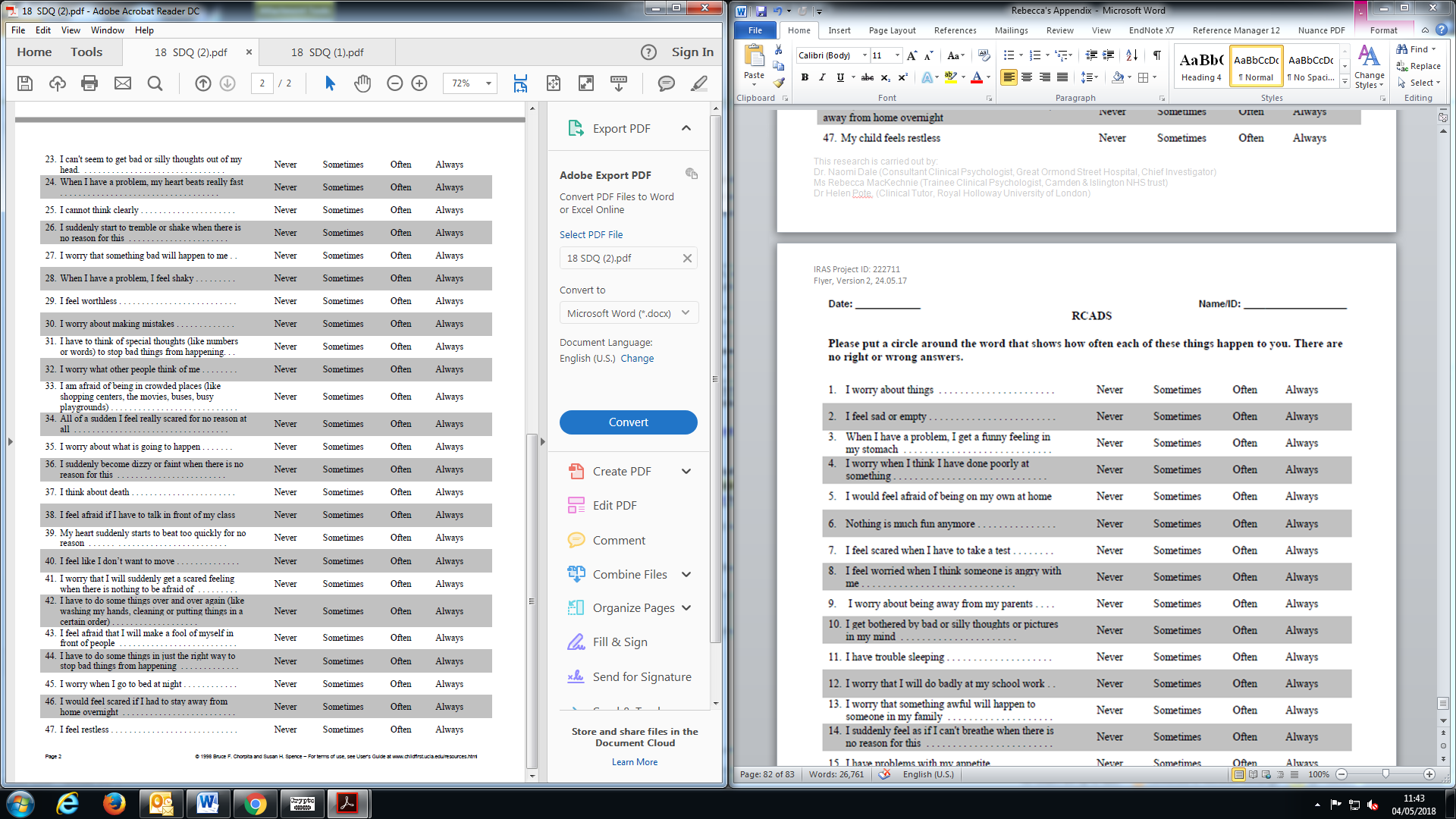
**Please put a circle around the word that shows how often each of these things happens for your child.**





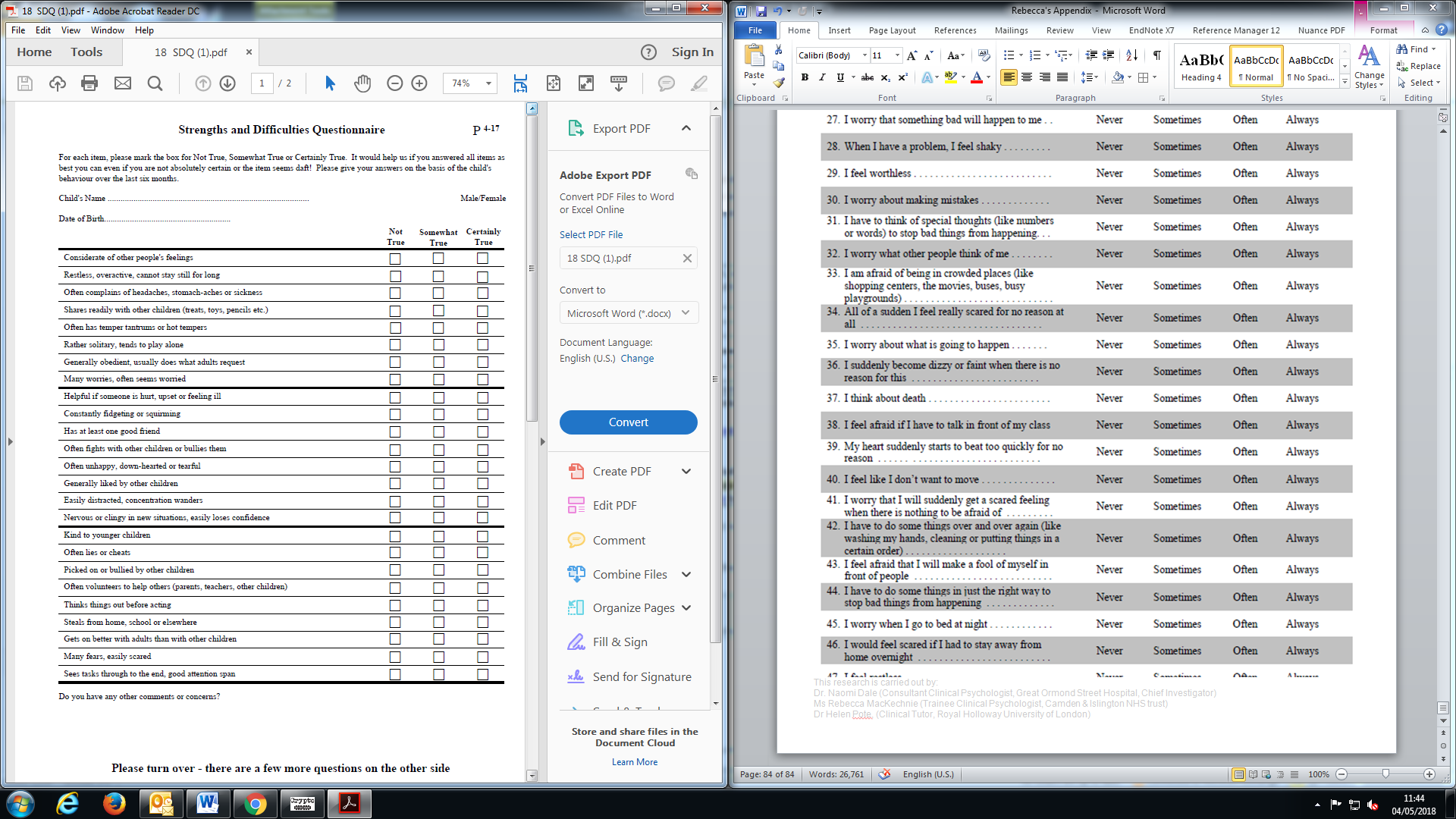
Appendix C

Revised Children’s Anxiety and Depression Scale (RCADS)



Appendix D

Strengths and Difficulties Questionnaire (SDQ)



Appendix E

Pediatric Quality of Life Inventory

Pediatric Quality of Life Inventory Parent Report (PedsQL)

Varni, J. W., Seid, M., & Rode, C. A. (1999). The PedsQL™: measurement model for the pediatric quality of life inventory. *Medical care*, *37*(2), 126-139.

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Appendix F

Pediatric Quality of Life Inventory

Pediatric Quality of Life Inventory Child Report (PedsQL)

Varni, J. W., Seid, M., & Rode, C. A. (1999). The PedsQL™: measurement model for the pediatric quality of life inventory. *Medical care*, *37*(2), 126-139.

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Appendix G

Adaptive Behavior Assessment System–Third edition

Adaptive Behavior Assessment System–Third edition (ABAS-3)

du Preez, J. (2017). Adaptive Behavior Assessment System–Third edition (ABAS-3).

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Appendix H

Social Repsonsiveness Scale

Social Repsonsiveness Scale (SRS)

Constantino, J. N., & Gruber, C. P. (2012). *Social responsiveness scale (SRS)*. Torrance, CA: Western Psychological Services.

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Appendix I

Demographic Form

Demographic Questionnaire

**General information**

|  |  |
| --- | --- |
| **1.** Child’s gender: | **F / M** |
| **2.** Child’s date of birth: | **\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_** |
| **3.** Weeks gestation at birth: | **\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_** |
| **4.** Child’s birth weight: | **\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_** |
| **5.** How many siblings does your child have? | **\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_** |
| **6.** Please give the ages of any other children in your family: | **\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_** |

**School/Childcare**

What kind of school does your child attend?

|  |  |
| --- | --- |
|  | Mainstream school without additional support |
|  | Mainstream school with additional support\* |
|  | Specialist school for children with visual impairment\* |
|  | Any other\* **\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_** |
|  | \*please specify any additional information: **\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_** |

Does your child receive support in the classroom?

|  |  |
| --- | --- |
|  | 1 to 1 TA/ LSA |
|  | Shared TA |
|  | No individual support |
|  | Any other\* **\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_** |

How many hours of support per week does your child receive in school? \_\_\_\_\_\_\_\_\_\_\_

Is your child supported by a VI specialist in school? **Yes/No**

Please specify \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_

Is your child visited by a VI specialist at home? **Yes/No**

Please specify \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_

To be filled out with information on child’s **mother**:

**Maternal Education (Please tick the highest level you have obtained):**

|  |  |
| --- | --- |
|  | Primary |
|  | Secondary (before A levels) |
|  | A Levels |
|  | Some university |
|  | University graduate |
|  | Some postgraduate |
|  | Master’s degree |
|  | PHD or professional doctorate/MD |

**Are you currently employed?**

**What does the organisation you work for mainly make or do?**

Particularly interested in key words (e.g. ‘manufacturing’/’distributing’), main goods involved, materials used.

**What is your main job?**

Job title:

**What did you mainly do in your job?**

Please specify your broad duties, and any qualifications/training required for your job.

**Are you working as an employee or are you self-employed?**

Employee – **Do you have any formal responsibility for supervising the work of other employees?**

Employee – **How many people worked for your employer at the place where you worked? –** Asking about the total number at the workplace – not just department. Referring to local unit (geographical location).

|  |  |
| --- | --- |
|  | 1-24 |
|  | 25-499 |
|  | 500+ |

Self-employed **- Do you work on your own or do you have employees?**

* **How many people do you employ at the place where you work?**

|  |  |
| --- | --- |
|  | 1-24 |
|  | 25-499 |
|  | 500+ |

Mother’s ethnicity:

White or White British

Asian or Asian British

Black / African / Caribbean / Black British

Mixed / multiple ethnic groups

Other (specify)\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_

* Were you born in the UK? Y / N
* **If no**, how many years you have lived in the UK: \_\_\_\_\_\_\_\_\_\_\_\_\_
* **If no**, country of origin: \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_
* Is English your first language? Y / N
* **If no**, how many years you have spoken English: \_\_\_\_\_\_\_\_\_\_
* Do you speak English at home? Y / N
* **If no**, what language is spoken at home: \_\_\_\_\_\_\_\_\_\_\_\_\_
* How many languages does your child hear at home? \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_
* Mother’s marital status:

Single/never married; Married; Separated; Divorced; Widowed; Living with partner

To be filled out with information on child’s **father**:

**Paternal Education: (Please tick the highest level you have obtained)**

|  |  |
| --- | --- |
|  | Primary |
|  | Secondary (before A levels) |
|  | A Levels |
|  | Some university |
|  | University graduate |
|  | Some postgraduate |
|  | Master’s degree |
|  | PHD or professional doctorate/MD |

**Are you currently employed?**

**What does the organisation you work for mainly make or do?**

Particularly interested in key words (e.g. ‘manufacturing’/’distributing’), main goods involved, materials used.

**What is your main job?**

Job title:

**What did you mainly do in your job?**

Please specify your broad duties, and any qualifications/training required for your job.

**Are you working as an employee or are you self-employed?**

Employee – **Do you have any formal responsibility for supervising the work of other employees?**

Employee – **How many people worked for your employer at the place where you worked? –** Asking about the total number at the workplace – not just department. Referring to local unit (geographical location).

|  |  |
| --- | --- |
|  | 1-24 |
|  | 25-499 |
|  | 500+ |

Self-employed **- Do you work on your own or do you have employees?**

* **How many people do you employ at the place where you work?**

|  |  |
| --- | --- |
|  | 1-24 |
|  | 25-499 |
|  | 500+ |

Father’s ethnicity:

White or White British

Asian or Asian British

Black / African / Caribbean / Black British

Mixed / multiple ethnic groups

Other (specify)\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_

* Were you born in the UK? Y / N
* **If no**, how many years you have lived in the UK: \_\_\_\_\_\_\_\_\_\_\_\_\_
* **If no**, country of origin: \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_
* Is English your first language? Y / N
* **If no**, how many years you have spoken English: \_\_\_\_\_\_\_\_\_\_
* Do you speak English at home? Y / N
* **If no**, what language is spoken at home: \_\_\_\_\_\_\_\_\_\_\_\_\_
* How many languages does your child hear at home? \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_
* Father’s marital status:

Single/never married; Married; Separated; Divorced; Widowed; Living with partner

Appendix J

Child Medical Form



**Project:** An investigation of mental health, social-emotional and behavioural profiles in children with congenital ophthalmological disorders and visual impairment.

*The parent has given consent for the following information to be disclosed to our research team.*

**Confidential Patient contact details:**

|  |  |
| --- | --- |
| **Child’s Surname:** |  |
| **Child’s First Name:** |  |
| **Hospital number:** |  |
| **Date of Birth:** |  |

**GP Contact details**

|  |  |
| --- | --- |
| **Professional Title:** |  |
| **Name:** |  |
| **Hospital Address:** |  |
|  |  |
| **Post code:** |  |
| **Telephone number:** |  |

**We kindly ask you to please fill in the following information:**

**Today’s Date: \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_**

**Child’s Gender: \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_**

**Tick**

|  |
| --- |
|  |

**Vision disorder:** Peripheral visual disorder (globe, retina, anterior optic nerve)

Describe:

**Tick**

|  |
| --- |
|  |
|  |
|  |
|  |

**Vision:** No vision

Light perception only

Aware of near objects only (non-light reflecting)

Vision for small objects or distant objects

|  |
| --- |
|  |

**Visual acuity (if known)**

|  |
| --- |
|  |

**Method of measuring acuity**

|  |
| --- |
|  |

**Paediatric condition**

**Tick**

|  |
| --- |
|  |

**No known hearingimpairment or motor impairment**

**Other Medical Information**

|  |  |  |
| --- | --- | --- |
|  | **No** |  |

1. **Does your child have a genetic condition? Yes If yes, please name gene/condition**

|  |
| --- |
|  |

|  |  |  |
| --- | --- | --- |
|  | **No** |  |

1. **Does your child have any pituitary problems? Yes If yes, please give details**

|  |
| --- |
|  |

|  |  |  |
| --- | --- | --- |
|  | **No** |  |

1. **Is your child taking any medication? Yes**

**If yes, please give details and reasons**

|  |
| --- |
|  |

|  |  |  |
| --- | --- | --- |
|  | **No** |  |

1. **Does your child have epilepsy? Yes**

**If yes, please give details and any medication**

|  |
| --- |
|  |

|  |  |  |
| --- | --- | --- |
|  | **No** |  |

1. **Was your child born at full term? Yes**

**If no, please provide details of your child’s gestational age at birth and about any birth complications**

|  |
| --- |
|  |

1. **What is your child’s education level in relation to their classmates. Please give details of their curriculum level (e.g. key stage 1/2/3, SATs results or information from school reports)?**

|  |
| --- |
|  |

|  |  |  |
| --- | --- | --- |
|  | **No** |  |

1. **Does your child have an intellectual (learning) disability?**

**Yes**

1. **If yes, please provide further details:**

|  |
| --- |
|  |

1. **Does your child have any social communication or language difficulties?**

|  |  |  |
| --- | --- | --- |
|  | **No** |  |

**Yes**

1. **Has this lead to any diagnosis e.g., language disorder, autism, learning difficulties? If yes, please describe**

|  |
| --- |
|  |

1. **Does your child have any sleeping difficulties?**

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
|  | **No** |  | **Mild** |  |

**Significant**

1. **Are they on any medication for sleep e.g., melatonin? Please describe**

|  |
| --- |
|  |

1. **Has your child ever accessed any support for mental health / emotional / social / behavioural difficulties?**

|  |  |  |
| --- | --- | --- |
|  | **No** |  |

**Yes**

1. **Which type of input has your child received? Please tick all that apply**

|  |  |
| --- | --- |
|  | School Counsellor |
|  | Other School Input |
|  | CAMHS Input (psychology, psychiatry, family therapy, play therapy etc) |
|  | Specialist psychologist input from an NHS clinic specialising in children with visual impairment |
|  | Other Psychology input |
|  | Counsellor or other input from charitable organisation |
|  | Psychiatry |
|  | Other |

1. **Please provide further details about which professional(s) (e.g. SENCO, QTVI, psychologist) helped your child and the types of services (e.g. NHS, school) you accessed:**

|  |
| --- |
|  |

1. **Child’s ethnicity:**

|  |  |
| --- | --- |
|  | White or White British |
|  | Asian or Asian British |
|  | Black / African / Caribbean / Black British |
|  | Mixed / multiple ethnic groups |
|  | Other ethnic group (please specify): \_**\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_** |

***Thank you!***

**PLEASE RETURN WITH THE PREPAID ENVELOPE**

Appendix K

Information form for parents



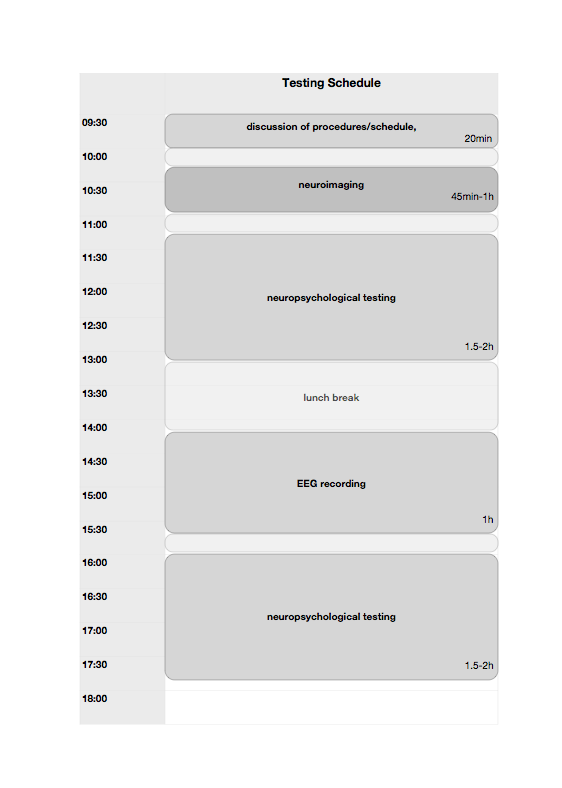
**An investigation of mental health, social-emotional and behavioural profiles in children with congenital ophthalmological disorders and visual impairment.**

**Chief Investigator: Dr Naomi Dale**

**Participant Information Sheet for Mothers of child**

We are a group of clinicians and researchers from the Developmental Vision Clinic and Ophthalmology Department at Great Ormond Street Hospital for Children. Our group specialises in the care of children with visual impairment. An important part of this work involves research about the challenges and difficulties faced by children with congenital visual impairment, as they grow up. This research enables us to ensure that we give the very best care and advice to children, families, practitioners, educators and doctors.

You are invited to take part in a research study on the wellbeing of children with congenital visual impairment. This study is carried out at Great Ormond Street Hospital for Children by Ms Rebecca MacKechnie, Dr. Naomi Dale and Dr Helen Pote.

Before you consider being involved, we want to make sure you know why we are doing this research and what we will be asking you to do. Please read the following information carefully and discuss it with other people if it helps. You can call or email us if anything is not clear or if you have more questions.

We are aware that some parents taking part in the study may also have a visual impairment. If preferred, we can arrange for information about the study to be sent to you electronically. Please let the research team know if you would like to arrange this.

**What is the purpose of this study?**

Children with long term health conditions are at high risk of mental health difficulties. They are more likely to have social, emotional and behavioural difficulties than other children. Some research has shown that children with congenital visual impairments are also at risk for a range of difficulties, but there has been very little research in this important area.

This study aims to investigate the strengths and difficulties of children with congenital visual impairment, in detail. The findings will highlight specific areas that children with congenital visual impairment might need extra help with.

This study is being conducted as part of an educational project for a clinical doctorate which Rebecca MacKechnie is undertaking.

**Why are we asking for your help?**

We would like to work with children aged 8 to 11 years who have a mild to moderate or severe to profound visual impairment that they have had from birth (or congenital) and their parents We are not including children who also have significant learning difficulties on this occasion.

**What will happen to us if we take part?**

In this study, we are asking for Mothers to complete questionnaires about their child. The project will involve Mothers completing several questionnaires, which are designed to ask about your child’s strengths and difficulties. Mothers will also be asked to complete a demographics questionnaire. We would also like your child to complete two questionnaires about their feelings. An information sheet for children has been included to help explain the purpose of the study. We would encourage you to discuss the study with your child, to find out if they are happy to participate. If your child does not want to take part, then we would still like Mothers to take part. We would also like to collect demographic information about the Father, with his consent. We would still like you to participate, even if the child’s Father does not wish to give his demographic information.

If you agree to take part, then your contact details will be passed onto the clinical researcher. The researcher will contact you at a convenient time to discuss any questions you have about the project. The researcher will obtain written consent from you to participate in the project. We will also require written consent from the Father, so that we can include his demographic information. After you consent to taking part in the project, the questionnaires will be sent to you in the post. Completing the questionnaires may take up to 90 minutes. The questionnaires will be sent to you in the post, with a prepaid return envelope. If preferred, a telephone call with the clinical researcher can be arranged so that the questionnaires can be administered verbally. If your child would like the child questionnaire to be administered verbally by the clinical researcher via the telephone, then this can also be arranged.

**What are the risks, benefits and burdens in participating?**

This work involves minimal foreseeable risk.

There are no direct benefits to you or your child from the study. You will receive a letter summarising your child’s strengths and difficulties, according to their questionnaire scores. This is not a clinical report and therefore we will not provide exact scores. We will offer a description of areas of relative strengths and weaknesses which may give you useful information about your child and can be shown to other professionals involved in your child’s care. We will also send you the results of the study upon completion if you wish to receive them. You will also be invited to attend a parent feedback meeting, which may be interesting and informative.

We hope that the study findings will help us gain a better understanding of the needs of children with congenital visual impairment and their parents.

Participating in the study will involve your time. The questionnaires may take up to 90 minutes to complete. We will ensure that you are given ample time to complete the questionnaires in your home environment. You will be provided with a prepaid envelope to return the questionnaires. We can also arrange to take your questionnaire responses over the phone if this is more convenient for you.

It is possible that some parents or children may find filling in the questionnaires stressful. If you become upset or distressed whilst completing the project, then please let the research team know so that they can offer you support. If you or your child becomes upset or distressed, you might also find it helpful to contact your GP so that you can discuss your concerns. If you or your child become very upset or distressed and require urgent support, then you may need to go to your local Accident and Emergency Hospital Department to access immediate support.

**Who will have access to my child’s medical/research records?**

All participants will be first approached about this study by a member of your child’s direct clinical care team (normally a doctor or a psychologist). The researchers will not have access to your personal details (e.g. phone number, address) until you express an interest to find out more about the study and give permission for the research team to contact you. Your personal details will then be saved on a secure server, which can only be accessed by the research team. These details will be destroyed as soon as the study is finished.

Only the researchers involved in this study and individuals from the Sponsor and Regulatory Authorities will have access to your child’s medical records and data collected during this study. The Sponsor and Regulatory Authorities will require access to such data and medical records to monitor and audit the conduct of the study.

We will tell people what we have learned in the study in reports and publications, but nobody will learn anything personal about your child, or any other child by reading these reports or publications. Your name or your child’s name will not appear in any reports or publications. We are following the government’s strict rules about how information like this must be stored, to keep it secure.

All of your data will be stored in an anonymised way – each participant will be assigned a code and this is how their data set will be labelled. This anonymous project data will be stored on the secure Great Ormond Street Hospital computer system (NHS) and also on an encrypted drive kept in secure storage office space in University College London Institute of Child Health (UCL-ICH). The anonymised data may be saved on the UCL-ICH network for short periods of time in order to analyse the data, but this will only take place with the encrypted drive attached to the computer. Anonymised data may be transferred electronically between the research team during data analysis of the project via encrypted USB stick/drive.

The key that links the code and the participant name together will be kept on a highly secure server that can only be accessed by the research team.

Your questionnaire forms will be labelled with an anonymised code and stored manually in secure office storage in secure office space in UCL-ICH. We may need to keep the research data for up to 15 years, but this will be kept anonymised.

With your consent, we would like to separately store your personal information (contact details) securely. This is so that we can contact you again in the future to see if you would like to take part in any related future studies.

**Who will know that we are taking part?**

When you consent to participate in the project, we will ask for your child’s GP details. We may, with your consent, ask your child’s GP to send the researchers information about your child’s vision. This could include: vision diagnosis, paediatric diagnosis and vision level and acuity (if relevant).

If during the project we identify any clinical or developmental concerns about your child, the Principal Investigator of the project will telephone you and discuss contacting the child’s GP/paediatrician. In this case, you may find it helpful to share the report that we have sent you to the relevant professional.

**Do we have to take part?**

No, it’s entirely up to you. Even if you sign the consent form to say you want to be involved, you can change your mind at any time without telling us why. If you decide not to take part in the study, your child’s clinical care will not be affected in any way.

**What if something goes wrong?**

It is unlikely that anything should go wrong in this study. If you feel that you have been harmed due to someone’s negligence, then you may be able to claim compensation. This will require you to prove fault in a court of law and you will need to bear the legal costs.

If you wish to complain about the conduct of this study, then the usual complaints process applies to you. Please discuss any issues with Dr Naomi Dale in the first instance. If you are still not satisfied and wish to complain formally please contact the Great Ormond Street Hospital Foundation Trust, PALS (Patient Advisory Liaison Service) on 020 7829 7862 or email [pals@gosh.nhs.uk](mailto:pals@gosh.nhs.uk).

**What will happen to the results of the study?**

We expect to get the results in 2018, and we will publish them in scientific and practitioner journals. The results will be shared at conferences and services for children with visual impairments. Nobody will learn anything personal about your child, or any other child, by reading these reports or publications or by attending any conferences. Your child’s name will not be shared.

**Who is paying for the study to take place?**

We are funded by Royal Holloway University of London and Great Ormond Street Hospital. This study is being conducted as part of an educational project for a clinical doctorate, which Rebecca MacKechnie is undertaking.

**Who has sponsored and reviewed the study?**

The study is being sponsored by Great Ormond Street Hospital and has been approved by the London-West London & GTAC Research NHS Ethics Committee (REC ref: 17/LO/0481.

**Contact for further information**

Thank you for your time and for considering taking part in the study. If you decide to take part, you will be given a signed consent form to keep.

If you require further information with regards to the study, please contact:

Miss Rebecca MacKechnie

Trainee Clinical Psychologist

[rebecca.mackechnie.2015@live.rhul.ac.uk](mailto:rebecca.mackechnie.2015@live.rhul.ac.uk)

Dr. Naomi Dale,

Consultant Clinical Psychologist,

Psychological Services

Great Ormond Street

London

WC1N 3JH

[naomi.dale@gosh.nhs.uk](mailto:naomi.dale@gosh.nhs.uk)

Appendix L

Information sheet for children



**What are the feelings of children who are sight impaired?**

*IRAS ID: 222711 REC Number:17/LO/0481 R&D Number:16NC33*

**Information for children**

**This information sheet should be read with the assistance of a parent**

We would like you and your parents to take part in our project. We call this a research project, as you and your parents help us to do the learning.

**We would like to learn more about your feelings. Sometimes children feel happy or excited or relaxed. Most children also have times when they feel sad or cross, or worried. We would like to ask you to tell us about your feelings. We would also like to ask you some questions about your everyday life.**

**This is because we want to understand more about the feelings of children who are sight impaired. This will help us to understand better how we can support children with sight impairment to have happy and interesting lives.**

**We would like to send you two questionnaires to fill in about your feelings. You can ask your parent to help you with this. You will do this at home. If you prefer, we can arrange to ask you the questions from the questionnaire over the phone.**

**We would also like to send your parents some questionnaires to fill in about your feelings and skills. We may also like to ask your parents some questions on the phone.**

Please let your parent know if there is anything you are unhappy about and if you are happy to join our project.

Thank you for learning about our project and for thinking about taking part.

Appendix M

Expression of interest form



**EXPRESSION OF INTEREST FORM**

**Project:** An investigation of mental health, social-emotional and behavioural profiles in children with congenital ophthalmological disorders and visual impairment.

Researchers' Names:

Dr. Naomi Dale (Chief Investigator, Consultant Clinical Psychologist)

Rebecca MacKechnie (Trainee Clinical Psychologist)

Dr. Helen Pote (Clinical Tutor)

**Thank you for taking the time to find out about this project!**

If you would like to hear more about participating in the project, please fill out the information below and send this form back to the research team. Once we have received your contact details, we will call you to discuss the project in more detail and to answer any questions you might have.

**Contact details:**

|  |  |
| --- | --- |
| **Child’s Full Name:** |  |
| **Child’s Date of Birth:** |  |
| **Child’s gender:** |  |
| **Your Full Name:** |  |
| **Relationship to Child:** |  |
| **Telephone:** |  |
| **Best Time to Contact:** |  |
| **Where did you hear about the project:** |  |

**Signed: \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_ Date: \_\_\_\_\_\_\_\_\_\_\_\_\_**

**PLEASE RETURN EXPRESSION OF INTEREST FORM IN PRE-PAID ENVEOPE**

**For further information please contact**

Rebecca MacKechnie

Trainee Clinical Psychologist

Clinical Neurosciences

Developmental Neurosciences Programme

UCL Institute of Child Health

30 Guilford Street, London

WC1N 1EH

Tel: 020 7905 2970 Email: Rebecca.mackechnie@nhs.net

**Details of how to contact the chief investigator:**

Dr. Naomi Dale

Senior Consultant Clinical Psychologist,

Psychological Services

Great Ormond Street Hospital NHS Trust

London WC1N 3JH

Tel: 020 7405 9200, extension 1199 Email: naomi.dale@gosh.nhs.uk

Appendix N

CONSENT FORM FOR MOTHER



**CONSENT FORM FOR MOTHER of child**

**Project:** An investigation of mental health, social-emotional and behavioural profiles in children with congenital ophthalmological disorders and visual impairment.

Researchers' Names:

Dr. Naomi Dale (Chief Investigator, Consultant Clinical Psychologist)

Rebecca MacKechnie (Trainee Clinical Psychologist)

Dr. Helen Pote (Clinical Tutor)

**Name of Child**: \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_ \_\_\_\_\_\_\_\_

For parent(s) to fill in:

|  |  |
| --- | --- |
| **Please initial** |  |
|  | I confirm that I have read and understand the information sheet for the above study (10/05/2017, Version 2) and have had the opportunity to ask questions. |
|  | I understand that my participation and my child’s participation is voluntary, and that I am free to withdraw at any time, without giving any reason, without my legal rights being affected. |
|  | I understand that relevant sections of my child’s medical notes and data collected during the study may be looked at by individuals from the sponsor organisation, regulatory authorities or from the NHS Trust where it is relevant to my taking part in this research. I give permission for these individuals to have access to my child’s records. |
|  | I am aware that the data collected as part of this study will be stored in anonymised form for up to 15 years and might be used in future studies. |
|  | I give permission for my child’s Paediatrician/Ophthalmologist/GP (if appropriate) to send information on my child’s vision and paediatric condition and vision and other paediatric test results (if available) to the Chief Investigator. |
|  | I understand that if the research team are concerned about a risk of harm to myself or my child, then the Great Ormond Street Hospital Policy for Safeguarding Children will be followed. |
|  | I give permission for my personal information (contact details) to be stored securely by the Chief Investigator of the Study so that I can be contacted with information about any related future studies. |
|  | I give permission for my child to participate in the above study. |
|  | I give my consent to participate in the above study. |

*A signed copy of this consent form is to be retained by the parent and the Chief Investigator of the project.*

\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_ \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_ \_\_\_\_\_\_\_\_\_\_\_\_ **Parent's Name** **Parent's Signature Date**

\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_ \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_ \_\_\_\_\_\_\_\_\_\_\_\_

**Investigator's Name** **Investigator's Signature Date**

Appendix O

Ethical approval



**London - West London & GTAC Research Ethics Committee**

The Old Chapel

Royal Standard Place

Nottingham

NG1 6FS

**Please note: This is the favourable opinion of the**

**REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval**

06 June 2017

Dr Naomi Dale

Psychological Services

Great Ormond Street

London

W1CN 3JH

Dear Dr Dale

|  |  |
| --- | --- |
| **Study title:** | **An investigation of mental health, social-emotional and behavioural profiles in children with congenital ophthalmological disorders and visual impairment.** |
| **REC reference:** | **17/LO/0481** |
| **IRAS project ID:** | **222711** |

Thank you for responding to the Committee’s request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to make a request to postpone publication, please contact [hra.studyregistration@nhs.net](mailto:hra.studyregistration@nhs.net) outlining the reasons for your request.

**Confirmation of ethical opinion**

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

**Conditions of the favourable opinion**

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission must be obtained from each host organisation prior to the start of the study at the site concerned.

*Management permission should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).*

*Guidance on applying for NHS permission for research is available in the Integrated Research Application System,* [*www.hra.nhs.uk*](http://www.hra.nhs.uk) *or at* [*http://www.rdforum.nhs.uk*](http://www.rdforum.nhs.uk)*.*

*Where a NHS organisation’s role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.*

*For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.*

*Sponsors are not required to notify the Committee of management permissions from host organisations*

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact [hra.studyregistration@nhs.net](mailto:hra.studyregistration@nhs.net). The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

**It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).**

**Ethical review of research sites**

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

**Approved documents**

The final list of documents reviewed and approved by the Committee is as follows:

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
| *Document* | *Version* | | *Date* | |
| Costing template (commercial projects) [Cost Form] | 1 | | 18 November 2016 | |
| Covering letter on headed paper [Responses to REC comments] | | | | |
| Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Indemnity Letter] | | 1 | | 01 August 2016 |
| GP/consultant information sheets or letters [Letter to GP] | | 1 | | 10 May 2017 |
| IRAS Application Form [IRAS\_Form\_01032017] | |  | | 01 March 2017 |
| IRAS Application Form XML file [IRAS\_Form\_01032017] | |  | | 01 March 2017 |
| IRAS Checklist XML [Checklist\_01032017] | |  | | 01 March 2017 |
| IRAS Checklist XML [Checklist\_13032017] | |  | | 13 March 2017 |
| IRAS Checklist XML [Checklist\_26052017] | |  | | 26 May 2017 |
| Letter from funder [Funding approval] | | 1 | | 16 December 2016 |
| Letter from statistician [Statistician Letter] | | 1 | | 05 May 2017 |
| Letters of invitation to participant [Flyer] | | 1 | | 16 February 2017 |
| Letters of invitation to participant [Flyer, TRACKED CHANGES] | | 2 | | 24 May 2017 |
| Letters of invitation to participant [Flyer, Clean Version] | | 2 | | 24 May 2017 |
| Non-validated questionnaire [Demographics Questionnaire] | | 1 | | 16 February 2017 |
| Non-validated questionnaire [Child Medical Information] | | 1 | | 16 February 2017 |
| Other [Research Report Template] | | 1 | | 13 March 2017 |
| Other [Research Protocol, TRACKED CHANGES] | | 2 | | 24 May 2017 |
| Other [Research Protocol, CLEAN VERSION] | | 2 | | 24 May 2017 |
| Participant consent form [Consent Form - Father] | | 1 | | 10 May 2017 |
| Participant consent form [Consent Form] | | 1 | | 16 February 2017 |
| Participant consent form [Consent Form, Mother, TRACKED CHANGES] | | 2 | | 10 May 2017 |
| Participant consent form [Consent Form, Mother, CLEAN VERSION] | | 2 | | 10 May 2017 |
| Participant information sheet (PIS) [Expression of Interest] | | 1 | | 16 February 2017 |
| Participant information sheet (PIS) [Participant Information Sheet - Mother, TRACKED CHANGES] | | 2 | | 10 May 2017 |
| Participant information sheet (PIS) [Participant Information Sheet - Mother, CLEAN VERSION] | | 2 | | 10 May 2017 |
| Participant information sheet (PIS) [Participant Information Sheet - Father] | | 1 | | 10 May 2017 |
| Participant information sheet (PIS) [Information sheet for children] | | 1 | | 03 May 2017 |
| Referee's report or other scientific critique report [Critique and responses – Royal Holloway University of London] | | 1 | | 18 November 2016 |
| Referee's report or other scientific critique report [Project Approval - RHUL] | | 1 | | 16 December 2016 |
| Referee's report or other scientific critique report [CRAC approval] | | 1 | | 28 February 2017 |
| Summary CV for Chief Investigator (CI) [Naomi Dale CV] | | 1 | | 01 March 2017 |
| Summary CV for student [Student CV] | | 1 | | 28 February 2017 |
| Summary CV for supervisor (student research) [Helen Pote CV] | | 1 | | 09 March 2017 |
| Validated questionnaire [RCADS] | | 1 | |  |
| Validated questionnaire [SDQ] | | 1 | | 28 February 2017 |
| Validated questionnaire [SCORE-15] | | 1 | | 28 February 2017 |
| Validated questionnaire [ABAS III] | | 1 | | 13 March 2017 |
| Validated questionnaire [SRS-2] | | 1 | | 09 March 2017 |
| Validated questionnaire [RCADS Child Report] | | 1 | | 25 May 2017 |
| Validated questionnaire [PedsQL Parent] | | 1 | | 25 May 2017 |
| Validated questionnaire [PedsQL Child] | | 1 | | 25 May 2017 |

**Statement of compliance**

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

**After ethical review**

Reporting requirements

The attached document *“After ethical review – guidance for researchers”* gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

* Notifying substantial amendments
* Adding new sites and investigators
* Notification of serious breaches of the protocol
* Progress and safety reports
* Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

**User Feedback**

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: <http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/>

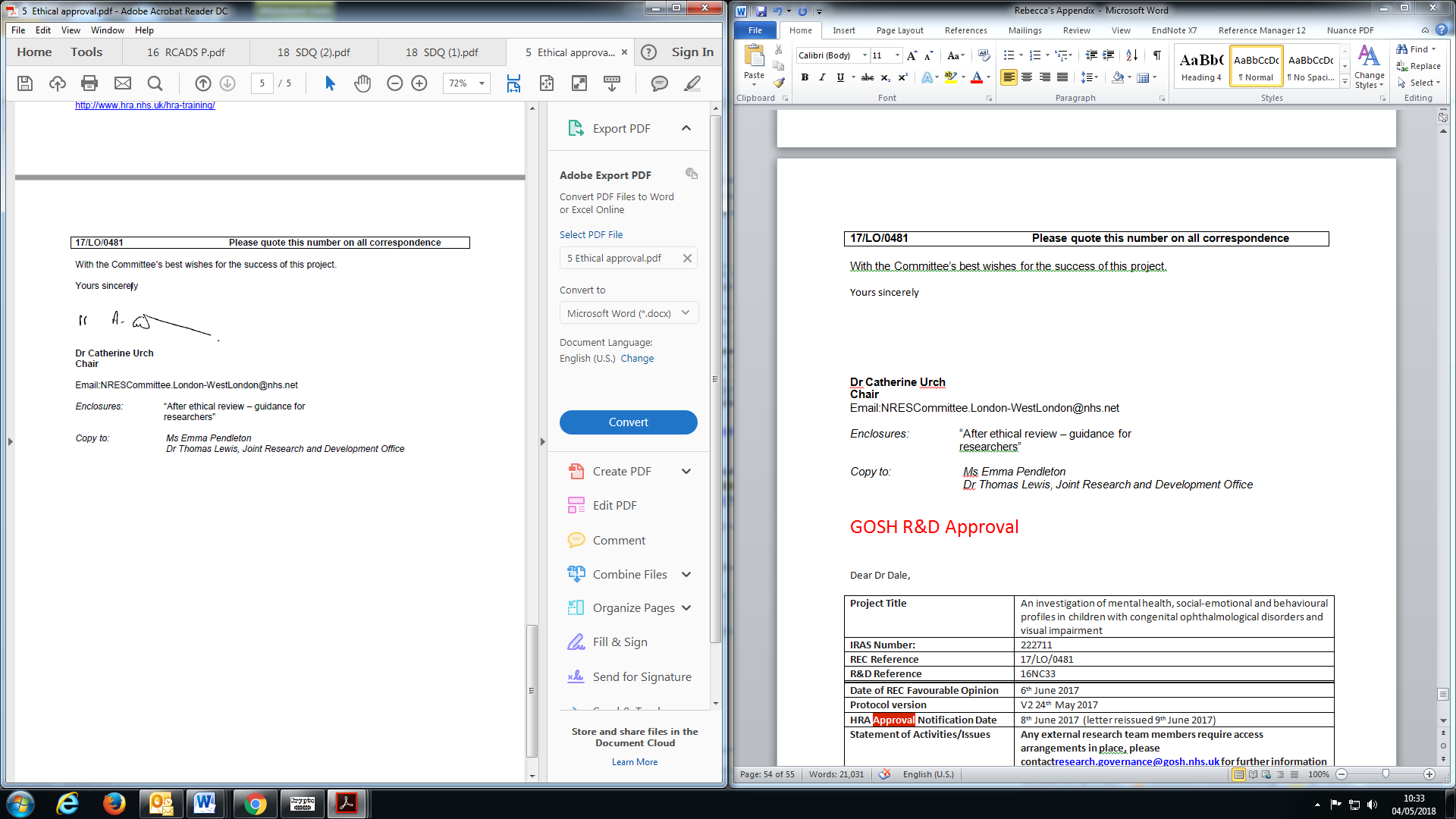
**HRA Training**

We are pleased to welcome researchers and R&D staff at our training days – see details at <http://www.hra.nhs.uk/hra-training/>

**17/LO/0481 Please quote this number on all correspondence**

With the Committee’s best wishes for the success of this project.

Yours sincerely



**Dr Catherine Urch**

**Chair**

Email:NRESCommittee.London-WestLondon@nhs.net

*Enclosures:* “After ethical review – guidance for

researchers”

|  |  |
| --- | --- |
| *Copy to:* | *Ms Emma Pendleton*  *Dr Thomas Lewis, Joint Research and Development Office* |

Appendix Q

GOSH R&D Approval

Dear Dr Dale,

|  |  |
| --- | --- |
| **Project Title** | An investigation of mental health, social-emotional and behavioural profiles in children with congenital ophthalmological disorders and visual impairment |
| **IRAS Number:** | 222711 |
| **REC Reference** | 17/LO/0481 |
| **R&D Reference** | 16NC33 |
|  |  |
|  |  |
| **Date of REC Favourable Opinion** | 6th June 2017 |
| **Protocol version** | V2 24th May 2017 |
| **HRA Approval Notification Date** | 8th June 2017 (letter reissued 9th June 2017) |
| **Statement of Activities/Issues** | **Any external research team members require access arrangements in place, please contact**[**research.governance@gosh.nhs.uk**](mailto:research.governance@gosh.nhs.uk)**for further information if necessary.**    **Research procedures at ICH (including data analysis/storage) will require an ICH risk assessment approval to be in place. Please contact**[**kate.thornton@ucl.ac.uk**](mailto:kate.thornton@ucl.ac.uk)**for further advice if necessary.** |

This research project has been reviewed locally at Great Ormond Street Hospital NHS Foundation Trust for capacity and capability. After review we can confirm a **Notification of No Objection** to this research being conducted at GOSH .

Please contact us using the contact details below if you require any further information.

We wish you all the best with your research.

Kind regards

Dr Thomas Lewis

Appendix R

Moorfields HRA Approval

Dear Sponsor Representative,

**RE: IRAS 222711 - Confirmation of Capacity and Capability at Moorfields Eye Hospital NHS Foundation Trust.**

|  |  |
| --- | --- |
| **Full Study Title:** | An investigation of mental health, social-emotional and behavioural profiles in children with congenital ophthalmological disorders and visual impairment. |
| **Site Local Collaborator** | Dr  Ngozi Oluonye |
| **Protocol version:** | V2.0 24.05.17 |
| **Latest HRA Approval date:** | 08.06.17 – re-issued 09.06.17 |

This email confirms that **Moorfields Eye Hospital NHS Foundation Trust** has the capacity and capability to deliver the above referenced study. Please find attached the agreed Statement of Activities  as confirmation.

Please note, in line with the national HRA approvals process, you will no longer receive a NHS R&D Approval/Permission letter.

If you have any questions please do not hesitate to get in touch.

Kind regards

**Tania West**

Research Portfolio Manager

Research and Development, Moorfields Eye Hospital NHS Foundation Trust, 162 City Road, London, EC1V 2PD

**Tel:** 020 7253 3411 x2036 | **Email:**  Tania.west@moorfields.nhs.

Appendix S

Research Report Template



**Research report**

\* IMPORTANT INFORMATION ABOUT THIS RESEARCH REPORT \*

You completed some questionnaires about your child as part of a scientific research study, rather than for clinical purposes. The results of research studies may be less accurate and less detailed than a comprehensive clinical assessment. Therefore**, the results of these questionnaires cannot directly contribute to an individual’s healthcare or education.** For a more detailed explanation of how research and clinical assessments differ, and why this is important, please see the appendix at the end of this report. You may share this report with the professionals involved in your child’s care if you wish but we cannot enter into discussion of these questionnaire scores as this is not a clinical assessment. If any area has been indicated as possibly needing more attention, a further clinical assessment may be suggested.

|  |  |
| --- | --- |
| **Name:** <NAME> | **Date of assessment:** XXX |
| **Date of birth:** XXX | **Researcher:** Rebecca MacKechnie |
| **Age:** X years, X months | **Researcher position:**  Clinical Researcher |

**Research project:** An investigation of mental health, social-emotional and behavioural profiles in children with congenital ophthalmological disorders and visual impairment.

**Research project lead:** Dr Naomi Dale, Head of Psychology (Neurodisability) Great Ormond Street Hospital & Reader, UCL Institute of Child Health, Developmental Neurosciences Programme

**Your research contact:** Rebecca MacKechnie

Email: Rebecca.mackechnie.2015@live.rhul.ac.uk

Clinical Neurosciences Section

Developmental Neurosciences Programme

UCL Institute of Child Health

30 Guilford Street | London | WC1N 1EH

**Ethical approval:** This research project was reviewed and given ethical approval by the NRES Committee London – West London Research Ethics Committee (ethics reference number 17/LO/0481).

**Aims of the research project:** Children with long term health conditions are more likely to have mental health difficulties than children who are healthy. Some research has shown that children with congenital visual impairments, who have had a visual impairment since birth, are also at risk for a range of difficulties including social, emotional and behavioural difficulties. There has been very little research in this important area.

This study aims to investigate the strengths and difficulties of children with congenital visual impairment, in detail. The findings will highlight specific areas that children with congenital visual impairment might need extra help with. This will help clinicians, educators and doctors to know how best to support children with congenital visual impairment and their families.

This is a research report and is not a clinical report. It is based on the parent’s responses to a range of standard questionnaires. Information has been given of whether the child’s ratings are in the average range for their age or whether they are in the ‘raised’ area, which may suggest some clinical concern. For further information please see the study information sheet included with this report, or contact the research team directly.

|  |  |  |
| --- | --- | --- |
| **Questionnaires** | | |
| **Measure** | **What does this assess?** | **range** |
| General strengths and difficulties | Child’s behaviour report according to average range for age or ‘raised’ if in the clinical range and may be of concern | Appeared… |
| Emotional mood | Child’s anxiety and mood report according to average range for age or ‘raised’ if in the clinical range and may be of concern | Appeared… |
| Social responsiveness | Child’s social abilities report according to average range for age or ‘raised’ if in the clinical range and may be of concern | Appeared… |
| Adaptive behaviour | Child’s adaptive/independent skills report according to average range for age or ‘raised’ if in the clinical range and may be of concern | Appeared… |

**Things to note:**

<FLAG UP ANY DOMAINS WHICH MIGHT REQUIRE FURTHER ASSESSMENT BY A CLINICIAN (those in the clinically significant range) e.g. NAME’S score on a measure of anxiety was in the ‘raised’ range and may be of clinical concern. . This may require further assessment or support by a clinician. For information on how to arrange this, please see the ‘Further Information’ section at the end of this report.>

**Further Information**

**I have questions about this report. Who should I contact?**

Please do not hesitate to contact the research team directly (contact details are provided on page 1).

**I am concerned about my child’s report, what can I do now?**

Depending on the age of the child, discuss this report with your child’s GP and school, and any other health or educational professionals involved in his or her care. Ask whether a detailed assessment by a Clinical or Education Psychologist or other clinician would be helpful. If so, a doctor or teacher should be able to arrange this.

**A doctor or a teacher has questions about this report. Who should they contact?**

Please ask them to contact the research team directly (contact details are provided on page 1).

**Appendix**

The way clinicians and researchers perform psychological assessments differs in important ways. These differences influence how the results can be interpreted and used, and are described below.

**A clinician will…**

* Perform a comprehensive assessment based on an individual’s own needs.
* Take a detailed history from the parent and other professionals involved with the child and consider their performance in different environments e.g. at home and school.
* Use tests directly with the individual, as well as giving questionnaires to parents to complete.
* Test a range of abilities so they can build up a picture of an individual’s strengths and weaknesses.
* Use clinical judgement and expertise to interpret the results and their significance for the child.

**A researcher will…**

* Perform a limited set of assessments based on the aims of their study. It might be that the specific assessments a researcher performs happen to test the things a participant finds particularly easy or particularly difficult. This only provides part of the picture, and might not accurately reflect how someone is doing overall.
* Send all of the questionnaire to parents to complete in one go. Results may therefore be influenced by distraction, fatigue or poor concentration.
* A range of other situational factors may also influence results. All of these things mean there is some uncertainty around how accurate the results are. This level of uncertainty is ok for researchers, because they only look for differences between *groups* of people. However, this level of uncertainty does make it difficult to make confident conclusions for an individual participant.
* We are using measures that have not been used before with children with visual impairment so we are not sure about their validity and they need interpreting cautiously.

1. ‘Complicated’ CDPVS describes CDPVS conditions which are part of a neuroophthalmic condition, such as cataracts in children who have Down Syndrome. [↑](#footnote-ref-1)
2. Low vision’ is defined as vision acuity (the finest detail that can be seen) of less than 6/18 (logMAR 0.5) but better than 3/60 (logMAR 1.4) and ‘blindness’ is defined as vision acuity of less than 3/60 (logMAR 1.4). [↑](#footnote-ref-2)
3. ‘Complicated’ CDPVS describes CDPVS conditions which are part of a syndromic condition involving the brain, such as cataracts in children who have Down Syndrome. [↑](#footnote-ref-3)
4. The following items were removed and prorated: ‘Looks at other people’s face when they are talking to him or her’, ‘Looks both ways before crossing a street or parking lot’ and ‘Looks at pictures or read books or magazine’s during free time’. [↑](#footnote-ref-4)
5. Parents of children who used braille were asked to complete the following items as if the question asked about braille skills: ‘Writes of prints his or her first and last name’, ‘Reads his or her name when printed’, ‘Reads and obeys common signs’, ‘Tells time correctly, using a watch or clock with hands’, ‘Reads menus at restaurants’, ‘Writes his or her address, including zip code’. [↑](#footnote-ref-5)
6. The following items were removed and prorated: ‘Avoids eye contact or has unusual eye contact’, and ‘stares or gazes off into space’. [↑](#footnote-ref-6)
7. (0.2 = small, 0.5= medium, 0.8 = large). [↑](#footnote-ref-7)
8. ICC’s are rated as; 0-0.40 poor agreement, 0.41-0.60 moderate agreement, 0.61-0.80 good agreement and 0.81-100 excellent agreement. [↑](#footnote-ref-8)
9. Strength of correlation was described as weak (r=.10 to .29), moderate (r=.30 to .49) or strong (r=.50 to 1.0). [↑](#footnote-ref-9)
10. Socioeconomic Status was calculated based on The National Statistics Socio-economic classification (NS-SEC, Office for National Statistics). For each child, details of the parent with the highest NS-SEC classification was used as a marker of socio-economic status. [↑](#footnote-ref-10)
11. In relation to vision disorders, children may have more than one disorder, but the primary disorder only is reported here. [↑](#footnote-ref-11)
12. Two children had ‘potentially simple’ CDPVS which were later onset and one child had ‘potentially simple’ CDPVS with a comorbid severe intellectual disability. [↑](#footnote-ref-12)
13. *(Total Sample aged 8-12, USA, Ebesutani et al., 2011).* [↑](#footnote-ref-13)
14. *(Total Sample aged 5-15, UK, Goodman 2001).* [↑](#footnote-ref-14)
15. *(Total Sample aged 5-16, USA, Varni, Burwinkle, Seid & Skarr, 2003).* [↑](#footnote-ref-15)
16. *(Total Sample aged 5-8, UK, Wigham, McConachie, Tandos & Couteur, 2012).* [↑](#footnote-ref-16)
17. (Total Sample, aged 0-89 years, USA, Harrison & Oakland, 2013). [↑](#footnote-ref-17)
18. CDPVS refers to the origin of VI in the globe, retina or anterior optic nerve and ‘potentially simple’ refers to VI with no known neurological involvement [↑](#footnote-ref-18)
19. logMAR 0.5 to 1.3 [↑](#footnote-ref-19)
20. logMAR 1.4 or worse [↑](#footnote-ref-20)