

The nature of the anxiety in children with Selective Mutism.

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20th August 2015

*This thesis is presented in partial fulfilment of the requirements for the degree of Doctor
of Psychology (Clinical Psychology)*

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Thesis Abstract

Research demonstrates that children with selective mutism (SM) present with symptoms of social anxiety and there is a high comorbidity between SM and social phobia (SP). As a result some researchers have questioned whether SM is a variant of SP rather than a distinct anxiety disorder. This thesis sought to clarify the association between SM and SP. Participants were 25 children with a primary diagnosis of SM, 17 children with a primary diagnosis of SP and 15 children identified as “easy-going” who did not meet criteria for a DSM-IV diagnosis. Children were aged between 4 to 8 years. Data included mother, father, teacher and child reports, a semi-structured clinical interview with the child’s mother, and an observational assessment with the child and their mother.

The first paper in the thesis reports on the development and psychometric properties of the child self-report measure of mutism, Selective Mutism Questionnaire – Child (SMQ-C). The SMQ-C was adapted from a similar adult measure and was designed for children aged from 4 years. The SMQ-C showed good internal consistency, convergent and divergent validity. Even young children were able to provide a clear picture of how much they spoke in various contexts.

The second paper systematically compared children with SM, SP and non-clinical controls on social anxiety symptomatology and behavioural inhibition. Observational measures showed that children in the SM group were more verbally and nonverbally inhibited than children in the SP group and nonclinical controls, and parent report revealed that they were more fearful of negative evaluation than children in the SP group who did not differ significantly from non-clinical controls. These findings indicate that children with a primary diagnosis of SM show similar or even greater

social phobia symptomatology than children with a primary diagnosis of SP and both are more socially anxious than non-clinical controls.

The third paper explored relationships between social anxiety, parents' negative affectivity, and parenting practices among children with SM, SP and controls. No significant differences in the levels of negative emotion or social phobia symptomatology among mothers and fathers were found between the three groups, nor did the study find any significant differences in parent-child interaction with one exception. Mothers and fathers of children with a primary diagnosis of SM reported more thoughts of frustration with their child's reticence in a novel social situation. Correlational analyses showed a significant association between maternal social phobia and child anxiety in the SM and non-clinical groups.

Overall, the findings support the idea postulated in the literature that SM is a more severe form of SP observed in younger children. Speech is the primary form of communication in all social interactions, the high social anxiety together with the spoken and unspoken pressures to speak contribute to maintenance of the disorder.

Declaration

I certify that this thesis contains no material which has been accepted for the award of any other degree in any university or other tertiary institution other than Macquarie University and, to the best of my knowledge and belief, contains no material previously published or written by other persons, except where due reference has been made in the text.

I certify that this thesis is an original piece of work and that it has been written by me. For each primary study, I was responsible for the formulation of the research questions, ethics application, study and project management, data preparation, statistical analysis and preparation of the papers. Professor Ron Rapee, as the Primary Supervisor, and Dr Lorna Peters, as my Associate Supervisor, supervised the research development, preparation and writing of each paper.

The research was conducted predominately in the time that I was employed as a Clinical Psychologist at Redbank House, Western Sydney Local Health District.

I give consent to this copy of my thesis, when deposited in the Macquarie University Library, being made available for loan subject to the provisions of the Copyright Act 1968.

The conduct of this research was approved by the Western Sydney Local Health District Human Ethics Committee under approval number *HREC/10/WMEAD/179* *SSA/11/WMEAD/190*, NSW Department of Education and Communities (*SERAP Number 2010175*), and the Catholic Education Diocese of Parramatta (letter dated 24-11-2011). External approval was noted by the Ethics Secretariat of the Human Research Ethics committee of Macquarie University (*REF 5201100751*).

Signed: _____ DATE: _____

Maria Ivanka Milić

Acknowledgements

The traditional saying “it takes a community to raise a child” describes my journey. It has taken a community to support me to achieve my goal of translating my clinical work and learning into research. There are many people to thank.

Firstly, mama i tata (my parents), bez vaše ljubavi, pomoći, podrške i molitve nebi ovo postigla. My dear children who have watched, helped in their own little ways, and waited for mama to finish this journey to begin the adventure of working through the long list of fun things put on hold with the statement “when you finish uni mama can we...”. My muž, Mario, who has supported me on this journey. My dear brothers, Filip and Niko, and their partners/my dear sister-in laws, Rita and Anđa, for your constant support and faith that I could do this. Also, the never ending cuddles from my beautiful nieces and nephews, Jake, Anica, Luke, Alana, Isabelle and Liliana. My dear extended family, kumovi (Vera, Lena, Nellie and Natalie and their families), and valued friends for your unconditional support, many hugs, unconditional faith “you can do it”, and the everyday help with mama duties and other tasks for which there was not enough time. Thank you for all the extra cuddles and care you gave my girls in this time.

Secondly, a HUGE thank you to my supervisor Ron, for helping me translate my clinical learning into a research project. Without your patience, calm reflections, immense knowledge, input, curious questions and quiet confidence throughout the journey, I would not have achieved my goal. Thank you also to Lorna for the input and support in the early stages, especially the emotional ups and downs of trying to juggle being a researcher whilst being a mama of littlies.

Thirdly, Dr Megan Chambers and Dr John Brennan, for creating the opportunity to pursue my interest in working with children with Selective Mutism. Megan, your confidence in my abilities kept me going, particularly at those times that when it got tough and I became uncertain. And the dear friendships that started at work, Michelle, Holly, Hris, Antoinette, Melissa and many more, thank you.

Finally, a huge thank you to Chris Basten, Jody and the team at Basten and Associates.

Hvala Blažena Djevica Marija, čuvala si mene i moju obitelj kroz ovaj dug put.

CHAPTER 1

GENERAL INTRODUCTION

1. Definition of Selective Mutism

The defining criterion for a diagnosis of Selective Mutism is the persistent failure to use spoken language in select social situations where speech is anticipated and/or expected (DSM-5: APA, 2014). Children with Selective Mutism (SM) understand and are able to use spoken language. They speak freely to immediate family members when alone in their own home. The extent to which they fail to speak varies. In the more severe presentations, children fail to use spoken language with adults and children at (pre)school, in community social activities, with extended family and even with immediate family in the vicinity of other people (Black & Udhe, 1995; Kearney, 2010; McHolm, Cunningham & Vanier, 2005; Roe, 2015). The prolonged nature of the mutism is usually first noticed after a period of attendance in an educational setting, either preschool or school (Sharp, Sherman & Gross, 2007). There is often an expectation belief that the lack of speech is a normal developmental behaviour that the child will outgrow (Andersson & Thomson, 1998).

For a formal DSM-5 (*Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition*: APA, 2014) diagnosis of SM, the mutism needs to have been present for at least one month, not including the first month of (pre)school. The diagnosis is not given if the prolonged failure to speak can be understood as being part of a speech and language delay or Autism Spectrum Disorder (APA, 2014). For children learning a new language, the diagnosis is commonly withheld whilst the child builds competency and confidence in speaking the new language, and mutism needs to be present in the primary and new language (Busse & Downey, 2011; Krynski, 2003; Tancer, 1992; Toppelberg, Tabors, Coggins, Lum and Burger, 2005). The final criterion for diagnosis, and the reason why families seek treatment, is that the prolonged mutism impacts on a

child's friendships; their level of academic, social and community participation; and achievement of developmentally appropriate independence (APA, 2014).

Treatment is generally sought several years after adults first notice the lack of speech (Black & Udhe, 1995; Dummit, Klein, Tancer, Asche, Martin & Fairbanks, 1997). Treatment-seeking children are typically in the early years of formal schooling, and the efforts of parents and teachers have not been successful in helping the child begin to use their voice (Johnson & Wintgens, 2001). Given the early onset, duration of the mutism and the impact on functioning, early intervention is imperative. Effective treatment is dependent on a clear, evidence-based understanding of why some children remain mute in select social interactions.

2. A historical overview of Selective Mutism

The first published account of children failing to speak in select situations appeared in 1877 with Kussmaul's description of three cases of "asphasia voluntaria", which implied the failure to speak was voluntary (Cline, 2015; Halpern, Hammond & Cohen, 1971; Lesser-Katz, 1986). Various other terms were used in the first half of the 1900's to describe this same presentation, including speech shyness, speech avoidance, negativism, functional mutism and psychogenic mutism (Adam and Glasner, 1954; Halpern et al., 1971; Kratochwill, Brody & Peirsel, 1979). The descriptive term "Elektiver Mutismus bei Kindern" or Elective Mutism was coined by Tramer in 1934 to describe cases of children who spoke with immediate family members and same age friends but were silent with everyone else (Salfield, 1950). Tramer noted the lack of speech had no organic basis. Like Kussmaul, he hypothesised the mutism was volitional and the child was "electing" not to speak (Elson, Pearson, Jones & Schumacher, 1965; Kratochwill, 1981).

From the 1940's to early 1980, Elective Mutism (EM) was the most commonly used descriptive label for this presentation. Published research consisted of single case studies (Brison, 1966; Chethik, 1973; Mora, DeVault & Schopler, 1962), case study series (Adams & Glasner, 1954; Hayden, 1980; Meijer, 1979; Morris, 1953; Pustron & Speer, 1964; Reed, 1963; Salfield, 1950) and retrospective reviews of clinical cases (Adam and Glasner, 1954; Hayden, 1980; Kolvin & Fundudis, 1981; Krohn, Weckstein Wright, 1992; Parker, Olsen & Throckmorton, 1960; Wergerland, 1979; Wright, 1968). Prolonged mutism in certain situations and/or with certain people was identified as the primary symptom of EM. There were discrepancies between studies on inclusion criteria. These discrepancies continued following the addition of Elective Mutism as a distinct disorder in the "Other Disorders of Infancy, Childhood or Adolescence" of the *Diagnostic and Statistical Manual of Mental Disorders, Third Edition* (DSM-III: APA, 1980). Discrepancies included:

- (i) differences in which relationships the mutism occurred (e.g. not speaking to immediate family vs not speaking outside immediate family) (Hayden, 1980; Morris, 1953; Paniagua & Saeed, 1988; Salfield, 1950),
- (ii) duration of the mutism (from six months to two years)(Carlson, Kratochwill & Johnson, 1994; Clive & Baldwin, 1984),
- (iii) degree of mutism (total mutism for a distinct period and/or mutism in select situations and with select persons) (Clive & Baldwin 1994; Paniagua & Saeed, 1988), and
- (iv) inclusion of cases with individual, family and environmental symptoms (Adam & Glasner, 1954; Browne, Wilson & Laybourne, 1963; Hayden, 1980; Herbert, 1959; Morris, 1953; Parker et al, 1960; Reed, 1963; Salfield, 1950) that would be

excluded under the *Diagnostic and Statistical Manual of Mental Disorders* (DSM) criteria.

Despite the discrepancies, there were commonalities in the descriptions of the children identified as being EM. Most consistently, the mutism was seen as not having a neurological cause. The child's disposition or temperament was described as "sensitive"/"hypersensitive", "timid", "shy", "slow to warm up", "negativistic" and/or "inhibited" (Adam & Glasner, 1954; Halpern et al., 1971; Hayden, 1980; Meijer, 1979; Morris, 1953; Salfeld, 1950; Wergeland, 1979). The children were socially avoidant, and for the majority of cases, the mutism occurred in relationships outside of the immediate family. The mutism was first observed in the toddler years when the children began to enter situations in which they were expected to speak with unfamiliar people (Salfeld, 1950; Lesser-Katz, 1986). There were many commonalities between the description of EM and the diagnostic criteria of Avoidant Disorder of Childhood and Adolescence, a childhood disorder introduced into the "Other Disorders of Childhood and Adolescence" category of *Diagnostic and Statistical Manual of Mental Disorders Third Edition* (DSM-III: APA, 1980) at the same time as Elective Mutism. Unlike Avoidant Disorder of Childhood and Adolescence where the restricted speech was seen as an anxious response to persons outside the immediate family, clinicians and researchers continued to see the mutism in EM as volitional. They hypothesised that the child was "electing" not to speak to suppress the expression of negative emotions (most commonly thought to be anger, hostility, defiance or depressive feelings), arising from either the intra-psychic conflict in the child or in response to hostile and/or unsafe family interactions and/or parent behaviour (Adam & Glasner, 1954; Browne et al, 1963; Busse & Downey, 2011; Elson et al., 1965; Hesselman, 1983; Hultquist, 1965; Mora et al., 1968; Parker et al, 1960; Salfeld, 1950; Wergeland, 1979; Wright, 1968).

Clinical assumptions were made about the contributing causal and maintaining factors including: a traumatic event preceding and/or during language development, potential and/or real loss of the maternal relationship, parent mental health problems, and ongoing family and/or marital relationship problems (Adams & Glasner, 1954; Browne et al, 1963; Halpern et al, 1971; Hayden, 1980; Hesselman, 1973; Krolian, 1988; Morris, 1953; Parker et al, 1960; Pustrom & Speers, 1964; Salfeld, 1950; Sluckin, 1977).

Treatment up to mid 1960's focused on resolving the hypothesised causal and maintaining factors. Speech was not the central focus of therapy. Treatment predominately consisted of intensive individual therapy with the child; long individual inpatient admissions (of one or more years) with infrequent, time limited contact with the family; and/or special school placement. In some cases there was concurrent individual therapy with the child's mother or family therapy (Wright, Holmes, Cuccaro & Leonhardt, 1994). The general consensus was that the symptom of mutism was difficult to treat (Kratowill et al, 1979). In cases where there was an observed improvement in the child's speech, these improvements occurred outside the therapy (Pustrom & Speers, 1964) and in context where there was no pressure to speak (Reed, 1963).

From the mid 1960's, the emergence of Behaviour Therapy saw speech become the focus of treatment. The assumption of intentionality was removed and the mutism was viewed as a learned pattern of behaviour (Porjes, 1992). Treatment, which consisted of a single behavioural technique or multimodal behavioural therapy, was implemented in outpatient clinic sessions and at the child's regular school with the active involvement of the teacher. Behavioural strategies included: contingency and token reward programmes, response costs, shaping, self-modelling, escape-avoidance

and stimulus fading. The interventions were observed to be more effective than previous individually focused treatments as there was an observed increase in speech, and the observed gains in speech were attributed to the intervention rather than natural remission over time (Anstendig, 1999; Asutad, Sininger & Stricklin, 1980; Bauermeister & Jemail, 1975; Colligan, Colligan & Dilliard, 1977; Kratochwill et al, 1979; Nash, Thorpe, Andrews & Davies, 1979; Rasbury, 1974; Reid, Hawkins, Keutzer, McNeal, Phelps, Reid & Mees, 1967; Rosenbaum & Kellman, 1973; Sanok & Striefel, 1979; Tancer, 1992).

The shift from viewing the mutism as a volitional behaviour with the child “electing” not to speak to viewing the mutism as a behavioural response that occurred in “select” social interactions (Dummit et al 1997; Krynski, 2003) was incorporated into the *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition* (DSM-IV) in 1994 (APA, 1994). The name of the disorder was changed to Selective Mutism and a minimum duration of the mutism (one month not including the first month of school) was specified. From the early 1990’s methodologically improved research studies began to emerge comparing larger groups of children with SM to other clinical disorders and controls. The studies used well-validated semi-structured interviews, questionnaires and behavioural experiments. Information was sought from various sources including the child, their parents, teachers and clinician observations. These studies provided strong empirical evidence that SM was an anxiety based disorder, and the anxiety emerged in social interactions with adults and children outside of the home (Bögels, Alden, Beidel, Clark, Pine, Stein & Voncken, 2010; Cohan, Price & Stein, 2006; Viana, Beidel & Rabian, 2009). This was exemplified through the reclassification of SM in *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition* (DSM-5) in 2014 to the

section on “Anxiety Disorders” (APA, 2014). The criteria for a diagnosis of SM remained unchanged from DSM-IV to DSM-5.

3. Descriptive features of Selective Mutism

3.1. Demographic features of Selective Mutism

3.1.1. Prevalence

Early reports suggested the disorder was relatively rare affecting less than 0.08% of the population (Browne and Lloyd, 1975; Cline and Baldwin, 1994). Methodologically improved prevalence studies reveal rates ranging between 0.7% to 3.3% of children in the first three years of school (Karakaya, Sismanlar, Memik, Coskun, Agaoglu and Yavuz, 2008; Kopp and Gillberg, 1997; Kumpulainen, Räsänen, Raaska and Somppi, 1998), 0.4% to 0.76% in the preschool population (Elizur and Perednik, 2003, Ezpeleta, de la Osa & Doménech, 2014) and 1.5% in a study of all 3 year olds in a district of New York (Bufferd, Dougherty, Carlson and Klein, 2011). Additionally, a higher percentage of children with SM than community children spoke a primary language that was discordant with the primary language spoken in the community (Bergman, Gonzalez, Piancentini & Keller, 2013; Bradley and Sloman, 1975; Brown and Lloyd, 1975; Elizur and Perednik, 2003).

There is a strong likelihood that the preschool and school studies are an underestimate of prevalence. Children identified in these studies as having SM had a more chronic form of mutism as they had failed to speak for more than six months. The diagnosis of SM requires the mutism be present for one month, not including the first month of school. Browne and Lloyd (1975) using a stricter criterion of no speech at school, found a prevalence rate of 0.7% eight weeks after starting Kindergarten and this

declined to 0.08% at 6 months and 0.01% at 12 months. This would suggest that a number of children display a more “transient” form of mutism, lasting less than six months, who were not included in the abovementioned prevalence studies.

3.1.2. Gender

Both epidemiological and larger clinical studies show mixed results with regard to gender distribution. There appear to be fewer studies showing a near equal distribution of males and females (Anderson & Thomsen, 1998; Bergman et al, 2013; Brown & Lloyd, 1975; Carbone, Schmidt, Cunningham, McHolm, Edison et al, 2010; Karakaya et al, 2008; Kristensen & Oerbeck, 2006; Nowakowski, Cunningham, McHolm, Evans, Edison et al, 2009) than studies that have a greater proportion of females to males in the SM sample, with the ratio of females to males ranging from 1.4:1 to 3:1 (Cohan, Chavira, Shipon-Blum, Hitchcock, Roesch & Stein, 2008; Black & Udhe, 1995; Dummitt et al., 1997; Ezpeleta et al, 2014; Ford, Sladeczek, Carlson & Kratochwill, 1998; Kristensen, 2002; Kumpulainen et al, 1998; Levin-Decanini, Connolly, Simpson, Suarez & Jacobs, 2013; Steinhausen & Juzi, 1996).

3.1.3. Age at Onset

SM is a disorder with an early mean age of onset, ranging from approximately three years of age to five or six years of age (Bergman et al, 2013; Black & Udhe, 1995; Dummit et al, 1997; Elizur & Perednik, 2003; Remschmidt et al., 2001). The upper age of onset coincides with the first year of formal schooling which varies between countries (Anderson and Thomson, 1998; Ford et al., 1998; Kumpulainen et al, 1998; Steinhausen & Juzi, 1996). Descriptive studies found that parents reported their child had always been socially reticent, suggesting an insidious onset for a proportion of the

children with SM (Anderson & Thomson, 1998; Krohn et al., 1992; Black & Udhe, 1995; Steinhausen & Juzi, 1996).

3.1.4. Referral path

Few studies included information about the average duration of mutism prior to referral. Where this information is included, it shows children are referred two to five years post onset (Black & Udhe, 1995; Bergman et al, 2013; Dummit et al, 1997; Kristensen, 2002; Krohn et al, 1992). Treatment is generally sought after a period of formal schooling (Krohn et al, 1992; Bergman et al, 2013) because the mutism severely compromises the child's participation in classroom-based group learning, thwarts assessment of academic progress and inhibits children seeking assistance when required (Cline, 2015; Ford et al., 1998).

3.2. Symptom profile

3.2.1. Speaking behaviour:

The severity of the mutism varies between children with SM. Parent and teacher reports show that children with SM vary on:

- (i) The number of contexts in which they fail to speak. Mutism is most commonly observed at school followed by the community. Children speak most freely at home when only their immediate family is present (Andersson & Thomsen, 1998; Bergman, Piacentini & McCracken, 2002; Krohn et al, 1992; Kumpulainen et al. 1998; Steinhausen & Juzi, 1996; Ford et al., 1998).
- (ii) The number of people they fail to speak with. A proportion of children are totally mute in the school environment. If they do speak at school, it is in select situations,

most often with one or two peers away from the classroom (Karakaya et al 2008; Kumpulainen et al, 1998; Sluckin et al, 1991; Steinhausen & Juzi, 1996).

- (iii) The frequency, duration and quality of speech with people with whom they do speak (e.g. parents, siblings, best friend) varies depending on the context and audience. Children often fail to speak with parents and siblings at school. If they do speak, their verbal responses are frequently restricted to single words or short phrases, and in most instances there is no or minimal spontaneous speech (Bergman, Keller, Piacentini & Bergman, 2008; Ford et al, 1998).
- (iv) The level of nonverbal responsiveness with both people with whom they do and do not speak, and the range of nonverbal responses, varies from being almost “frozen” in posture to nonverbally responsive and comfortable (Andersson & Thomsen, 1996; Kumpulainen et al.1998; Yeganeh, Beidel, Turner, Pina & Silverman, 2003).
- (v) The frequency, duration and quality of speech with immediate family at home is impacted when familiar or unfamiliar adults and children were present (Bergman et al, 2008; Ford et al., 1998; Steinhausen & Juzi, 1996).

How much children and adolescents agree with their parents’ and teachers’ reports about speaking behaviour is not known. The child and adolescent’s perspectives may differ as research shows poor to low concordance between parents, teachers and child/adolescent on ratings of internalising problems, and moderate concordance for more observable behaviours (Miller, Martinez, Shumka & Baker, 2014; Salbach, Klinkowski, Lenz & Lehmkuhl, 2009).

3.2.2. Internalising symptoms

Studies using parent-, teacher-, and child-report have consistently found that children with SM experience significantly higher levels of internalising symptoms,

general anxiety and social anxiety relative to nonclinical controls (Alyanak, Kili aslan, Harmanci, Demirkaya, Yurtbay & Vehid 2013; Bergman et al., 2002; Elizur and Perednik, 2003; Manassis, Tannock, Garland, Minde, McInnes & Clarke, 2007; Melfsen, Walitza & Warnke, 2006; Vecchio & Kearney, 2005). The overlap in the behavioural manifestation of SM to social phobia (SP) together with the identified high social anxiety in children with SM, raises questions about how children with SM and SP differ. These studies reveal that children with SM show more severe social anxiety than children with SP when rated by clinicians (Yeganeh et al., 2003; Yeganeh, Beidel & Turner, 2006; Young, Bunnell & Beidel, 2012), whereas parents report no difference in the level of social anxiety in children with SM compared to children with SP (McInnes, Fung, Manassis, Fiksenbaum & Tannock, 2004; Young et al., 2012). Researchers have questioned whether lack of speech is erroneously interpreted as higher anxiety by adult observers given the observation that some children with SM are nonverbally socially engaged and responsive (Carlson, Mitchell & Segool, 2007; Krohn, 1993; Yeganeh et al., 2003; Young et al., 2012). Interestingly, self-report ratings by children with SM show similar levels of social anxiety as reported by children with SP (Carbone et al., 2010; Manassis, Fung, Tannock, Sloman, Fiksenbaum & McInnes, 2003; McInnes et al., 2004; Yeganeh et al., 2003, 2006; Young et al., 2012) with the exception of one study that found that children with SM reported lower levels of social anxiety relative to children with SP (Melfsen et al., 2006). A methodological difficulty in the research is that measures of SP contain items related to speaking and hence will produce spurious overlap with SM. Only one study to date has excluded speech items during the analysis and shown that children with SM display more nonverbal social anxiety compared to non-clinical controls (Bergman et al. 2002). Comparison between children with SM and

SP using measures that distinguish between verbal and non-verbal social anxiety has not been conducted.

3.2.3. Externalising symptoms

Research comparing children with SM to community controls has not supported early clinical hypotheses that viewed the mutism behaviour as an expression of anger, oppositionality or hostility (Browne, Wilson & Laybourne, 1963; Mora et al., 1962; Parker et al, 1960; Pustrom & Speers, 1964). Indeed, on measures of externalizing behaviour, studies found either no difference between children with SM and community controls (Cunningham, McHolm, Boyle & Patel, 2004; Elizur and Perednik, 2003; Vecchio & Kearney, 2005), or a higher expression of externalizing behaviour in only a small percentage of the children with SM. This subgroup of children with SM who displayed higher levels of externalizing behaviour also scored highly on the internalizing measures. Clinicians and researchers have suggested that the externalising behaviour was reflective of underlying anxiety, and an attempt to avoid the anxiety-provoking social situations (Alyanak et al, 2013; Cohan et al, 2008; Kristensen, 2001; Yeganeh et al, 2006).

3.2.4. Comorbidity

For children with SM, Social Phobia or Avoidant Disorder of Childhood was the most common comorbidity, although estimates have varied widely between 18.2% and 100% (Edison, Evans, McHolm, Cunningham, Nowakowski, Boyle, Schmidt, 2011; Carbone et al., 2010; Chavira, Shipon-Blum, Hitchcock, Cohan & Stein, 2007; Dummit et al, 1997; Manassis et al., 2007; Yeganeh et al., 2006). Other common comorbidities in children with SM include Overanxious Disorder/Generalised Anxiety Disorder,

Separation Anxiety Disorder and Specific Phobia (Black & Udhe, 1995; Chavira et al, 2007; Dummit et al, 1997; Kristensen, 2000; Levin-Decanini et al, 2013). Low levels of comorbidity were found between SM and the other disorders of childhood including disruptive behaviour and mood disorders (Elizur and Perednik, 2003; Kristensen, 2001; Levin-Decanini et al, 2013; Steinhausen & Juzi, 1996).

3.2.5. Course of the disorder

As Chavira et al (2007) note, “little is known about the naturalistic course of SM” (p. 1464). In a review of available follow-up studies, Remschmidt et al. (2001) concluded that the average rate of remission of SM symptoms after a mean follow-up period of 5 years was 74%. The remission rate for children who had not received treatment was lower (Remschmidt et al, 2001). Furthermore, the children, adolescents and young people classified as being in remission (either with or without treatment) continued to speak less than average (Bergman et al, 2002). They also continued to experience higher levels of social anxiety that caused them distress and interfered with their level of functioning at school, socially and vocationally (Bergman et al, 2013; Ford et al, 1998; Oerbeck, Stein, Pripp & Kristensen, 2015; Omdal, 2007; Remschmidt et al. 2001; Sluckin et al, 1991; Steinhausen & Juzi, 1996). This finding further reinforces a link between SM and social anxiety.

4. Risk factors for Selective Mutism

Our understanding of why some children fail to speak for prolonged periods of time is in its infancy. A number of risk factors are hypothesised to contribute to the development and maintenance of SM.

4.1. Internal risk factors

4.1.1. Genetic

Childhood anxiety research clearly shows that genetic factors play a modest but significant role in the development of anxiety in children (Czajkowski, Røysamb, Reichborn-Kjennerud & Tambs, 2010; Hettame, Neale & Kendler, 2001). Consistent with these findings, clinical interviews reveal a higher prevalence of Social Anxiety Disorder and Avoidant Personality Disorder in families of children with SM (Black & Udhe, 1995; Brown & Lloyd, 1979; Kristensen, 2000; Remschmidt et al., 2001; Steinhausen & Adamek, 1997) and a higher prevalence of personality styles reflecting some degree of social fearfulness and reticence (Kristensen & Torgersen, 2001; Steinhausen & Adamek, 1997). However, these studies have mostly lacked a comparison group. In a recent study, mothers of children with SM did not differ significantly from mothers of children with mixed anxiety and non-clinical controls on self-report measures of general or social anxiety. Yet, clinicians noted that the mothers of SM children appeared anxious during the laboratory assessment with their child, suggesting they may have underreported their level of anxiety (Edison et al., 2011). Chavira et al. (2007) found that Generalised Social Phobia and Avoidant Personality Disorder was three to four fold more common among SM parents than parents of community controls, and child SM severity predicted parental Generalised Social Anxiety. However, in this study it was the fathers of children with SM who were significantly more likely to have a diagnosis of Generalised Social Phobia and Avoidant Personality Disorder than fathers of community control children, and the difference between mothers did not reach significance. Research on anxiety in pre-schoolers shows that maternal anxiety does confer a risk to the development of child anxiety (Hudson, Dodd, Lyneham & Bovopoulos, 2011). Thus, further research of parental anxiety in

children with SM is required to shed light on whether the finding of no difference between mothers of children with SM and comparison groups is due to low sample size and whether there are differences in genetic vulnerability associated with parent gender (Chavira et al., 2007).

4.1.2. Temperament

There are many similarities between the behavioural manifestation of SM and the temperament style of Behavioural Inhibition (BI) (Stein, 2001; Bergman et al, 2002; Carlson et al, 2008). BI describes a relatively stable tendency that is observed in 10 to 15 percent of young children (Kagan, Reznick & Gibbons, 1989). Preschoolers with high BI respond to unfamiliar situations, objects and people with fear, avoidance, or quiet restraint and reticence (Fox, Henderson, Marshall, Nichols & Ghera, 2005). BI is associated with an increased risk for developing an anxiety disorder (Clauss & Blackford, 2012; Rapee, Schniering & Hudson, 2009). Indirect evidence of a link between BI and SM comes from the strong resemblance between SM and BI, the descriptions of children with SM being slow to warm to new things (Ford et al. 1998) and the finding that parents of SM children rate them as displaying less sociability and higher shyness on measures of temperament than non-referred controls (Kristensen & Torgersen, 2002). Muris and Ollendick (2015) conclude in the most recent literature review on SM that “although the link between behavioural inhibition and SM seems evident, there is actually no direct empirical support for this relationship” (p. 8).

4.1.3. Speech and language skills

Numerous studies point to a higher incidence of speech and language difficulties among children with SM (Anderson & Thomsen, 1998; Black & Udhe, 1995; Ford et al,

1998; Kristensen, 2000; Kumpulainen et al, 1998; Steinhausen & Juzi, 1996) and one study demonstrated that severity of SM was associated with language performance (Manassis et al., 2007). Studies investigating the nature of these difficulties show that the receptive vocabulary skills of children with SM are within the average range however, their mean receptive vocabulary score is in the lower range of average relative to non-clinical controls (Kristensen, 2000; Manassis et al., 2003; Nowakowski et al., 2009). Expressively, children with SM give linguistically simpler and shorter narratives than children with SP (McInnes, Fung, Manassis, Fiksenbaum and Tannok, 2004). Klein, Armstrong and Shipon-Blum (2012) identified that 42% of the children with SM in their study performed at or below the fifth percentile on a measure of expressive narrative language skills. Thus, difficulties with spoken language may exacerbate the social anxiety experienced in situations where speech is anticipated and/or expected.

4.1.4. Auditory processing abilities

Researchers identified that some children with SM have abnormalities in their auditory efferent feedback pathways (Bar-Haim, Henkin, Ari-Evan-Roth, Tetin-Schneider, Hildesheimer, Muchnik, 2004). Their ability to simultaneously speak and process incoming auditory signals about quality of voice and speech is impaired, resulting in an abnormal subjective experience of their voice. Anecdotally, some children with SM have reported that their voice sounds different and strange (Henkin & Bar-Haim, 2015; Johnson & Wintgens, 2001; Muris & Ollendick, 2015). Auditory efferent abilities are further compromised at times of stress (Arie, Henkin, Lamy, Tetin-Schneider, Apter et al; 2007) and in noisier environments (Henkin & Bar-Haim; 2015). It is hypothesised that for a subgroup of children with SM, the difficulties with auditory

processing and the associated negative thoughts about the quality of one's voice may be another factor involved in the aetiology of the disorder (Henkin & Bar-Haim, 2015).

4.1.5. Social performance skills

As previously noted, children with SM vary in the degree to which they interact and communicate with same age peers (Kumpulainen et al, 1998). Furthermore, studies have found that children with SM display fewer social skills in their interactions with peers than children with a clinical anxiety disorder and non-clinical controls (Carbone et al., 2010; Yeganeh et al., 2003). Teachers report some children with SM are rejected by their classmates (Kumpulainen et al, 1998) and they experience more social difficulties than children with mixed anxiety and nonclinical controls (Levin-Decanini et al., 2013). In contrast, parents of the children with SM perceive their children have fewer social problems than did parents of children with mixed anxiety (Levin-Decanini et al., 2013). Fewer successful and positive experiences with peers may exacerbate the level of social anxiety (Rapee & Spence, 2004) and mutism. How these difficulties impact a child with SM's capacity to form and sustain friendships is not known. Studies show that children with SP experience more difficulties forming friendships and they have fewer friends (Scharfstein, Alfano, Beidel & Wong, 2011). One would anticipate that children with SM will experience similar if not greater difficulties with friendships than children with SP. This area warrants further research.

4.2. Environmental factors

4.2.1. Parent-child interaction

Parenting is one aspect of the parent-child interaction that has been shown to account for a modest proportion of the variance in childhood anxiety (McLeod, Wood & Weisz, 2007). Parents of children with SM do not differ significantly from controls on measures of discipline practice (Alyanak et al., 2013; Cunningham, McHolm, Boyle & Patel, 2004; Yeganeh et al., 2006), overprotective behaviour (Alyanak et al., 2013; Yeganeh et al., 2006), or parental acceptance (Yeganeh et al. 2006). However, McLeod and colleagues (2007) identified that questionnaire measures were less sensitive to detecting differences in parenting relative to observational studies. A recent observational study identified that mothers of children with SM displayed more parental control (i.e., granted their children less autonomy and were more directive) in comparison to mothers of children with anxiety and no-anxiety (Edison et al., 2011). Furthermore, maternal control increased the higher the observed child anxiety and the younger the child (Edison et al., 2011). This is consistent with anecdotal reports in clinical practice of parents quickly jumping in to direct their wary and reticent child to answer or participate in the activity. Replication of this finding with additional observational studies would be valuable.

4.2.2. Negative life events

Contrary to early hypotheses, most studies report that children with SM do not experience more personal, family or environmental negative life events (Black & Udhe, 1995; Brown & Lloyd, 1975; Kopp and Gilberg, 1997; Steinhausen & Juzi, 1996) however, these studies lacked a comparison group. For children who have experienced a

negative life event, there is no evidence of a temporal or causal relationship between the negative life event and the development of SM (Andersson & Thomson, 1998; Black & Udhe, 1995).

5. Behavioural/Cognitive-Behavioural treatment

A number of resource manuals and structured treatment programs for SM have been developed (Bergman, 2013; Kearney, 2010; Johnson & Wintgens, 2001; McHolm et al., 2005; Perednik, 2011). The programs share four core features: (i) the primary goal of intervention is increased speech, (ii) speech is seen as being on a continuum from non-verbal communication to speaking freely, (iii) the main intervention is exposure-based practice in feared situations in which speech is expected, and (iv) the child's school is actively involved in the treatment. Bergman et al. (2013) and Oerbeck et al. (2015) demonstrated that 50 to 67% of children with SM improved in response to a structured treatment program. The primary improvement was increased functional speech. However, despite the improvements in speaking, the majority of children in continued to experience significant levels of social anxiety (Bergman et al., 2013; Oerbeck et al., 2015).

6. Relationship of Selective Mutism to other Anxiety Disorders:

There is widespread support for the decision to reclassify SM as an Anxiety Disorder. Anxiety is a prominent feature of SM and there is a high prevalence of comorbid anxiety disorders in children with SM. Furthermore, both SM and other anxiety disorders share risk factors (genetics, behavioural inhibition) and children with SM respond to treatments that have been found to be most effective in the treatment of anxiety disorders. However, how Selective Mutism fits alongside other anxiety

disorders and in particular, its relationship to Social Anxiety Disorder is unclear. The overlap in symptomatology and commonalities in risk factors have led some researchers and clinicians to question whether SM is a variant of SP rather than a distinct anxiety disorder.

One suggestion is that SM is a developmental variant of Social Phobia (Bergman et al., 2002; Black & Udhe, 1992 & 1995; Dummitt et al., 1997). Children with SM fail to speak in situations that older children and adolescents with SP identify as the most anxiety provoking and the situations they are most likely to avoid (Rao, Beidel, Turner, Ammerman, Crosby & Sallee, 2007). The more commonly avoided situations among older children and adolescents with SP include: speaking to unfamiliar people, initiating/joining conversations, asking a teacher a question, giving oral reports/presentations, and speaking to adults. As a young child it is not possible to avoid these situations. Not speaking is a “more natural form of social avoidance for younger children” (Bögels et al., 2010: p. 178). Furthermore, a long latency to spontaneously speak is a defining feature of BI and research suggests that children with SM may be more behaviourally inhibited based on indirect evidence. Thus, not speaking is part of common social avoidance. Mutism reduces exposure to the feared social situation and attenuates the intensity of social anxiety experienced by the child with SM (Letamendi, Chavira, Hitchcock, Roesch, Shipon-Blum & Stein, (2008), Ford et al., 1998; Yeganeh et al., 2003; Young et al., 2012). However, the failure to speak draws attention to the speaking behaviour and there are spoken as well as unspoken pressures to speak. This environmental response may exacerbate the fear and avoidance of speaking. Black & Udhe (1992, 1995) have additionally suggested that SM may be a more severe presentation of SP, as children “frozen” in posture.

An alternative idea that has been proposed is that SM is a social fear that is specific to expressive speech (Omdal & Galloway, 2008). Proponents of this position note that a proportion of children with SM appear nonverbally socially engaged and confident (Carlson et al., 2007; Yeganeh et al., 2003). Furthermore, one retrospective study of adults with a history of SM found the adults did not report fear of negative evaluation in social interactions; rather their fear was specific to speaking behaviour (Omdal & Galloway, 2008). The idea that SM is a social fear that is specific to expressive speech is more consistent with the current guidelines in the DSM-5 for the differential diagnosis between SP and SM which reads: “Individuals with SM may fail to speak because of fear of negative evaluation, but they do not fear negative evaluation in social situations where no speaking is required (e.g., nonverbal play)” (APA, 2014: p. 207).

The current research evidence for children with SM does not provide a clear answer to the question of where SM sits in relation to SP. Partly because studies of social anxiety in children with SM have not assessed verbal and non-verbal social anxiety separately.

7. Summary and aims of the thesis

The initial response of mutism and wariness observed in toddlers and young children is a developmentally normal reaction in new and less familiar situations with adults and children. However, about one in every hundred children fails to speak for a number of years. The duration, severity and contexts in which the mutism occurs, together with the impairment in social and academic functioning (Wakefield, 2010), forms the criteria for a diagnosis of SM. The failure to speak is not volitional rather it is a fear response. Children with SM are more socially anxious, present with a higher

comorbidity of other anxiety disorders, and a small proportion display higher levels of externalising behaviour. The externalising behaviour is understood to be an avoidance of speaking situations. Whilst our understanding of why some children fail to speak is in its infancy, the evidence thus far clearly shows that SM is an anxiety disorder. This knowledge prompted the reclassification of SM as an anxiety disorder. The overlap in the behavioural manifestation of SM and SP has prompted questions about whether SM should be included as a distinct anxiety disorder or a variant of SP. However, there is a dearth of evidence from which to draw conclusions about how similar the presentation of SM is to SP.

This thesis sought to clarify the association between SM and SP and to address some of the identified gaps in our understanding of SM. Children aged 4 to 8 years with SM were compared to children with SP and children who were identified as “easy-going”. The child, their parents and their teacher were invited to participate in the study.

The first paper in this study pertains to the development and psychometric evaluation of the Selective Mutism Questionnaire-Child. This questionnaire is an adaption of parent-report and teacher-report measures of speaking behaviour. It is designed for children aged from 4 years. A child self-report measure will provide information about how the children see their mutism and how this compares to adult observations. As social reticence is a symptom of both SM and SP, the children’s responses will also provide information about how the children with SM and SP compare on speaking behaviour.

The second study systematically compares children with SM, SP and non-clinical controls on social anxiety symptomatology and behavioural inhibition. The primary aim of this study is to clarify the nature of the anxiety experienced by children with SM. In particular, in what ways are children with SM similar, and how do they

differ from children with SP on verbal social anxiety, non-verbal social anxiety and non-social anxiety? The study will also assess the level of behavioural inhibition in children with SM, SP and non-clinical controls to confirm whether BI is associated with SM. Finally, the study will investigate whether disinterest in social interactions is a contributing factor in observed social avoidance.

The third paper in this thesis is an exploratory study examining whether mothers and fathers of children with SM, SP and non-clinical controls differ in their levels of negative emotion, anxiety and parenting. The study will also examine whether there is an association between the child's level of anxiety and mutism and parental anxiety. The other point of interest in this study is whether there is an association between parenting and child and/or parental anxiety. These findings may provide further details about similarities and differences between children with SM and SP, and contribute to the knowledge about risk factors for the development of mutism.

Thus, the main aim of the current thesis was to systematically compare children with SM and SP. This information will clarify our understanding of the association between SM and SP. This increased understanding of the nature of the anxiety in children with SM may provide some guidance on ways to improve the effectiveness of treatment for children with SM.

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CHAPTER 2:

Paper 1

THE DEVELOPMENT AND PSYCHOMETRIC PROPERTIES OF THE SELECTIVE MUTISM QUESTIONNAIRE – CHILD (SMQ-C)

Abstract

The Selective Mutism Questionnaire (SMQ) is a psychometrically sound measure of symptoms of selective mutism completed by parents (Bergman, Keller, Piacentini & Bergman, 2008). It is supported by a parallel teacher-report measure, the School Speech Questionnaire (SSQ) (Bergman, Gonzalez, Piacentini & Keller, 2013). The current study reports the development and psychometric properties of a child-report measure to accompany these two existing scales. The Selective Mutism Questionnaire-Child (SMQ-C) provides the child's perspective on how much they talk in select situations with adults and children. It was designed for children aged from 4 years and completion of the measure does not require written or verbal responding. The SMQ-C was administered to three groups of children aged 4 to 8 years: children with Selective Mutism, children with Social Phobia and a non-clinical control group. Parents and teachers completed questionnaires on speaking, anxiety and externalising behaviour. The three-factor structure from the existing parent measure was retained. Results indicated high internal consistency for all factors and the total score of the SMQ-C. As predicted, the SMQ-C correlated moderately with parent and teacher measures of children's anxiety, and did not correlate significantly with parent and teacher reports of externalising behaviour. High correlations were found between the child SMQ-C, parent SMQ and teacher SSQ. Young children are able to provide a valid picture of their mutism behaviour, providing an additional perspective in the assessment of selective mutism and creating a clinically useful method to engage the child in assessment.

Introduction

Children with Selective Mutism (SM) fail to speak with adults and children in select situations. These children can understand and produce spoken language and they speak comfortably with their immediate family at home (APA, 2014; Muris & Ollendick, 2015). The failure to speak is generally observed at school, in the community, and in the clinical context (Bergman, Keller, Piacentini & Bergman, 2008; Bergman, Piacentini & McCracken, 2002; Black & Udhe, 1995). For a formal diagnosis of SM, the failure to speak cannot be accounted for by a speech and language disorder, lack of knowledge of the language or autism spectrum disorder. The mutism must have been present for one month (excluding the first month of attendance in an education-type setting) and be interfering with the child's functioning (APA, 2014).

Prevalence rates indicate that SM is not a "rare disorder" as previously thought (APA, 1980; Wright, Holmes, Cuccaro & Leonhardt, 1994). Epidemiological studies have reported SM prevalence rates of 0.6% - 3.3% in children in the first four years of school (Bergman et al., 2002; Browne & Lloyd, 1975; Karakaya, Sismanlar, Memik, Coskun, Agaoglu and Yavuz, 2008; Kumpulainen, Räsänen, Raaska and Somppi, 1998), 0.4% - 0.8% in preschool populations (Elizur & Perednik, 2003; Ezpeleta, de la Osa & Doménech, 2014), and 1.5% in 3 year-olds (Bufferd, Dougherty, Carlson and Klein, 2011). Speaking a primary language that is discordant with the primary language spoken in the child's community appears to be a common correlate of SM (Bergman, Gonzalez, Piacentini & Keller, 2013; Bradley & Sloman, 1975; Elizur & Perednik, 2003; Steinhausen & Juzi, 1996). Findings in regards to gender distribution have been mixed, with some studies reporting a near equal distribution between girls and boys (Bergman et al., 2002; Elizur & Perednik, 2003), while other studies have reported that

SM was more common in girls than boys (Dummit, Klein, Tancer, Asche, Martin & Fairbanks, 1997).

The dominant view that has emerged over the past two decades is that SM is an anxiety disorder (Anstendig, 1999; Bögels, Alden, Beidel, Clark, Pine et al., 2010; Muris & Ollendick, 2015). This has been exemplified through the reclassification of SM in the *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition* (DSM-5: APA, 2014) to the section on “Anxiety Disorders” from “Other Disorders of Childhood and Early Adolescents” where it had been placed when the diagnosis was first included in the *Diagnostic and Statistical Manual of Mental Disorders, Third Edition* (DSM-III) as ‘Elective Mutism’ (APA, 1980). The core diagnostic criterion for a diagnosis of SM, the absence of speech, has remained constant from DSM-III through to DSM-5. The absence of speech occurs in social situations and social anxiety is a prominent feature of the child’s presentation (Muris & Ollendick, 2015). The overlap with social anxiety indicates that in addition to failing to speak, there is a greater likelihood that children may be behaviourally more reserved with a new adult in unfamiliar environments such as the treatment clinic.

This inhibition may be further enhanced by the high prevalence of comorbid anxiety disorders amongst children with SM (Bögels et al., 2010; Muris & Ollendick, 2015). For children with SM, Social Phobia or Avoidant Disorder of Childhood is the most common comorbidity, although estimates have varied widely between 18.2% and 100% (Black & Udhe, 1995; Carbone, Schmidt, Cunningham, McHolm, Edison, St.Pierre & Boyle, 2010; Chavira, Shipon-Blum, Hitchcock, Cohan & Stein, 2007; Dummit et al., 1997; Edison, Evans, McHolm, Cunningham, Nowakowski, Boyle & Schmidt, 2011; Manassis, Tannock, Garland, Minde, McInnes & Clarke, 2007; Yeganeh, Beidel & Turner, 2006). Other common comorbidities in children with SM

include Overanxious Disorder/Generalised Anxiety Disorder, Separation Anxiety Disorder and Specific Phobia (Black & Udhe, 1995; Chavira et al., 2007; Dummit et al., 1997; Kristensen, 2000; Levin-Decanini, Connolly, Simpson, Suarez & Jacob, 2013). In contrast, comorbidity with other disorders that present in childhood is low (Black and Udhe, 1995; Cohen, Price & Stein, 2006; Elizur and Perednik, 2003; Kristensen, 2001; Levin-Decanini et al., 2013; Steinhausen & Juzi, 1996).

A critical challenge for the assessment of SM is the developmental age of the child at presentation. The failure to speak is often first noticed when a child is aged around 3 years old, which coincides with entry to an education/daycare type setting (Elizur & Perednik, 2003; McHolm, Cunningham & Vanier, 2005). Treatment is usually sought two to three years post onset (Anderson & Thomsen, 1998; Ford, Sladeczek & Carlson, 1998) at a time when children are just beginning to build their reading skills and are thus, unable to independently complete questionnaires about their experience.

Added to the challenge of trying to assess a younger child with developing reading skills, high levels of social anxiousness and inhibition as well as the possibility of a different primary language, is the evidence indicating a higher incidence of speech and language difficulties among children with SM (Alyanak, Kiliñcaslan, Harmanci, Demirkaya, Yurtbay, & Vehid, 2013; Anderson & Thomsen, 1998; Dummit et al., 1997; Elizur & Perednik, 2003; Kristensen, 2000; Manassis et al., 2007). Investigation of the nature of these difficulties suggests that the problems lie in their expressive language skills while receptive vocabulary skills appear to be within the average range (Nowakowski, Cunningham, McHolm, Evans, Edison et al., 2009; Klein, Armstrong & Shipon-Blum, 2012; Manassis, Fung, Tannock, Sloman, Fiksenbaum & McInnes, 2003). McInnes, Fung, Manassis, Fiksenbaum and Tannok (2004) compared the

expressive narrative skills of children with SM to children with Social Phobia. They found that when children with SM retold a story to their parent either at home or in the clinic, their narratives were shorter, linguistically simpler and less detailed than those given by children with Social Phobia. Similarly, Klein et al. (2012) identified that 42% of the children in their study performed at or below the fifth percentile on a measure of expressive narrative language skills. Children included in both studies had attained normal scores on measures of cognitive and receptive language skills and the second study by Klein and colleagues also excluded children who were bilingual. Hence, in addition to the impact of anxiety on a child's ability to engage in a clinical interview, a proportion of children with SM may experience difficulties finding the words to tell a narrative about their experience.

As a result of these limitations in verbal production, clinical information about the child with SM is generally obtained from the child's parents and teacher via interview and questionnaire. Questionnaires most commonly used for assessment have been measures of internalizing behaviour (e.g. Child Behavior Checklist (CBCL): Elizur & Perenik, 2003), anxiety (e.g. Multidimensional Anxiety Scale for Children (MACS): McInnes et al., 2004) and more specifically social anxiety (e.g. Social Anxiety Scale for Children–Revised (SASC-R): Bergman et al., 2002; Manassis et al., 2003). Studies of parent-child concordance on such measures have consistently shown low agreement between parents and children (Miller, Martinez, Shumka & Baker, 2014; Salbach-Andrae, Klinkowski, Lenz & Lehmkuhl, 2009). Predictors of poor parent-child concordance include younger age of the child, and measurement of internal emotional experiences rather than observable behaviours (LaGreca, 1999; Miller et al., 2014; Morris, Hirshfeld-Becker, Henin & Storch, 2004). Another predictor of poorer concordance is parent anxiety and/or depression, which has been shown to result in an

over-reporting of symptoms (Morris et al., 2004). This is noteworthy given the high incidence of social anxiety in parents of children with SM (Chavira et al. 2007). Finally, poor parent-clinician concordance has also been demonstrated in the assessment of younger children, where parents tend to report less concern for behavioural and emotional difficulties that may be considered of clinical concern (Morris et al., 2004). Given these issues, being able to obtain the child's perspective on their SM symptoms using age-appropriate clinical tools would provide a beneficial addition to clinical assessment and in research with children with SM, and it would provide an avenue to compare how the child's perspective compares to adult reports of their mutism.

There are currently no self-report measures for children to describe their frequency and reluctance to speak in different social situations. One parent-report questionnaire has begun to be more widely used to assess SM, the Selective Mutism Questionnaire (SMQ) (Bergman et al., 2002, 2008). The SMQ is designed to assess the core symptom of SM, that is, the frequency of speaking behaviour with adults and children across the three functional domains of childhood: school, home and social situations in the community. Psychometric studies of the SMQ have revealed acceptable to high internal consistency (Cronbach α .65 to .97) (Bergman et al., 2008; Letamendi, Chavira, Hitchcock, Roesch, Shipon-Blum and Stein, 2008). A three-factor solution (comprising School, Home/Family and Public/Social) best explained the data, accounting for greater than 50% of the variance (Bergman et al., 2008; Letamendi et al., 2008). The measure has good construct and incremental validity (Bergman et al., 2008; Letamendi et al., 2008). SMQ correlated significantly with related constructs, that is, parent-report measures of children's social anxiety (Bergman et al., 2008) and the clinician severity rating for SM on a semi-structured diagnostic interview (Letamendi et al., 2008). The SMQ as expected did not correlate significantly with unrelated

constructs including measures of non-social anxiety (Bergman et al., 2008). The SMQ was able to distinguish children with SM from children with Social Phobia, other anxiety disorders and non-clinical controls (Bar-Haim, Henkin, Ari-Even-Roth, Tetin-Schneider, Hildesheimer & Muchnik, 2004; Bergman et al., 2008; Manassis et al., 2003).

The “school” factor from the SMQ was modified into a teacher-report questionnaire, the School Speech Questionnaire (SSQ). The SSQ asks the child’s teacher to rate how much the child speaks with adults and children in various school situations. Preliminary analyses of the SSQ have shown acceptable internal consistency (Cronbach α of 0.94) (Bergman et al., 2002, 2013).

The present study reports on the development and psychometric evaluation of the Selective Mutism Questionnaire–Child (SMQ-C). Items from the SMQ and SSQ were incorporated and modified to form the SMQ-C. To allow very young and speech restricted children to complete the measure, the SMQ-C does not require speech, reading or written responses but is completed by the child with the clinician acting as the reader and scribe. Three groups of children were invited to participate in the current study: children with SM, children with social phobia, and a group of non-clinical controls. The SMQ-C was compared against the same (mutism), related (social anxiety, emotional distress) and unrelated constructs (hyperactivity, externalising behaviours).

Method

Participants:

Children aged 4 to 8 years were recruited via distribution of flyers and emails to child mental health services, child care services and government/private/independent

schools in the Sydney metropolitan area (38/54, 70%), direct referral from educational and health professionals (14/54, 26%), and a magazine advertisement (2/54, 4%). Children were excluded if the parent reported the child had a diagnosed developmental disorder (e.g. Intellectual Delay, Autism Spectrum Disorder) or if the child's mother could not read and understand primary school level English.

The sample consisted of 54 children aged between 4.0 and 8.1 years (See Table 1), their parents and teachers. The three groups were: children with a primary DSM-IV diagnosis of Selective Mutism (SM group, n=22), children with a primary DSM-IV diagnosis of Social Phobia (SP group, n=17), and children who were identified by their parent as "easy-going" and on assessment did not meet any DSM-IV diagnostic criteria (C group, n=15). DSM-IV criteria were used as this study commenced prior to the publication of the DSM-5 (See Table 2). For children who met DSM-IV criteria for Selective Mutism, a comorbid diagnosis of Social Phobia was given if the child displayed a clinically significant level of social anxiety and avoidance in situations where use of voice was not the sole expectation of the interaction [e.g. "working or playing with a group", "sport class", "meetings (scouts, team sports)", "musical or athletic performances", "attending parties, dances or activities"]. For children who met DSM-IV criteria for both Selective Mutism and Social Phobia, Selective Mutism was identified as the primary diagnosis as the failure to speak was associated with the greatest impairment in functioning. The sample contained more female participants (n=38, 70%) than male participants (n=16, 30%), with an average age of 72.2 months (SD = 16.3 months, range = 48-107 months) (Table 1). Two (4%) children were in a day care setting, 17 (31%) in a preschool program, 12 (22%) in Kindergarten (the first year of formal schooling in New South Wales), and 23 (43%) were in primary school (grade 1 and above). The ethnic composition of the sample was: 42.5% Oceanian or

North West European (where English is the primary language), 20.4% South/Eastern European, 14.8% East Asian, 11.1% South/Central Asian, 7.4% Middle Eastern and 3.8% another non-English speaking country.

Table 1: *Demographic Characteristics of the children in the study.*

Demographic variable	SM (n = 22)	SP (n = 17)	C (n = 15)	Total (n = 54)
Children's age (mths) [mean (SD)]	70.5 (14.0) _{ab}	80.0(17.8) _a	65.9(15.01) _b	72.2 (16.3)
Child Gender: Female [% (n)]	81.8% (18)	52.9% (9)	73.3% (11)	70.4% (38)
Primary language not English [% (n)]	31.8% (7)	11.8% (2)	0% (0)	16.77% (9)
PPVT-IV standard score (SD)	96.4 (11.0) _a	105.2 (13.7) _{ab}	114.1 (14.9) _b	104.1 (14.7)
Speech Therapy Intervention (n)	6	2	0	8

Note: Means sharing subscripts are not significantly different ($p > 0.05$).

Table 2: *Primary and comorbid diagnoses in the two clinical groups, SM and SP.*

	SM (n = 22)	SP (n = 17)
Comorbid Disorder(s) [Mean number (SD)]	1.68 (0.10)	0.94 (1.02)
Social Phobia [n (%)]	20 (91%)	-
Separation Anxiety Disorder [n (%)]	6 (24%)	4 (24%)
Generalised Anxiety Disorder [n (%)]	5 (20%)	7 (41%)
Specific Phobia [n (%)]	4 (18%)	3 (18%)
Selective Mutism (Prior History) [n (%)]	-	3 (18%)
Attention-Deficit Hyperactivity Disorder [n (%)]	0 (0%)	1 (6%)
Oppositional Defiant Disorder [n (%)]	1 (4%)	0 (0%)
Sleep Disorder NOS [n (%)]	0 (0%)	1 (6%)

All mothers (mean age = 38.2 years, $SD = 9.64$) of the 54 children and 50 fathers (93%, mean age = 39.1 years, $SD = 5.41$) participated in the study. Fifty-one (94%) children lived with both biological parents, who were in a married ($n = 46$, 85.1%) or de-facto ($n = 5$, 9.3%) relationship. Ninety-two percent of families (47 of 51 families who disclosed) identified middle to above average family income. Eighty-five percent of mothers and fathers had completed further education post high-school. Most mothers in this study were either employed part-time ($n=27$, 50%) or were at home by choice ($n=16$, 29.6%), while most fathers were in full-time employment ($n = 50$, 94.3%).

Fifty-two (96%) teachers participated in the study. They had taught the identified child for an average of nine months ($SD = 7.8$ months).

Measures:

(a) Child report

Development of the SMQ-C: Psychometrically sound measures that had been developed for use with children from age four years were identified and reviewed (Dubi, Lavalle & Schneider, 2012; French & Mantzicopoulos, 2007; Harter & Pike, 1984; and Muris, Meesters, Mayer, Bogie, Luijten, et al., 2003). Commonalities were found in the structure and format of these measures including:

- (i) The clinician administered the questionnaire individually.
- (ii) Each item was matched with a colour picture that represented the context of the item (question). The picture helped hold the child's attention and interest, and it facilitated understanding of language.
- (iii) A two-step response was required for each item with a pictorial representation provided for each step. The child was first asked a binary choice question (e.g.

yes or no). The child was then asked to quantify how much the item matched their experience using a pictorial Likert scale that had two to three levels.

These commonalities were included in the development of the SMQ-C as they corresponded to the first goal of having a measure that did not require speech, reading or a written respond.

The SMQ-C contained 20 items. The first 17 items of the SMQ and three items that had subsequently been added to the SSQ that asked about nonverbal communication in the school context were included. The word “talk/speak” was modified to “talk with your voice” and “talk with your body” (with the clinician modelling head and hand gestures when saying talk with your body) to differentiate more clearly the different ways of communicating. The wording of each item was scripted into a two-part question, and the child responded by pointing to a pictorial response scale. The pictorial response scale was two-sided. On the first side were pictures for “yes” or “no” and on the opposite side there was a three-point pictorial scale from “a little”, “a lot” to “always”. The response wording was consistent with the SMQ and SSQ scales.

Prior to the administration of the SMQ-C, three to five practice items about the child’s likes and dislikes were asked to train the child in the use of the 2-stage response and the 3-point pictorial Likert scale. After the child demonstrated an understanding of the response procedure, the SMQ-C items were administered in the same order for each child. There was a pictorial representation for each item. The picture was placed on the table. For some items there was a simple statement the assessor said aloud as they placed the picture on the table (eg. item 14, “*Your parents have friends that you see a lot and you know them well*”). Once the picture was placed on the table, the assessor asked the child the first part of the question: “Do you ...(*talk with your voice to your*

parents friends that you know well, item 14)?”. If the child indicated a “yes”, the second part of the question was asked: “How much do you...(*talk with your voice to your parent’s friends that you know well, item 14)?*”. If the child indicated “no”, the assessor asked the next item. Two items in the SMQ contained the response option of “non applicable” (ie. item 15, babysitter, and item 20, extra curricular activities), and this response option was retained in the SMQ-C by asking the child before presenting the item whether they had a babysitter or engaged in extracurricular activities. The responses were converted to a 4 point scale from 0 (no talk) to 3 (always).

The study did not have an adequate number of child participants to conduct factor analyses. Thus, the three factor structure of the SMQ: school, home/family and public/social was retained in the scoring of the SMQ-C. A mean score was calculated for each factor, and the mean of the factor scores formed the total score. Lower scores on the SMQ-C reflected less talking behaviour.

The *Peabody Picture Vocabulary Test, Fourth Edition (PPVT-IV)* (Dunn and Dunn, 2007), is a nonverbal measure of receptive vocabulary skills for children aged from two years and six months. The PPVT-IV was administered prior to the SMQ-C as it has been identified as a useful screener for vocabulary development and for detecting language impairments (Dunn & Dunn, 2007). The children’s PPVT-IV Standard Score was used for comparison.

(b) Parent report

The *Selective Mutism Questionnaire (SMQ)* (Bergman et al., 2002, 2008) consists of 23 items, with the first 17 items assessing frequency of speech in the preceding month across three factors: (i) School, (ii) Home/Family, and (iii) Public/Social. The final 6 items measure level of interference/distress arising from the

speech failure and these responses were not included in the total score nor considered in this study. For the first 17 items, parents rated frequency of speech from 0 (never) to 3 (always). The mean score for the three factors and the total score were included in analyses. As with the SMQ-C, a lower score indicated less speaking behaviour. The internal consistency of the three factors and total score remained high in this study (Cronbach α : SDQ School: .96 for mothers and .95 fathers, SDQ Home/Family .82 for mothers and .88 for fathers, SDQ Public .95 for fathers and .93 for mothers; and SDQ Total .96 for mothers and .97 for fathers).

The *Family Demographic Questionnaire* consisted of 34 questions that collected demographic data on the child and their immediate family. The child's mother completed the questionnaire.

The *Anxiety Disorders Interview Schedule for DSM-IV: Child Version, Parent Interview Schedule (ADIS:C/P)* (Silverman & Albano, 1996) is a methodologically sound and widely used semi-structured interview that assesses for the presence of the major DSM-IV anxiety, mood and externalising disorders experienced by children and adolescents. The interview was conducted with the child's mother, either in person (n=25, 46%) or by telephone (n=29, 54%). Previous studies have found the ADIS:C/P can be used reliably as a diagnostic tool with parents of preschoolers (Edwards, Rapee, Kennedy & Spence, 2010; Hudson, Dodd, Lyneham & Bovopoulos, 2011; Rapee, 2014) and it can be administered by telephone (Lyneham & Rapee, 2005; Letamendi et al., 2008). The first author, an experienced registered clinical psychologist, conducted the interviews. All interviews were audio recorded. Clinical Severity Ratings (CSRs) on a scale of 0 to 8 were assigned based on number of settings, severity of symptoms and impact on functioning. A diagnosis was considered "clinical" if the CSR was four or greater. Nineteen percent (10/54) of the interviews were coded for inter-rater reliability

by a second rater trained in the ADIS:C/P and blind to group status. Interrater agreement was as follows: primary diagnosis ($\kappa = 1.00$) and secondary diagnosis ($\kappa = 1.00$). Intraclass correlations for the clinician severity rating were: primary diagnosis ($ICC = .97$) and secondary diagnosis ($ICC = .75$).

Social Anxiety Scale for Children – Revised/Parent Version (SASC-R/P) (La Greca, 1999) is a 22 item parent measure of the child's social anxiety in their relationship with peers. Eighteen of the items group to form three subscales: SASC—R/P:New (social avoidance and distress with new and less familiar social people), SASC-R/P:FNE (fear of negative evaluation or response by peers) and SASC-R/P:General (social avoidance and distress generally when interacting with others). The parent rates how true each statement is for their child on a 5-point Likert scale from 1 (not at all) to 5 (all the time). Some items in the SASC-R/P:New and SASC-R/P:General asked about speaking. The SASC-R/P:New was calculated with and without these items. The SASC-R/P:General subscales could not be calculated without speaking items as only one item remained in the subscale. The SASC-R/P measure has been shown to have good internal consistency (SASC-R/P New $\alpha = .87$, SASC-R/P FNE $\alpha = .91$, SASC-R/P General $\alpha = .78$) and moderate convergent validity (La Greca, 1999). In the current sample, Cronbach α for the SASC-R/P-New(ns, no speech) was .85 for mothers and .89 for fathers, and for the SASC-R/P-FNE was .94 for mothers and fathers.

The *Strengths and Difficulties Questionnaire (SDQ)* (Goodman, 1997) is a 25 item measure screening for behavioural and emotional problems in children aged 4 to 16 years. Three of the five subscales: Emotion Symptoms, Conduct Problems and Hyperactivity/Inattention were included in the study. Studies of Australian children show that while there are questions about the factor structure of the SDQ (Mellor &

Stokes, 2007) it has sound psychometric properties (Hawes & Dadds, 2004) and is an effective screening instrument (Mathai, Anderson and Bourne, 2004; Mellor & Stokes, 2007). The Cronbach α 's in this study were as follows: emotion symptoms - .77 for mothers and .73 for fathers, conduct problems .66 for mothers and .62 for fathers, and hyperactivity/inattention .75 for mothers and .77 for fathers.

(c) Teacher report

Teachers completed a brief Background Information form asking about the structure of the learning environment, and the length of time (in months) they had (a) known and (b) taught the child. The teachers then completed the following three questionnaires.

School Speech Questionnaire (SSQ) (Bergman et al., 2002) is a 7 item measure. The first six items ask teachers to rate frequency of speaking with children and adults in the most common social situations at school and the seventh item asked how much the failure to speak interferes with functioning. Teachers rated the first six items on a four-point Likert scale from 0 (never) to 3 (always). The internal consistency in this study of the 6 items measuring speaking was $\alpha = .95$ with all item-total correlations .80 or above.

Child Behavior Scale (CBS) (Ladd, 2010) is a 59 item teacher-report measure of the child's behaviour and relationship with peers in school contexts. There are six subscales of which three were included in this study: the internalizing subscale (Anxious-Fearful) and two externalizing subscales (Aggressive with Peers and Hyperactivity-Distractibility). Teacher's rated how applicable the description for each item was to the child on a 3-point Likert scale from 1 (Doesn't apply, seldom displays this behaviour) to 3 (Certainly applies, often displays this behaviour). The measure has been shown to be internally consistent and relatively stable across time with acceptable

validity when used with young children from age 5 (Ladd, 2010). Contact was made with the author of the questionnaire to confirm appropriateness for use with children aged 4 years. The internal consistency of the three subscales of the CBS remained good in this study (Cronbach α : Aggressive to peers $\alpha = .74$, Hyperactive Distractible $\alpha = .82$, and Anxious-Fearful $\alpha = .78$).

Teacher Rating Scale for Social Anxiety – “junior” (TRSSA) (Bokhorst, Goosens & de Ruyter, 2001) is a 9 item scale asking teachers to rate the level of observed social anxiety or failure to socially participate in school social contexts. Teachers rated each item on a 5-point Likert scale from 0 (not at all/never) to 5 (very/always). A higher total score indicated more social anxiety. The scale has demonstrated high internal consistency ($\alpha = .87$ to $.90$) and good convergent and divergent validity when used with children aged 5 to 6 years of age (BokHorst et al., 2001). Internal consistency of the scale ($\alpha = .92$, with item-total correlations of $.48$ - $.86$) was moderate to high in this study.

(d) Child Observation

Children participated in a laboratory observation that included a Verbal Social Interaction Task (Asendorpf, 1990; Kennedy, Rapee & Edwards, 2009). The child was asked a series of questions beginning with closed questions related to novel toys on the table in front of the child and followed by open questions about the child (e.g. age, likes). Quantitative data were collected on: latency to say the first spoken word, total talk time, latency to the first nonverbal response and number of nonverbal responses. The videos were coded by the first author. A second rater, who was blind to group membership, independently scored a random 30% ($n = 19$) of the assessments. Inter-rater correlations for all the variables was high ($r = .97$ to 1.0).

Procedure

Ethics approval for this study was granted by the Health Department (Western Sydney Local Health Network), Education (NSW Department of Education and Communities, and Catholic Education Diocese of Parramatta) and Macquarie University Human Research Ethics Committee. The parents provided informed written consent for their child's and their own involvement in the study. Each parent completed a package of questionnaires and then the child's mother participated in the semi-structured diagnostic interview. Following this, the child and their mother attended the clinic for a Behavioural Observation interview during which time the child completed the self-report questionnaires with the clinician. Once the family had completed their role in the study, the child's (pre)school was invited to participate. The child's teacher completed questionnaires and provided informed written consent for their involvement in the study.

Families in this study were part of a broader study of selective mutism. Parents of children in the SM and SP groups received a clinical report with recommendations for support services. As the clinical report was not relevant to children in the non-clinical control group, families in the non-clinical control group received a shopping voucher as reimbursement for their time.

Statistical Analyses

Internal consistency of the SMQ-C was evaluated using the Cronbach's coefficient alpha and the descriptive statistics of the individual items of the SMQ-C are reported. There was a small percentage of missing data for parents due to item non-response, and where the number of participants varied this is noted in the results table. Pearson's product-moment correlation coefficient was used to measure the strength of

the relationship between the SMQ-C and measures of other constructs. Given the number of analyses, the Type-1 error rate for significance was corrected by applying the Bonferroni adjustment to each correlation matrix and the critical alpha is noted. Analysis of Covariance was used to determine whether SMQ-C could differentiate between the groups. Age and the PPVT-IV standard score were included as covariates in these analyses. Where a statistical difference was found between groups, pairwise comparisons were conducted using the estimated marginal means and the Bonferroni adjustment for multiple comparisons.

Results

Demographic characteristics of the sample:

There was a statistically significant difference between groups on age, $F(2,51) = 3.50$, $p = 0.04$, with the mean age of children in the SP group being just over one year greater than for children in the C group (see Table 1). Gender distribution did not differ significantly between groups, $X^2(1, N = 54) = 0.56$, *ns*.

In terms of language skills, 81% ($n=44$) of the families identified English as the primary language spoken at home with the child. A statistically significant difference was found between groups on English as the primary language spoken at home, $X^2(1, N = 54) = 6.50$, $p = 0.03$. For 31.8% of the children in the SM group and 11.8% of the children in the SP group English was not the primary language spoken at home. English was the primary language spoken at home for all the children in the non-clinical group. The PPVT-IV standard score for 93% ($n=50$) of the children fell within the Average or Moderately High Range. However, a statistically significant difference was found

between groups on the PPVT-IV standard score, $F(2,51) = 8.22$, $p = 0.001$, with the mean PPVT-IV standard score for children in the SM group being in the lower range of Average compared to the C group who performed closer to the Moderately High range (see Table 1). Eight children in the study (15%) had previous speech therapy involvement and most (75%) were from the SM group.

Internal consistency and item analysis:

On the SMQ-C, 23 of the children (42.6%) had missing data rated “not appropriate” on item 15 (speaking with the babysitter) and/or item 20 (extra-curricular activities) and one participant had only completed the school subscale. For the 30 participants who had full data on all 20 items, the SMQ-C total score showed good internal consistency (Cronbach’s $\alpha = .89$) and removal of any item did not increase the alpha. When items 15 and 20 were excluded from the analysis, the internal consistency remained consistent ($n=53$, Cronbach’s $\alpha = .88$).

The internal consistency of each factor was also calculated. Two factors, School and Public/Social, showed good internal consistency [Cronbach’s α for school ($n = 54$) was .82, Public/Social ($n=45$) was .80], whereas the internal consistency of the Home/Family factor was lower ($n=37$, Cronbach’s $\alpha = .67$). Internal consistency decreased when the items rated as “not applicable” were excluded. Exclusion of item 15 resulted in the Home/Family ($n = 53$) Cronbach α dropping to .61 and exclusion of item 20 from the Public/Social factor ($n = 53$) resulted in a Cronbach α of .71. Thus, even though there were children for whom items 15 and 20 were not applicable, for those children where these items were relevant, the items were internally consistent and contributed to the reliability of the scale.

Descriptive statistics showed that the individual items possessed sufficient variability to discriminate among children (see Table 3). Skewness varied between -1.25 and 1.39 , and kurtosis of individual items varied between -1.64 and 0.31 across items.

Table 3: *Item responses on the SMQ-C*

Factor	Item	Mean	Skewness	Kurtosis	Endorsed “Never” on item		
		(SD)	(SD)	(SD)	SM (%)	SP (%)	C (%)
School (n = 54)	1. Most peers at school.	1.57 (1.25)	-0.15 (0.33)	-1.64 (0.64)	63.6	17.6	0.0
	2. Selected peers at school.	2.04 (1.17)	-0.75 (0.33)	-1.02 (0.64)	36.4	5.9	0.0
	3. Answers teacher’s question.	1.46 (1.17)	0.20 (0.33)	-1.50 (0.64)	68.2	5.9	0.0
	4. Asks teacher question.	1.35 (1.21)	0.20 (0.33)	-1.55 (0.64)	77.3	11.8	0.0
	5. Most teachers/adults at school.	0.69 (1.06)	1.26 (0.33)	0.13 (0.64)	90.9	64.7	26.7
	6. Groups or in front of class.	1.22 (1.11)	0.40 (0.33)	-1.18 (0.64)	72.7	5.9	6.7
	7. Joins in nonverbally in class group activities.	1.57 (1.20)	-0.05 (0.33)	-1.47 (0.64)	38.1	29.4	0.0
	8. Answers teacher questions with gesture.	2.19 (1.02)	-0.85 (0.33)	-0.66 (0.64)	14.3	5.9	0.0
	9. Joins in nonverbally in class performances.	1.62 (1.21)	-0.30 (0.33)	-1.61 (0.64)	28.6	35.3	0.0
Home/ Family (n=53)	10. With immediate family at home when others present.	2.06 (1.08)	-0.78 (0.33)	-0.74 (0.64)	28.6	5.9	0.0
	11. With immediate family in unfamiliar places.	2.15 (0.97)	-0.84 (0.33)	-0.38 (0.64)	14.3	0.0	6.7
	12. With extended family not living at home.	2.34 (0.98)	-1.25 (0.33)	0.31 (0.64)	14.3	5.9	0.0
	13. With immediate family on the phone.	1.92 (1.21)	-0.60 (0.33)	-1.25 (0.64)	33.3	17.6	6.7
	14. With familiar family friends.	1.60 (1.25)	0.00 (0.33)	-1.68 (0.64)	42.9	23.5	0.0
	*15. With babysitter.	2.03 (1.04)	-0.68 (0.39)	-0.76 (0.76)	26.7	0.0	0.0
Public/ Social (n=53)	16. Unfamiliar peers.	0.68 (1.11)	1.39 (0.33)	0.37 (0.64)	85.7	76.5	26.7
	17. With unfamiliar family friends.	0.75 (1.05)	1.23 (0.33)	0.22 (0.64)	76.2	64.7	20
	18. With doctor and/or dentist.	0.87 (1.00)	0.87 (0.33)	-0.40 (0.64)	81.0	47.1	0.0
	19. With adults in shops and cafes.	0.75 (1.02)	1.20 (0.33)	0.28 (0.64)	71.4	58.8	26.7
	**20. With others in sports/activities outside (pre)school.	1.47 (1.22)	0.12 (0.35)	-1.57 (0.70)	73.3	13.3	0.0

Note: * n=37, ** n = 45.

Convergent validity

Pearsons Product-moment correlation coefficients were generated to assess the convergent validity of the SMQ-C with parent-report and teacher-report measures of the same construct (mutism) (Table 4) and related constructs (social anxiety and emotional distress) (Table 5).

Table 4: *Correlations between the SMQ-C, mother's SMQ, father's SMQ and SSQ.*

Measure	1	2	3	4	5	6	7.	8	9	10	11	12
SMQ-C School	-											
SMQ-C Home	.62**											
SMQ-C Public	.72**	.57**										
SMQ-C Total	.90**	.83**	.89**									
SMQ School (m)	.75**	.47*	.63**	.71**								
SMQ Home (m)	.46*	.41*	.51**	.52**	.62**							
SMQ Public (m)	.58**	.41*	.62**	.61**	.75**	.65**						
SMQ Total (m)	.70**	.49**	.67**	.70**	.92**	.82**	.91**					
SMQ School (f)	.70**	.39*	.59**	.64**	.89**	.60**	.67**	.83**				
SMQ Home (f)	.34*	.44*	.39*	.44*	.51**	.66**	.52**	.62**	.52**			
SMQ Public (f)	.66**	.45*	.75**	.70**	.71**	.54**	.76**	.77**	.76**	.60**		
SMQ Total (f)	.67**	.49*	.68**	.69**	.83**	.67**	.76**	.86**	.90**	.78**	.92**	
SSQ Total	.75**	.50**	.62**	.70**	.87**	.52**	.70**	.81**	.84**	.45*	.68**	.78**

Note: * $p < 0.01$; ** $p < 0.001$. Critical alpha set at $p < 0.001$ following Bonferroni adjustment.

(m) = mother, (f) = father

1 = SMQ-C School, 2 = SMQ-C Home, 3 = SMQ-C Public, 4 = SMQ-C Total, 5 = SMQ School (m), 6 = SMQ Home (m), 7 = SMQ Public (m), 8 = SMQ Total (m), 9 = SMQ School (f), 10 = SMQ Home (f), 11 = SMQ Public (f), 12 = SMQ Total (f)

On measures of mutism, statistically significant positive correlations were found between the child's, mother's and father's responses on the School factor, Public/Social factor and the Total scores (range $r = .54$ to $.89$, $p < 0.001$) (see Table 4). This was observed across informants and between the factor and total scores. A statistically significant positive correlation was also found between the teacher's SSQ Total score and child's School factor score ($r = .75$, $p < 0.001$) and the teacher's SSQ Total and the parents' School factor scores (mother $r = .87$, $p < 0.001$ and father $r = .84$, $p < 0.001$). Overall, high cross-informant correlations were found on frequency of talking on the School factor, Public/Social factor and Total score. With regard to the SMQ and SMQ-C Home/Family factor, moderate positive correlations (approaching statistical significance) were found between the child's score and mother's ($r = .41$, $p = 0.002$) and father's ($r = .44$, $p = 0.002$) responses.

Table 5: *Correlations between SMQ-C and parent- and teacher-report on related constructs.*

Respondent	Measure	SMQ-C School	SMQ-C Home/Family	SMQ-C Public/Social	SMQ-C Total
Mother	SASC-R/P New	-.42*	-.28	-.48*	-.46*
	SASC-R/P New (ns)	-.40*	-.30	-.45*	-.45*
	SASC-R/P General	-.48**	-.30	-.52**	-.50**
	SASC-R/P FNE	-.22	-.16	-.21	-.23
	SDQ Emotion Symptoms	-.30	-.21	-.28	-.31
Father	SASC-R/P New	-.56**	-.37	-.55**	-.57**
	SASC-R/P New (ns)	-.55**	-.36	-.53**	-.55**
	SASC-R/P General	-.44*	-.36	-.50*	-.50*
	SASC-R/P FNE	-.28	-.25	-.38	-.35
	SDQ Emotion Symptoms	-.23	-.33	-.32	-.34
Teacher	TRSSA	-.45*	-.30	-.40*	-.44*
	CBS Anxious/Fearful	-.26	-.15	-.25	-.25

Note: * $p < .01$; ** $p < .001$. Critical alpha set at $p < 0.001$ after the Bonferroni adjustment.

On measures of social anxiety, moderate negative correlations were found between the children's SMQ-C School, Public/Social and Total scores and the parent responses on measures of social avoidance (see Table 5). More specifically, the correlation for mother's and father's responses on the SASC-R/P-General were statistically significant however, this variable contained items that included a speaking component. For the SASC-R/P-New, where the items that included a speaking component could be removed, the correlations remained high and approached significance. Moderate negative correlations approaching significance were also found between the children's SMQ-C School, Public/Social and Total scores and the teacher's measure of social avoidance (TRSSA). The SMQ-C Home/Family factor again performed differently with lower moderate correlations with both parent and teacher measures of social avoidance. The parent measure of the cognitive aspect of social anxiety (SASC-R/P-FNE) and the general measures of anxiety (SDQ-Emotion Symptoms subscale and CBS Anxious/Fearful subscale) showed low to moderate negative correlations with the children's SMQ-C factor and Total scores.

In the Verbal Social Interaction Task (Table 6), an observational measure of speech with an unfamiliar person, moderate to high statistically significant negative correlations were found between the all the SMQ-C scores and duration of time it took the child to begin speaking with the clinician. The total duration of time the child spoke with the clinician shared a positive statistically significant correlation with the SMQ-C School and Public/Social factors as expected.

Table 6: *Correlations between SMQ-C and Verbal Social Interaction Task.*

Verbal Social Interaction Task	SMQ-C School	SMQ-C Home/Family	SMQ-C Public/Social	SMQ-C Total
Latency to say first spoken word	-.70**	-.47*	-.57**	-.65**
Total duration of child's speech	.56**	.34	.58**	.57**
Latency to first nonverbal gesture	-.28	-.29	-.27	-.30
Number of nonverbal gestures	-.09	-.17	-.11	-.15

Note: * $p < .01$; ** $p < .001$. Critical alpha set at $p < 0.01$ based on the Bonferroni adjustment

Discriminant Validity:

Discriminant validity of the SMQ-CA/A was assessed via correlations with parent-report and teacher-report measures of Hyperactive/Distractible and Externalising behaviours. As can be seen in Table 7, all correlations were low and non-significant as expected.

Table 7: *Correlations between SMQ-C and parent- and teacher-report on externalising behaviour measures.*

Respondent	Measure	SMQ-C School	SMQ-C Home/Family	SMQ-C Public/Social	SMQ-C Total
Mother	SDQ Conduct	-.16	-.04	.04	-.06
	SDQ Hyperactive/Inattentive	.13	.11	.25	.19
Father	SDQ Conduct	-.11	-.23	.04	-.11
	SDQ Hyperactive/Inattentive	.03	.03	.09	.05
Teacher	CBS Aggressive	.02	.18	.07	.09
	CBS Hyperactive/distractible	.06	.17	.02	.09

Note: * $p < .01$; ** $p < .001$. Critical alpha set at $p < 0.003$ following the Bonferroni adjustment.

Sensitivity of the SMQ-C:

As seen in Table 8, a statistically significant difference was found between the three groups on the SMQ-C Total Score and the SMQ-C School and Public/Social factors. On the SMQ-C School factor, the SM group communicated less than the SP group and both groups communicated less than the C group. On the SMQ-C Total score and SMQ-C Public/Social factor, the SM and SP groups communicated less than the non-clinical group, and the two clinical groups did not differ significantly. The largest effects were shown on the school and public factors and the smallest effect size was shown on the home factor.

Table 8: Comparison between groups on SMQ-C Total and Factor scores.

Domain	SM group (n=21) Estimated Mean (SE)	SP group (n = 17) Estimated Mean (SE)	C Group (n = 15) Estimated Mean (SE)	<i>F</i> value	$p\eta^2$
Total	1.04 (.11) _a	1.51 (.12) _a	2.07 (.13) _b	$F(2,53) = 15.84, p < 0.001$.398
School	0.98 (.12) _a *	1.59 (.13) _b	2.24 (.15) _c	$F(2,54) = 18.48, p < 0.001$.435
Home/Family	1.70 (.15)	2.10 (.16)	2.33 (.19)	$F(2,53) = 3.327, p = ns.$.122
Public/Social	0.39 (.12) _a	0.81 (.13) _a	1.62 (.15) _b	$F(2,53) = 18.42, p < 0.001$.439

Note: Means sharing subscripts are not significantly different.

* $n = 22$.

Discussion

The purpose of this study was to develop a scale that could be completed by children where the primary reason for referral to a mental health professional is the failure to speak in select situations, the core symptom of SM. These children fail to

respond with voice to direct questions in the clinical interview and the degree of nonverbal responsiveness varies greatly (Anstendig, 1999; Cohan et al., 2006). The primary sources of information about how much the children speak in various social situations are usually their parents and teacher and research and previous research has demonstrated low to moderate concordance between child and the adult ratings of the child's anxiety (Miller et al., 2014). A valid child measure would provide a clinical tool that the mental health professional could use to gain the child's perspective on how much they speak at home, school and in the community. The measure needed to be suitable for young children as treatment for children with SM is usually sought in their early school years (McHolm et al., 2005; Muris & Ollendick, 2015), a time when they are just beginning to develop their reading and writing skills. The SMQ-C was modelled on measures that had been developed for children from age 4 years that did not require speech, reading or writing (Dubi et al., 2012; French & Mantzicopoulos, 2007; Harter & Pike, 1984; and Muris et al., 2003). The SMQ-C was adapted from the published parent-measure, the SMQ, and the teacher measure, the SSQ. The factor structure and scoring method of the SMQ were retained as the small sample size precluded more complex statistical analyses. The psychometric properties of the SMQ-C were evaluated using two clinical samples, children with SM and children with SP, and a community sample of non-clinical children.

High internal consistency was found for two of the three factors (School and Public/Social) and for the Total score, and acceptable internal consistency for the third factor (Home/ Family). There was considerable evidence supporting the construct validity of the SMQ-C. High child-parent-teacher concordance was found between the SMQ-C, SMQ and SSQ. More specifically, inter-informant concordance for speaking at school, in public social situations and total talking was very high. Moderately high

parent-child concordance was found on frequency of talking in the Home/Family situations. Furthermore, speaking is an observable, quantifiable behaviour and the correlation between the Verbal Social Interactional Task and the SMQ-C were moderate to high. This provides the strongest evidence of the real validity of the SMQ-C.

The construct validity of the SMQ-C was further demonstrated as the relationship between the SMQ-C and related constructs was in the expected range. In general, significant negative relationships were shown between the SMQ-C and measures of observable verbal and nonverbal social avoidance behaviour (SASCR-R/P:New, SASC-R/P:New (ns), SASCR-R/P:General, TRSSA). Moderate non-significant correlations were found with measures of less observable and internal qualities of anxiety, the fear of negative evaluation (SASC:Fear of Negative Evaluation) and general symptoms of negative emotion/anxiety (SDQ Emotional symptoms and CBS Anxious/Fearful). Thus, children who spoke less were observed to display higher levels of social anxiety. This pattern is consistent with the dominant thinking that SM is an anxiety disorder that is most strongly associated with social anxiety (Bögels et al., 2010; Muris and Ollendick, 2015). In contrast, consistent with data showing a lack of externalising disorders among children with SM (Cohan et al., 2006; Cunningham et al. 2004; Elizur & Perednik, 2003), low, non-significant correlations were found between the SMQ-C and parent and teacher reports on these unrelated constructs of Hyperactivity/Inattentiveness and Disruptive behaviour.

Mutism behaviour is most commonly observed at school (Kumpulainen et al., 1998; Steinhausen & Juzi, 1996) and mutism in the school setting is identified as the best predictor of SM (Letamendi et al., 2008). Consistent with this finding, the largest effect size difference between groups appeared to be on the SMQ-C School factor. The SMQ-C scores for speaking at school were significantly lower among children in the

SM group than children in the SP group and both groups spoke less than the non-clinical controls. A fundamental difficulty within research with SM is the high comorbidity of SP among children with SM (Muris & Ollendick, 2015). In this study the majority of children with SM had a comorbid diagnosis of SP. The SMQ-C was able to differentiate between children with SM who had a comorbid SP diagnosis and children with SP without a current comorbid diagnosis of SM. Hence the SMQ-C subscale may assist clinical decision making about whether a child with SP also meets criteria for a comorbid diagnosis of SM. Furthermore, the School subscale may provide the best option for a brief screening measure in future SM research. The second subscale, Public/Social subscale, was able to discriminate effectively between children in the SM or SP group and non-clinical controls however, the SMQ-C did not differentiate between the two social anxiety based groups. The Public/Social items assessed amount of speaking with new and less familiar adults and children, a situation where children with SP are also highly likely to experience distress and subsequent restricted speech (Heiser, Turner, Beidel & Roberson-Nay, 2009; Rao, Beidel, Turner, Ammerman, Crosby & Sallee, 2015). More than half the children with SP endorsed they “never” talked in the majority of situations that formed the Public/Social subscale. One might predict that with time the child with SP will begin to use their voice in these situations in contrast to the child with SM however, the SMQ-C does not measure duration of mutism. Given children with SP do fail to speak in these situations, the SMQ-C may be a useful clinical measure when assessing impact of social anxiety on the child’s functioning. The third subscale of the SMQ-C, the Home/Family subscale, was not sensitive enough to differentiate between the groups. The subscale measures amount of speaking with immediate family in specific situations as well as speaking with familiar family and friends. One may expect that responding verbally to a parent in

the presence of others is less anxiety provoking than speaking directly to a teacher, classmate or less familiar non-family member (Bergman et al., 2002, 2008; Rao et al., 2015). Thus, this subscale is more helpful in clinical practice as it provides information about the severity of mutism.

Some limitations of the present study should be noted. The main limitation was the small sample size, which did not allow investigation of the dimensionality of the measure. The factor structure and scoring process of the parent-report SMQ was applied to the child SMQ-C. The assumption that the measures would share the same factor structure may not be valid, although the internal consistencies and other psychometric properties were found to be strong. Future studies with larger samples are required to determine whether the current three-factor solution from the parent measure provides the best factor structure for the child measure. Future studies should also examine whether all the original items of the SMQ-C need to be retained. In this study, all the items were retained. The children are aware of their failure to speak and the range of items provided a way of capturing how they saw their speaking behaviour. A 20-item questionnaire can use considerable resources and thus, a briefer assessment tool would be valuable, especially for research and screening purposes.

SM is a social anxiety based disorder that presents in the early preschool years and significantly impacts on a child's ability to join into play and learning. Being able to differentiate between normal developmental shyness and SM is important to ensure early identification and referral for treatment. The SMQ-C is the first available self-report measure of frequency of speech. Its strength is that it can be completed by children from age 4 years, which is the average age of onset of SM (Bergman et al. 2013, Black & Udhe, 1995; Dummit et al., 1997). Results from this study show that the SMQ-C is a reliable and valid measure to assess presence of SM symptoms. The SMQ-

C also provides the child with an opportunity to participate in the clinical assessment of their speaking behaviour in a more meaningful way.

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CHAPTER 3

Paper 2

SOCIAL PHOBIA SYMPTOMATOLOGY AND BEHAVIOURAL INHIBITION IN CHILDREN WITH SELECTIVE MUTISM: A COMPARISON WITH CHILDREN WITH SOCIAL PHOBIA AND NON-CLINICAL CONTROLS.

Abstract

A growing body of evidence points to a strong overlap between selective mutism (SM) and social phobia (SP) in children on behavioural characteristics such as social reticence and anxiety, yet few studies have directly compared these populations. The current study compared 25 children with a primary diagnosis of SM, 17 children with a primary diagnosis of SP and 15 non-clinical controls on expressions of social anxiety and inhibition. The child's mother, father, teacher and the child completed standard questionnaires, and the child participated in a behavioural assessment with their mother present. Children in the SM and SP groups were similar on most symptoms of social phobia and when they differed, children with SM showed higher scores. Children in the SM group and children in the SP group did not differ in non-verbal social anxiety or non-social anxiety and both groups were more anxious than non-clinical controls. On behavioural observation, children with SM were significantly more inhibited verbally and nonverbally than children in the SP group and nonclinical controls. Both clinical groups had fewer friends than non-clinical children and they experienced difficulties forming friendships. Children with SM also experienced greater difficulties sustaining friendships. The current findings support suggestions of strong similarities between SM and SP, with SM children perhaps showing greater severity than those with more general SP. The findings suggest that inclusion of cognitive behavioural skills to manage anxiety along with social skills training will improve treatment effectiveness for SM.

Introduction

Children with a diagnosis of Selective Mutism (SM) fail to speak in select social situations where speech is anticipated and/or expected (APA, 2014). These children understand spoken language, and they speak competently and confidently with immediate family and select others. The failure to speak is most commonly observed with adults and children at (pre)school (Black & Udhe, 1995; Steinhausen & Juzi, 1996; Letamendi, Chavira, Hitchcock, Roesch, Shipon-Blum & Stein, 2008). Referral for treatment is typically prompted by concerns that the prolonged mutism is impacting on the child's functioning in the classroom, playground, in extracurricular activities and/or with extended family (APA, 2014). Unlike other diagnoses where a constellation of symptoms is required to meet criteria, the lack of speech, its duration and impact are the sole and defining criterion for a diagnosis of SM. The prolonged failure to speak cannot be accounted for by another diagnosis such as Autism Spectrum Disorder or a speech and language delay/disorder (APA, 2014). Between 0.6 % to 3.3% of children in the first four years of school (Bergman, Piacentini & McCracken, 2002; Browne & Lloyd, 1975; Karakaya, Sismanlar, Memik, Coskun, Agaoglu and Yavuz, 2008; Kumpulainen, Räsänen, Raaska and Somppi, 1998) and 0.4% to 0.8% of pre-schoolers (Elizur & Perednik, 2003; Ezpeleta, de la Osa & Doménech, 2014) meet criteria for SM.

Research over the past 20 years has supported the view that SM is an anxiety-based disorder (Anstendig, 1999; Bergman et al, 2002; Bögels, Alden, Beidel, Clark, Pine, Stein & Voncken, 2010; Carbone, Schmidt, Cunningham, McHolm, Edison, St. Pierre & Boyle, 2010; Muris & Ollendick, 2015; Sharp, Sherman & Gross, 2007). This body of research lead to the reclassification of SM to the "Anxiety Disorders" section of the *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition* (DSM-5: APA, 2014). What remains unclear from current research is the specific relationship

between SM and the other anxiety disorders of childhood, and in particular the relationship between SM and Social Phobia (or Social Anxiety Disorder, as it has been re-labelled in DSM-5. The term Social Phobia (SP) will be used in this paper). Social anxiety is a prominent feature of SM (Bergman et al., 2002; Black & Udhe, 1992 & 1995; Bögels et al., 2010; Muris & Ollendick, 2015) and there is a high comorbidity of SP in children with SM (Kristensen, 2000; Levin-Decanini, Connolly, Simpson, Suarez & Jacobs, 2013; Manassis, Tannock, Garland, Minde, McInnes & Clarke, 2007; Muchnik, Hilesheimer, Arie, Bar-Heim & Henkin, 2013; Vecchio & Kearney, 2005). Furthermore, restricted speech is an observed behavioural response in SP (Heiser, Turner, Beidel & Roberson-Nay, 2009). This overlap in symptomatology has led some researchers to suggest that SM is a variant of SP rather than a distinct anxiety disorder - specifically that the persistent mutism is a more severe symptom of SP (Black and Udhe, 1995) or SM is an earlier childhood expression of SP (Bergman et al., 2002; Bögels et al., 2010). However, there is a dearth of evidence from which to draw conclusions about how similar the clinical presentation of SM is to SP in the earlier childhood years. The results from a handful of studies that have compared children with SM to children with SP on select aspects of the social phobia symptomatology are mixed and inconclusive. Furthermore, most such studies have assessed older children, with a mean age of 9 to 10 years (Yeganeh, Beidel, Turner, Pina & Silverman, 2003; Young, Bunnell & Beidel, 2012) and an age range up to 15 years old (McInnes, Fung, Manassis, Fiksenbaum & Tannock, 2004; Manassis, Fung, Tannock, Sloman, Fiksenbaum & McInnes, 2003; Yeganeh, Beidel & Turner, 2006).

Overall, findings have suggested that children with SM show more severe levels of social anxiety than children with SP when rated by clinicians (Yeganeh et al., 2003, 2006; Young et al., 2012;). However, parents do not report differences in the levels of

social anxiety between children with SM and children with SP (McInnes et al., 2004; Young et al., 2013). Based on observations that some children with SM are nonverbally socially engaged and responsive, some authors have argued that lack of speech may erroneously infer higher anxiety to adult observers (Carlson, Mitchell & Segool, 2008; Krohn, 1993; Yeganeh et al., 2003; Yeganeh et al., 2006; Young et al., 2013;). Interestingly, self-report ratings by children with SM have shown similar levels of social anxiety as reported by children with SP (Carbone et al., 2010; McInnes et al., 2004; Manassis et al., 2003; Yeganeh et al., 2003, 2006; Young et al., 2012), although one study found that children with SM reported lower levels of social anxiety relative to children with SP (Melfsen, Walitza & Warnke, 2006). A key methodological difficulty in this research is that most measures of social anxiety contain items related to speaking. Only one study to date has excluded speech items from their analysis and found that children with SM displayed more nonverbal social anxiety compared to non-clinical controls (Bergman et al., 2002). Comparison between children diagnosed with SM and SP using measures that distinguish verbal from non-verbal social anxiety has not been conducted.

Reluctance to spontaneously speak is identified as one of the most sensitive indices of behavioural inhibition (Kagan, Reznick & Snidman, 1987). Behavioural Inhibition (BI) refers to the initial extreme wariness and avoidance of unfamiliar and challenging objects, people and situations (Asendorpf, 1990; Kagan et al, 1987) and is widely supported as an early risk factor for development of SP (Rapee & Spence, 2004). A link between SM and BI has been postulated based on the similarities between SM and the pattern of behaviour observed in children with high BI (Lesser-Katz, 1986; Halpern, Hammond & Cohen, 1971). One idea has been that behavioural inhibition contributes to the initial avoidance of speech with new people and that social anxiety

plays a role in the persistence of the mutism (Muris, Hendriks & Bot, 2015). To date, the few studies that have examined BI or similar temperamental styles in children with SM have not included a comparison group (Ford, Sladeczek, Carlson & Kratochwill, 1998; Kristensen and Torgersen, 2002; Muris, Hendricks & Bot, 2015).

Fear of negative evaluation is a core feature of SP that is critical to its maintenance (Clarke & McManus, 2002; Ollendick & Hirschfeld-Becker, 2002; Rapee & Spence, 2004). Only one study has evaluated this construct in children with SM. Parents and children with either SM or SP completed the Fear of Negative Evaluation subscale of the Social Anxiety Scale for Children (SASC; La Greca, 1999) which revealed a trend towards higher scores among children with SP than children with SM (Manassis et al. 2003).

An issue of some discussion in the literature is whether children with SM lack interest in social relationships or whether their social wariness is triggered by social anxiousness. Descriptively, references are made to children with SM being interested in their peers (Lesser-Katz, 1986) and joining into social interactions nonverbally (Carlson et al, 2008; Kumpulainen et al, 1998). In contrast, Cholemkery and colleagues (2014) found that children with SM had significantly greater difficulties with social motivation (e.g. would rather be alone than with others, avoids starting social interactions with others) and social communication (e.g. ability to communicate feelings to others, trouble keeping up with the flow of a conversation) compared to children with SP (Cholemkery, Mojica, Rohrmann, Gensthaler & Freitag, 2014). These observed differences between children with SM and SP could be related to greater social anxiety and avoidance, a lower motivation or disinterest in engaging in social interaction, and/or lower social competency. As noted above, the available evidence on level of social anxiety in children with SM is mixed. There has been no direct assessment of

interest/motivation to join social interactions among children with SM. Several studies have however examined social competence. They found that parents and teachers rated children with SM as displaying significantly fewer social interaction skills when compared to children with Social Phobia, children with mixed anxiety and non-clinical controls (Cunningham, McHolm and Boyle, 2006; Carbone et al, 2010). Similarly, in behavioural assessments, clinicians rated children with SM as lower on social performance than children with SP (Young et al., 2013). A confounding factor in these behavioural assessments is that the children with SM failed to use their voice thus, the observed difference in competency may be a result of anxiety and concern about social evaluation (Hudson & Rapee, 2000; Klein, Armstrong & Shipon-Blum, 2012).

Summary and hypotheses

While there is a strong overlap in the behavioural representation of SM and SP, there are important gaps in the knowledge base of how children with SM are the same and how they differ from children with SP on the key features of social phobia. The present study sought to systematically compare children with a primary diagnosis of SM to children with a primary diagnosis of SP and a comparison group of non-clinical controls on the following aspects of social phobia: (i) the degree of verbal avoidance, (ii) level of non-verbal social anxiety and avoidance, (iii) fear of negative evaluation, (iv) intensity of verbal and nonverbal avoidance behaviour and inhibition, and (iv) interest, opportunities and skills to engage in social situations with familiar people. The study screened speech and language development, presence of externalising behaviours and presence of negative life events, as these factors had been identified as potential mediating or risk factors in either the SM or SP literature (Cohan, Chavira, Shipon-Blum, Hitchcock, Roesch & Stein, 2008; Rapee & Spence, 2004; Rapee & Szollos,

2002). The study focused on a younger sample of children from age 4 to 8 years, as age of onset of SM is typically 3 to 4 years (Bergman, Gonzalez, Piacentini & Keller, 2013; Black & Udhe, 1995; Dummitt et al.; 1997; Elizur & Perednik, 2003) and treatment is generally sought 2 to 5 years later (Bergman et al., 2013; Black & Udhe, 1995; Krohn, Weckstein & Wright, 1992). It was hypothesised that children with SM and SP would not differ on the variables being measured, and if a difference was found between children with SM and SP, children with SM would display more severe features of the variable being measured than children with SP.

Method

Participants

Participants were 57 children aged between 3 years 6 months and 7 years 11 months, their parent(s) and teacher (see Table 1). Children were recruited via distribution of flyers and emails to child mental health services, schools and child care services (38/57, 66.7% of the sample), direct referral from educational and health professionals (17/57, 29.8%), and a magazine advertisement (2/57, 3.5%). Three groups were formed: children with a primary DSM-IV diagnosis of Selective Mutism (SM, n=25), children with a primary diagnosis of Social Phobia (SP, n=17), and children who were identified as “easy-going” and on assessment did not meet any DSM-IV diagnostic criteria (C, n=15) (See Table 2). DSM-IV criteria were used as this study commenced prior to the publication of DSM-5. For children who met DSM-IV criteria for Selective Mutism, a comorbid diagnosis of Social Phobia was given if the child displayed a clinically significant level of social anxiety and avoidance in situations where use of voice was not the sole expectation of the interaction (e.g. “working or playing with a

group”, “sport class”, “meetings (scouts, team sports)”, “musical or athletic performances”, “attending parties, dances or activities”). For children who met DSM-IV criteria for both Selective Mutism and Social Phobia, Selective Mutism was identified as the primary diagnosis as the failure to speak was associated with the greatest impairment in functioning. Children in the three groups were not matched for gender or age. The majority of children lived with both biological parents (87.7%), or with one biological parent in a single-parent household or blended family (10.5%). The ethnic composition of the sample was: 40.3 % Oceanian or North West European (English speaking countries), 19.3% South/Eastern European, 17.5 % East Asian, 10.5% South/Central Asian, 8.8 % Middle Eastern and 3.6% other non-English speaking country. Four (7%) children attended day care, 17 (30%) were in preschool, 13 (23%) were in Kindergarten (first year of formal schooling in New South Wales) and 23 (40%) of the children were in primary school (Grade 1 and above).

Fifty-seven mothers (mean age = 38.21 years, $SD = 9.39$) and 52 fathers (mean age = 39.11 years, $SD = 5.29$) participated in the study. The majority of mothers in this study were either employed part-time (48.1%) or at home by choice (31.6%), while most father’s were in full-time employment (94.5%). Gross family income for 92.6% of families was in the middle to high income range.

Fifty-four teachers participated in the study. For all the children, their teacher had known and taught them for at least two months.

Measures

(1) Parent(s)-report measures about the child:

The *Anxiety Disorder’s Interview Schedule for DSM-IV: Child Version, Parent Interview Schedule (ADIS:C/P)* (Silverman & Albano, 1996) is a semi-structured

clinical interview, assessing the presence of DSM-IV anxiety, mood and externalising disorders experienced by children and adolescents. Only the parent interview was administered because of the age of the sample. Items relating to school were changed to preschool for the non-school age children. The interview was conducted with the child's mother either in person (n=30, 53%) or by telephone (n=27, 47%). Previous studies show the ADIS:C/P can be used reliably as a diagnostic tool with parents of preschoolers (Edwards, Rapee, Kennedy & Spence, 2010; Hudson, Dodd, Lyneham, & Bovopoulos, 2011; Rapee, 2014) and it can be administered by telephone (Lyneham & Rapee, 2005; Letamendi et al., 2008). The first author, a registered and experienced clinician, conducted the interviews and was not blind to potential group membership. The interviews were audio recorded and 18% (10/57) of the interviews were coded for inter-rater reliability by a second rater, an experienced clinician trained in the ADIS:C/P and blind to group status. Clinical Severity Ratings (CSRs on a scale of 0-8) were assigned based on number of settings, severity of symptoms and impact on functioning. A diagnosis was considered "clinical" if the CSR was four or greater. Primary and additional diagnoses were included. Interrater agreement was as follows: primary diagnosis ($\kappa = 1.00$), secondary diagnosis ($\kappa = 1.00$). The intraclass correlations for the clinician severity rating were: primary diagnosis ($ICC = .97$) and secondary diagnosis ($ICC = .75$).

The Interpersonal Relationships Module of the ADIS:C/P was examined to evaluate children's interpersonal functioning. The items included in the analysis were: number of friends (more/same/less) and whether they had troubles making friends or keeping friends (yes/no/undecided). This module has been shown to be sensitive to detecting differences in the number of friends in children with SP compared to anxious children without SP (Bernstein, Bernat, Davis & Layne, 2008).

Family Demographic Questionnaire: comprised of 34 questions that collected demographic data on the child and their immediate family. The child's mother completed the questionnaire.

The *Selective Mutism Questionnaire (SMQ)* (Bergman, Keller, Piacentini & Bergman, 2008) is a 23 item questionnaire assessing frequency of speech in children aged from 3 years as reported by parents. The first 17 items measure frequency of speech in the preceding month and the items load onto three factors: (i) school, (ii) home/family, and (iii) public/social situations outside of school. Parents rate frequency of speech from 0 (always) to 3 (never). A lower factor score indicates less speaking behaviour. The three factors of SDQ have acceptable internal consistency ($\alpha=.65$ to $.91$ in Letamendi et al., 2008), and moderate convergent and incremental validity (Bergman et al., 2008; Letamendi et al., 2008). The internal consistency of the three factors remained high in this study (Cronbach α : SDQ School: $.96$ for mothers and $.95$ fathers, SDQ Home/Family $.82$ for mothers and $.88$ for fathers, and SDQ Public $.95$ for fathers and $.93$ for mothers).

The *Revised Preschool Anxiety Scale (PAS-R)* (Edwards, Rapee, Kennedy & Spence, 2010) is a 28 item questionnaire that measures presence of anxiety symptoms in children from age 3 as reported by parents. The items load onto four factors consistent with DSM-IV diagnoses: social anxiety, generalised anxiety, separation anxiety and specific fears. Parents rate each item from 0 (not at all true) to 4 (very often true). Given the high frequency of "talking" items in the Social scale, this factor was excluded. The three remaining factors were summed to form a PAS-R Non-social Total Score. The PAS-R has acceptable internal consistency, strong stability over a 12-month period and moderate to high construct validity (Edwards et al., 2010). Cronbach α 's for the PAS-R Non-social Total in the current sample was $.91$ for mothers and $.89$ for fathers.

Social Anxiety Scale for Children – Revised/Parent Version (SASC-R/P)

(LaGreca, 1999) is a 22 item parent measure of their child's social anxiety in their relationship with peers. Eighteen of the items group to form three subscales: SASC-R/P-New (social avoidance and distress with new and less familiar social people), SASC-R/P-FNE (fear of negative evaluation or response by peers) and SASC-R/P-General (social avoidance and distress generally when interacting with others). The parent rates how true each statement was for their child on a 5-point Likert scale from 1 (not at all) to 5 (all the time). La Greca (1999) recommends use of subscale scores in research. The SASC-P/R-New and SASC-P/R-General contained items about speaking. The SASC-P/R-New was re-calculated without these items [SASC-P/R-new(ns)]. The SAD-General could not be recalculated as only one item remained after the speaking items were excluded thus, this subscale was not included in the analysis. The SASC-R/P measure has been shown to have good internal consistency and adequate validity (LaGreca, 1999). In the current sample, Cronbach α for the SASC-R/P-new(ns) was .85 for mothers and .89 for fathers, and for the SASC-R/P-FNE was .94 for mothers and fathers.

The *Strengths and Difficulties Questionnaire (SDQ)* (Goodman, 1997) is a 25 item measure screening for behavioural and emotional problems in children aged 4 to 16 years. The five subscales: emotion symptoms, conduct problems, hyperactivity/inattention, peer relationship problems and prosocial behaviours were included as separate variables in the current study. Parents rate each item on a three point Likert scale from 0 (not true) to 2 (certainly true). Studies of Australian children show that while there are questions about the factor structure of the SDQ (Mellor & Stokes, 2007) the SDQ has sound psychometric properties (Hawes & Dadds, 2004) and it is an effective screening instrument (Mathai, Anderson and Bourne, 2004; Mellor &

Stokes, 2007). In this study the Cronbach for the five factors were as follows:

Emotional symptoms: .77 for mothers and .73 for fathers; conduct problems: .66 for mothers and .62 for fathers; hyperactivity/inattention: .75 for mothers and .77 for fathers; peer relationship problems: .73 for mothers and .61 for fathers; and prosocial behaviour: .75 for mothers and .84 for fathers).

Short Temperament Scale for Children (STSC) (Sanson, Smart, Prior, Oberklaid & Pedlow, 1994) contains 30 items assessing four temperament dimensions: Approach (tendency to approach vs withdraw from novel situations and people), Inflexibility, Persistence and Rythmicity. Parents rate how well the item describes their child's recent and current behaviour on a 6 point Likert scale from 1 (Almost never) to 6 (Almost always). Higher scores indicated that the child had more of the temperament characteristic being measured. The four dimensions were shown to measure the same dimension of behaviour from 3 to 7 years, the age range of children in this study. The STSC has adequate validity and good internal consistency (Sanson et al., 1994). The internal consistencies of the subscales for this study were: Approach (mother $\alpha = .80$ and father $\alpha = .86$), Inflexibility (mother $\alpha = .86$ and father $\alpha = .84$), Persistence (mother $\alpha = .81$ and father $\alpha = .79$) and Rythmicity (mother $\alpha = .68$ and father $\alpha = .78$).

Child Social Preference Scale (CSPS) (Coplan, Prakash, O'Neil & Armer, 2004) is an 11 item measure that distinguishes between two forms of social withdrawal: shyness and social disinterest. Parents rate how well the item describes their child's general social behaviour on a 5 point Likert scale from 1 (not at all) to 5 (a lot). The scale has satisfactory internal consistency (Coplan et al., 2004) and good convergent validity (Coplan & Weeks, 2010). In this study, the internal consistency of the CSPS Shyness subscale was .68 for mothers and .87 for fathers, and Social disinterest subscale it was .78 for mothers and .72 for fathers.

The Life Events Scale (LES) is a measure adapted by Edwards, Rapee, Kennedy and Spence (2010) to include items relevant to preschool aged children. The questionnaire asks parents to rate whether 21 traumatic and stressful events had occurred for their family in the preceding 12 months. There is a 22nd open item allowing parents to identify other stressors. If an event had occurred, parents then rate whether they considered the impact to be positive or negative and the perceived impact of the event on the child from 0 (no impact) to 3 (extreme impact). The total number of negative events and the impact score of the events rated as negative were summed.

Developmental Profile 3 (DP-3) - Parent/Caregiver checklist (Alpern, 2007) is a standardised measure of child development and functioning from birth through to 12 years. The Socio-Emotional subscale was included as a screener of social competence skills. The Social-Emotional Standard Score was shown to have good reliability and adequate convergent validity (Alpern, 2007).

(2) Observation of the child's behaviour:

A *Behavioural Assessment* of the child's level of inhibition was conducted using observation of performance on a series of tasks used in previous research of B.I. (Kagan, Reznick & Gibbons, 1989; Edwards et al., 2010; Asendorpf, 1990). The first author (M.I.M.), who was unfamiliar to the child, conducted the interviews. The child's mother was present for the whole interview. The assessment was videotaped and quantified. The behavioural assessment consisted of:

1. "Waiting Time" (2 minutes). On arrival to the room, the child was directed to a small chair approximately one meter from their parent beside several toys. The child and parent were left unsupervised.

2. “Nonverbal social interaction” (3 minutes). An unfamiliar assessor sat opposite the child and unpacked a bag of toys without instigating any direct communication with the child. The clinician responded with a brief, neutral response if the child initiated conversation.
3. “Verbal social interaction” (3 minutes). The child was asked a series of questions by the assessor, beginning with closed questions about the toys and moving to open questions about the child (e.g. age, likes).
4. Three physical activities: (a) popping bubbles (the mother blew soap bubbles and the child was instructed to pop them with their nose whilst the assessor counted aloud the number of bubbles popped), (b) walking on stilts, and (c) popping a balloon with a pin or their body (i.e. sitting or standing on it).

The following variables were coded (in seconds or frequency counts) for the first three tasks: latency to speech/nonverbal gesture/touch a novel toy; total duration of the child’s speech; number of nonverbal gestures; total duration of time spent playing with novel toys; time spent within one arm’s length of parent; and frequency of times the child referenced their mother. For the fourth task, the child and parent rated the level of the child’s anxiety for each physical activity, and a rating of the child’s hesitancy for each physical task was assigned by the rater using a nine-point continuum ranging from zero to eight (with the middle rating of 4 representing a neutral point on the scale and a score of 8 indicting the child had not engaged in the activity).

Inter-rater reliability was determined by having a second coder independently score a random sample of 30% (n=19) of the assessments following a written manual. The coder, an experienced child clinical psychologist, and the first author coded the first video together to clarify the specifics of coding. The coder was blind to group membership. Interrater correlations were high for all variables ($r = 0.95$ to 1.0).

(3) Child-self report measures:

Peabody Picture Vocabulary Test, Fourth Edition (PPVT-IV) (Dunn and Dunn, 2007) is a nonverbal measure of receptive vocabulary skills in children from age two years and six months. The instrument is a useful screener for vocabulary development and for detecting language impairments (Dunn & Dunn, 2007). The children's standard score was included as a covariate as research suggests a higher prevalence of speech and language impairment among children with SM (Alyanak, Kiliñcaslan, Harmanci, Demirkaya, Yurtbay & Vehid, 2013; Anderson & Thomsen, 1998; Dummit et al., 1997; Elizur & Perednik, 2003; Kristensen, 2000; Manassis et al, 2007) and a link between social withdrawal and language performance (Coplan & Evans, 2009).

Play Choice Interview (PCI) (Coplan, Prakash, O'Neil & Armar, 2004) is a 13 item pictorial, nonverbal response measure that explores children's social interest/disinterest by asking the child with whom they would chose to play (another child, adult or alone) in a variety of common play activities. The number of times the child chose to play with a peer, adult or alone was summed. The internal consistency of the PCI for this study was $\alpha = .65$ ($n=54$).

Selective Mutism Questionnaire – Child (SMQ-C) [Milić & Rapee, 2015a] is a 20-item child-report measure of frequency of speech at school, home/family and in the community. Children use a two-stage pictorial response scale to indicate whether they communicate in the situation, and if they do communicate, how much they communicate from 1 (a little) to 3 (all the time). The three Factor structure of the parent-report SMQ was retained for scoring. The SMQ-C Factor scores have adequate to good internal consistency (Cronbach α = School .82, Home/Family . 67, Public/Social .80) and adequate congruent validity [Milić & Rapee, 2015a].

(4) Teacher report:

Demographic Information was collected about the structure of the classroom, and the length of time (in months) the teachers had (a) known the child and (b) taught the identified child.

School Speech Questionnaire (SSQ) (Bergman, direct communication, 2004) is a modified version of the school domain of the SMQ. The SSQ consisted of seven items. Six items measure frequency of speech with adults and children in the school context and the seventh item assesses interference. As with the SMQ, teachers used a 4-point Likert rating scale and a low score indicated less speech. The SSQ Cronbach α in this study was .96.

Teacher Rating Scale for Social Anxiety – “junior” (TRSA) (Bokhorst, Goosens & de Ruyter, 2001) is a 9-item scale asking teachers to rate the level of social anxiety in school social activities. Teachers rated each item on a 5 point Likert scale from 0 (not at all/never) to 5 (very/always) and a higher total score reflected greater social anxiety. The scale had demonstrated high reliability (internal consistency, $\alpha = .87$ to $.90$) and good convergent and divergent validity when used with children aged 5 to 6 years (BokHorst et al., 2001). Internal consistency of the scale continued to be high in this study ($\alpha = .92$).

Child Behavior Scale (CBS) (Ladd, 2010) is a 59 item teacher-report measure of the child's behaviour and relationships with peers in the school context. There are six subscales: an internalizing subscale (Anxious-Fearful), two externalizing subscales (Aggressive with Peers and Hyperactivity-Distractibility) and three subscales measuring peers relationships (Prosocial with Peers, Asocial with Peers, Excluded by Peers). Teacher's rated how applicable the description for each item was to the child on a 3 point Likert scale from 1 (Doesn't apply, seldom displays this behaviour) to 3

(Certainly applies, often displays this behaviour). The measure has been shown to be internally consistent and relatively stable across time with acceptable validity when used with young children from age 5 (Ladd, 2010). The internal consistency of the six subscales of the CBS were good (Cronbach α : Aggressive to peers $\alpha = .74$, Hyperactive Distractible $\alpha = .82$, Asocial with peers $\alpha = .94$, Anxious-Fearful $\alpha = .78$, Prosocial with peers $\alpha = .89$ and Excluded by peers $\alpha = .88$).

Procedure

Health (Western Sydney Local Health Network), education (NSW Department of Education and Communities, and Catholic Education Diocese of Parramatta) and the Macquarie University Human Research Ethics Committees granted approval for the study. Suitability for the study was assessed via a telephone screen interview. Children were excluded from the study if their parent reported a diagnosed developmental disorder (e.g. Intellectual Delay, Autism Spectrum Disorder) or their mother (as the primary adult participant) was unable to read and understand primary school level English. Data collection commenced with the parents and their child, and consisted of three steps: (a) parents completed a package of five questionnaires about the child [and three self-report questionnaires for a separate study: Milic & Rapee, 2015b], (b) the semi-structured diagnostic interview with the child's mother, and (c) the behavioural assessment with the child and their mother at the clinic following which the child completed the child measures with the clinician's support. The parent questionnaires were split into two packages and parents completed the second package of three questionnaires at point (c). Following the clinic appointment, the child's (pre)school teacher was invited to completed questionnaires. Consent to participate was received for the child and for each adult involved in the study. Parents of children in the SM and SP

group received a report containing recommendations on seeking support as reimbursement for their involvement in the study. As the clinical report was not relevant to children in the control group, families in the control group received a shopping voucher as reimbursement for their involvement in the study.

Statistical Analysis

Ten percent of the mothers and fathers who participated in the study failed to complete the second package of questionnaires and three children did not participate in the Behavioural Observation. Of the questionnaires returned, less than 1% of the data was missing, most commonly because of item nonresponse. Demographic characteristics were examined using one-way Analysis of Variance (ANOVA) and Fischer's exact test. As significant between groups differences were found for the child's age and their PPVT-IV Standard score, these two variables were entered as covariates in all Multivariate analyses of variance (MANOVA) and Univariate ANOVA. When the main effect for group was statistically significant, Univariate ANOVAs were computed and followed by pairwise comparisons to identify differences between groups. The Bonferroni adjustment was applied to each family of Univariate ANOVAs and pairwise comparisons. MANOVA could not be conducted on the behavioural assessment variables, as the assumption of homogeneity was violated thus a series of one-way ANOVA's was calculated. Fisher's exact test was used to calculate differences on the categorical data in the peer relationships section.

Results

Demographic variables

(i) Children: As shown in Table 1, the mean age of children differed significantly between the groups, $F(2,56) = 3.84, p = 0.03$. Children in the SP group were on average one-year older than children in the C group and close to a year older than children in the SM group. The sample contained significantly more female than male participants however; there was no significant difference between groups on gender distribution, $X^2(2, N = 57) = 3.48, ns$. A statistically significant difference was identified between groups on English as the primary language spoken at home, $X^2(2, N=57) = 8.32, p = 0.02$. English was not the primary language spoken at home for significantly more children in the SM group than children in the C group, $X^2(1, N=40) = 6.97, p = 0.02$. There was no significant difference between the SM and SP groups in number of children for whom English was not the primary language, $X^2(1, N=42) = 3.08, ns$. Furthermore, there was a significant between group difference on receptive vocabulary skills, $F(2,54)=9.10, p < 0.001$. The PPVT-IV standard score fell within the Average range or higher for the majority of children across all three groups (see Table 1). The SM children had a significantly lower Average PPVT-IV score relative to children in the SP and C groups, and these latter two groups did not differ significantly, $F(2,54) = 9.10, p < 0.001$. In addition, the groups differed significantly on whether previous services had been involved, $X^2(2, N=57) = 29.79, p < 0.001$. Significantly more children in the SM group received previous treatment than children in the C group, $X^2(1, N=40) = 16.44, p = 0.00$, or the SP group, $X^2(1, N = 42) 5.63, p = 0.02$. The main interventions were: school counsellor involvement (52%), paediatric assessment (36%), speech therapy (32%) and psychological treatment (24%). No

children were on medications for management of anxiety, mood or attentional difficulties.

(ii) Parents: Parent groups did not differ significantly on most demographic variables including: age, occupation, employment, reading ability, family income and paternal education. A statistically significant difference was found between groups on mother's education, $X^2(N=57) = 18.01, p = 0.009$. Mothers in the SM group differed significantly from mothers in the C group, $X^2(N=40) = 16.44, p = 0.001$, with 72% of mothers in SM group and 100% of mothers in the C group having completed further education post-high school. The mothers in the SM and SP group did not differ significantly on education, $X^2(N=42) = 5.63, ns$, and 88% of mothers from SP group had completed further education post-high school.

Table 1: *Demographic characteristics.*

Demographic variable	SM (n=25)	SP (n=17)	C (n=15)	Total (n=57)
Gender: Female [% (n)]	80.0% (20)	52.9% (9)	73.3% (11)	70.2% (40)
Children's mean age (mths) [mean (SD)]	68.4 (14.9) _{ab}	80.0 (17.8) _a	65.9 (15.0) _b	71.2 (16.6)
Primary Language spoken at home not English [% (n)]	36.0% (9)	11.8% (2)	0.0% (0)	19.3% (11)
PPVT-IV Standard Score (SD)	95.4 (12.0)* _a	105.2 (13.7) _{ab}	114.1 (14.9) _b	103.5 (15.2)
PPVT-IV Standard Score below 85 [% (n)]	12.0 % (3)	5.8% (1)	0.0% (0)	7.0% (4)
Previous services	84.0% (21)	47.1% (8)	0.0% (0)	50.9% (29)

Note: Means sharing subscripts are not significant different at $p \geq 0.05$.

* n = 23

Comorbidity

A two group (SM vs SP) ANOVA revealed no significant difference between children in the SM and SP groups on their ADIS-C/P clinician severity rating for their primary diagnosis, $F(1,40) = 0.55$, *ns* (see Table 2). Eight-eight percent of children with a primary diagnosis of SM met criteria for SP, and 18% of children with a primary diagnosis of SP had a prior history of SM. In the SM group, 44% met criteria for a comorbid anxiety disorder other than SP (see Table 2) and this percentage was not significantly different than the 52.9% comorbidity rate for children in the SP group, $F(1,40) = 0.26$, *ns*.

Table 2: *Primary and comorbid diagnoses in the two clinical groups, SM and SP.*

	SM (<i>n</i> = 25)	SP (<i>n</i> = 17)
Clinician Severity Rating for Primary Diagnosis [Mean number (<i>SD</i>)]	6.76 (0.97)	6.53 (1.00)
Comorbid Anxiety Disorder [Mean number (<i>SD</i>)]	1.56 (0.92)	0.82 (0.95)
Social Phobia [<i>n</i> (%)]	22 (88%)	-
Separation Anxiety Disorder [<i>n</i> (%)]	6 (24%)	4 (24%)
Generalised Anxiety Disorder [<i>n</i> (%)]	5 (20%)	7 (41%)
Specific Phobia [<i>n</i> (%)]	5 (20%)	3 (18%)
Selective Mutism (Prior History) [<i>n</i> (%)]	-	3 (18%)
Comorbid non-Anxiety Disorder		
Attention-Deficit Hyperactivity Disorder [<i>n</i> (%)]	0 (0%)	1 (6%)
Oppositional Defiant Disorder [<i>n</i> (%)]	1 (4%)	0 (0%)
Sleep Disorder NOS [<i>n</i> (%)]	0 (0%)	1 (6%)

Symptom Measures

(i) Speaking Behaviour: One-way MANOVA comparing groups on the SMQ, SMQ-C and SSQ revealed a significant main effect, Pillai's Trace = 1.23, $F(20,64) = 5.10$, $p < 0.001$, $\eta_p^2 = .62$. Follow up Univariate ANOVAs showed significant main effects for group on the SMQ (mothers and fathers) and SMQ-C School and Public subscales, the Teacher SSQ, and the SMQ Home subscale (mothers only) (see Table 3). Pairwise comparisons (Table 3) showed that on the mother-, father-, teacher- and child-reports for School, children in the SM group spoke significantly less frequently at school relative to children in the SP and C groups, and children in the SP group spoke significantly less at school compared to children in the C group. On the SMQ and SMQ-C Public subscales, pairwise comparisons revealed that children in the SM and SP group did not differ significantly from each other and that both clinical groups spoke significantly less when compared with children in the C group. Finally, pairwise comparisons revealed that on the mother's SMQ Home subscale, children in the SM group were rated as speaking significantly less than children in the C group, whereas children in the SP group did not differ significantly from either the SM or the C groups on mothers' ratings of frequency of speech at home. No significant main effect for group was found on either the fathers SMQ or child SMQ-C Home subscales.

(ii) Non-verbal social Anxiety: The results of the one-way MANOVA for mother-, father- and teacher-reports on the social anxiety measures revealed a main effect for group, Pillai's Trace = .73, $F(10,68) = 3.93$, $p < 0.001$, $\eta_p^2 = .37$. Follow-up Univariate tests revealed significant main effects for group on mother and father SASC-P/R:New(ns) and SASC-P/R:FNE subscales, and also on the teacher TRSSA. As shown in Table 3, parents and teachers rated children in the SM and SP groups as displaying significantly more nonverbal social anxiety and avoidance [ie. SASC-P/R-New (ns),

TRSSA] than children in the C group, and the SM and SP groups did not differ significantly from each other. On the SASC-P/R:FNE, mothers and fathers rated children in the SM group as experiencing significantly greater fear of negative evaluation than children in the C group. FNE scores for children in the SP group did not differ significantly from either the SM or C group.

(iii) Non-social Anxiety: The one-way MANOVA for mother-, father- and teacher-reports on non-social anxiety measures revealed a main effect for group, Pillai's Trace = .41, $F(10,78) = 2.01$, $p = 0.04$, $\eta^2 = .21$. Subsequent Univariate ANOVA's showed a significant main effect on only the mother's SDQ Emotional subscale (see Table 3). Pairwise comparisons revealed that mothers rated children in the SM and SP groups as displaying significantly more emotional symptoms than children in the C group, and mothers' ratings for the SM and SP groups were not significantly different (Table 3). The main effect for group on father's SDQ Emotional scale ($p = 0.007$) did not reach significance after the Bonferroni correction. Univariate ANOVA revealed no significant main effect for group on the other non-social subscales (see Table 3).

Table 3: *Internalising symptoms and temperament*

Theme	Source	Measure	SM Estimated Mean (SE)	SP Estimated Mean (SE)	C Estimated Mean (SE)	F-value
Speech*	Mother	SMQ School	0.41 (0.11) _a	1.56 (0.12) _b	2.35 (0.14) _c	$F(2,50) = 55.22, p < 0.001, \eta_p^2 = .69$
		SMQ Home	1.82 (0.12) _a	2.08 (0.14) _{ab}	2.72 (0.16) _b	$F(2,50) = 9.17, p < 0.001, \eta_p^2 = .27$
		SMQ Public	0.43 (0.11) _a	0.76 (0.12) _a	1.98 (0.13) _b	$F(2,50) = 39.17, p < 0.001, \eta_p^2 = .61$
	Father	SMQ School	0.39 (0.10) _a	1.57 (0.12) _b	2.30 (0.29) _c	$F(2,44) = 64.00, p < 0.001, \eta_p^2 = .74$
		SMQ Home	1.99 (0.15)	2.07 (0.17)	2.71 (0.19)	$F(2,45) = 4.52, ns, \eta_p^2 = .17$
		SMQ Public	0.36 (0.13) _a	0.90 (0.15) _a	2.01 (0.16) _b	$F(2,45) = 28.74, p < 0.001, \eta_p^2 = .56$
	Child	SMQ-C School	0.97 (0.12) _a	1.60 (0.13) _b	2.25 (0.15) _c	$F(2,49) = 20.50, p < 0.001, \eta_p^2 = .46$
		SMQ-C Home	1.71 (0.15)	2.10 (0.16)	2.32 (0.18)	$F(2,48) = 3.26, ns, \eta_p^2 = .12$
		SMQ-C Public	0.37 (0.12) _a	0.82 (0.13) _a	1.64 (0.15) _b	$F(2,48) = 20.21, p < 0.001, \eta_p^2 = .46$
	Teacher	SSQ	0.59 (0.14) _a	1.67 (0.15) _b	2.63 (0.18) _c	$F(2,48) = 38.01, p < 0.001, \eta_p^2 = .61$
Social Anxiety **	Mother	SASC-P/R New (ns)	15.75 (0.64) _a	15.09 (0.72) _a	8.39 (0.77) _b	$F(2,47) = 27.96, p < 0.001, \eta_p^2 = .54$
		SASC-P/R FNE	21.42 (1.41) _a	19.48 (1.57) _{ab}	12.81 (1.69) _b	$F(2,47) = 7.28, p = 0.002, \eta_p^2 = .24$
	Father	SASC-P/R New (ns)	15.25 (0.81) _a	13.88 (0.89) _a	7.58 (0.91) _b	$F(2,41) = 19.99, p < 0.001, \eta_p^2 = .49$
		SASC-P/R FNE	21.25 (1.57) _a	18.99 (1.73) _{ab}	13.41 (1.77) _b	$F(2,41) = 5.21, p = 0.01, \eta_p^2 = .20$
	Teacher	TRSSA	29.69 (1.68) _a	28.88 (1.79) _a	17.24 (2.18) _b	$F(2,46) = 10.76, p < 0.001, \eta_p^2 = .32$
	Mother	PAS-R Nonsocial	33.36 (3.52)	35.52 (3.96)	21.46 (4.47)	$F(2,50) = 2.95, ns, \eta_p^2 = .11$
		SDQ Emotional	4.55 (0.50) _a	5.11 (0.55) _a	1.54 (0.62) _b	$F(2,49) = 9.74, p < 0.001, \eta_p^2 = .28$
Non- Social Anxiety **	Father	PAS-R Nonsocial	33.06 (3.34)	31.30 (3.81)	22.54 (4.13)	$F(2,45) = 1.92, ns, \eta_p^2 = .08$
		SDQ Emotional	3.52 (2.44)	4.27 (1.70)	1.43 (1.34)	$F(2,45) = 5.50, ns, \eta_p^2 = .20$
	Teacher	CBS Anxious/ Fearful	1.67 (0.10)	1.59 (0.11)	1.25 (0.13)	$F(2,48) = 3.01, ns, \eta_p^2 = .11$
Temper- ament ***	Father	STSC Approach	32.74 (6.00) _a	30.20 (7.04) _a	18.36 (3.08) _b	$F(2,50) = 24.56, p < 0.001, \eta_p^2 = .50$
	mother	STSC Approach	32.33 (6.23) _a	32.18 (4.07) _a	19.13 (5.11) _b	$F(2,45) = 24.55, p < 0.001, \eta_p^2 = .52$

Note: Subscripts indicate difference at the critical value.

* Bonferroni adjustment for Speech (10 dependent variables): F value critical alpha $p < 0.005$, pairwise comparisons critical value $p \leq 0.002$.

** Bonferroni adjustment for the Social Anxiety and Non-Social Anxiety groupings (5 dependent variables): F value critical alpha $p < 0.01$, pairwise comparisons critical alpha $p \leq 0.003$

*** Bonferroni adjustment for Temperament (8 dependent variables): F value critical alpha $p \leq 0.006$, pairwise comparisons critical value $p \leq 0.002$.

(iv) Non-anxiety variables: Means and standard deviations for the mother-, father- and teacher-report measures of externalising behaviour and mother and father responses on the Life Events Scale are depicted in Table 4. The MANOVA did not reveal any significant main effect for group on measures of externalising behaviour, Pillai's Trace = .37, $F(12,76) = 1.47$, ns , $\eta_p^2 = .19$, or negative life events, Pillai's Trace = .06, $F(4,80) = 0.60$, ns , $\eta_p^2 = .03$.

Table 4: *Non-anxiety variables:*

Theme	Source	Measure	SM Mean (SD)	SP Mean (SD)	C Mean (SD)
Externalising	Mother	SDQ Hyperactive/Inattentive	3.19 (2.39)	2.86(2.23)	3.27(2.52)
		SDQ Conduct	2.27 (1.96)	1.59 (1.50)	1.40 (1.69)
	Father	SDQ Hyperactive/ Inattentive	3.67 (2.08)	3.80 (2.83)	3.07 (2.92)
		SDQ Conduct	2.05 (2.01)	1.60 (1.30)	1.86 (1.83)
	Teacher	CBS Hyperactive/Distractible	1.31 (0.43)	1.37 (0.58)	1.16 (0.33)
		CBS Aggressive	1.12 (0.26)	1.06 (0.12)	1.07 (.16)
Negative events	Mother	LES Total	1.48 (1.94)	1.56 (2.00)	0.80 (1.15)
	Father	LES Total	1.17 (2.04)	1.36 (1.45)	0.64 (0.84)

Behavioural inhibition:

(i) Parent-report: The MANOVA for mothers' and fathers' responses on the STSC revealed a significant main effect for group, Pillia's Trace=.92, $F(16,78) = 4.15$, $p < 0.001$, $\eta_p^2 .46$. Follow-up Univariate ANOVA found no main effect for group on either parent's STSC Inflexibility subscale, mothers: $F(2,50) = 3.01$, ns , $\eta_p^2 , .11$ and father: $F(2,45) = 2.36$, ns , $\eta_p^2 .11$; Persistence subscale, mother: $F(2, 50) = 0.89$, ns , $\eta_p^2 .03$ and fathers $F(2,45) = 0.86$, ns , $\eta_p^2 .04$; or Rhythmicity subscale, mothers $F(2,50) = 0.51$, ns , $\eta_p^2 .02$ and fathers $F(2,45) = 0.34$, ns , $\eta_p^2 .02$. There was however, a

significant main effect for group on the STSC Approach subscale (see Table 3). Both mothers and fathers rated children in the SM and SP groups as displaying a significantly greater tendency to withdraw from novel situations and people than children in the C group, and the SM and SP groups did not differ significantly from each other.

(ii) Behavioural Observation Tasks: The MANOVA did not reveal any main effect for group on the clinician's ratings of hesitancy and parent and child's ratings of anxiety in the physical tasks (bubbles, stilts and balloon) (see Table 5), Pillai's Trace = 0.27, $F(18,72) = 0.62$, *ns*, $\eta_p^2 = .13$. A second MANOVA did not reveal any main effect difference between groups on the number of times the child referenced their mother in the "nonverbal social interaction" and "verbal social interaction" tasks, Pillai's Trace = 0.04, $F(4,94) = 0.53$, *ns*, $\eta_p^2 = .02$.

As previously noted, a series of Univariate ANOVAs were computed for the remaining dependent variables in the first three tasks: (1) "waiting time", (2) "nonverbal social interaction" and (3) "verbal social interaction" (see Table 5). With regard to the dependent variables measuring speech, Univariate ANOVA's revealed a significant main effect for group for the average latency to talk (in seconds). Pairwise comparisons showed that children in the SM group took significantly longer to spontaneously initiate speaking in the "verbal social interaction task" than did children in the SP and C groups. There was also a greater latency in time before children in the SM group responded with spoken words in the "non-social verbal interaction" than for children in the SP and C groups. The children in the SP and C groups did not differ significantly in time taken to initiate or respond with speech (see Table 5).

Univariate ANOVA's also showed a significant main effect for group in the number of times the child spoke with their mother in the "waiting room" time before the clinician entered the room. Pairwise comparison revealed that the children in the SM

group spoke less frequently with their mother compared to children in the SP group. Interestingly, there was no significant difference between the SM and C groups or between the SP and C groups in the frequency of times the child spoke with their mother during the “waiting room” time. In terms of the number of times the parent spoke with their child during the “waiting room” activity (i.e. before the clinician entered the room), a univariate ANOVA did not find a main effect for group (Table 5).

Once the clinician was present in the room, the univariate ANOVA for amount of talking initiated by the child during the second task, the “nonverbal social interaction”, approached the Bonferroni corrected alpha level for the main effect of group ($p = 0.01$). Pairwise comparisons revealed a trend towards significance showing that children in the SM group spoke less in this task than children in the C group ($p = 0.003$); children in the SP group did not differ significantly from either the SM or C groups. On the third task, the “verbal social interaction”, univariate ANOVA showed a main effect for group for the average amount of time the child spoke (in seconds). Pairwise comparisons revealed that children in the SM group spoke significantly less than children in the C group (see Table 5). Pairwise comparisons also revealed that the difference in amount of talking (in seconds) on the “verbal social interaction” task by children in the SM and SP groups approached significance ($p = 0.002$). No significant difference was found between the children in the SP and C groups in the amount of talking on this third task (Table 5).

With regard to the dependent variables measuring nonverbal responses (gestures), Univariate ANOVA’s revealed a significant main effect for group for average latency (in seconds) for the child to spontaneously initiate a gesture as a means of communication with either their mother or the clinician in the “nonverbal social interaction task” (see Table 5). Pairwise comparisons showed that a significantly longer

period of time passed before the children in the SM group initiated a gesture when compared to children in the SP and C groups and that the latter two groups did not differ significantly in latency to initiate a nonverbal gesture (Table 5). For latency to respond with a gesture on the “verbal social interaction task”, univariate analysis did not reveal a main effect for group. Similarly, univariate analysis did not show a main effect for group on the number of gestures the child used in their communication with their mother whilst waiting for the clinician or the number of gestures made in the “verbal social interaction task”.

Finally, univariate ANOVA revealed no significant main effect for group on proximity to mother across the three tasks (Table 5). With regard to the latency to touch the first novel toy, Univariate analyses did not reveal a main effect for group in the “waiting room” tasks when the child was alone with their mother. However, the univariate analysis for latency to touch the toy in the clinician’s presence (ie. “nonverbal social interaction”) revealed that the main effect for group approached significance ($p = 0.006$). Pairwise comparisons showed a trend towards significant for the time taken (in seconds) for children in the SM group to touch a toy on the table under the direct observation of the clinician when compared to children in the SP and C groups (SM vs SP and SM vs C, $p = 0.006$), and that children in the SP and C groups did not differ significantly in latency to touch a toy ($p > 0.05$).

Table 5: *Behavioural observation tasks.*

Dependent variable	SM Estimated mean (SE)	SP Estimated mean (SE)	C Estimated mean (SE)	F value
Talk (clinician or mother) (sec.)				
Task 2: Latency to initiate	172.90 (13.53) _a	98.68 (14.76) _b	54.26 (18.30) _b	$F(2,47) = 13.20, p < 0.001, \eta_p^2 = .36$
Task 3: Latency to respond	160.66 (9.99) _a	27.64 (10.89) _b	14.12 (13.51) _b	$F(2,47) = 49.93, p < 0.001, \eta_p^2 = .68$
Talk (frequency)				
Task 1: Total by mother with child	8.22 (0.83)	11.84 (0.91)	9.76 (1.13)	$F(2,47) = 4.17, ns, \eta_p^2 = .15$
Task 1: Total by child with mother	5.60 (0.94) _a	10.97 (1.02) _b	9.34 (1.27) _{ab}	$F(2,47) = 7.35, p = 0.002, \eta_p^2 = .24$
Talk (with clinician or mother) (sec.)				
Task 2: Total talk, initiated by child	0.94 (2.18)	5.19 (2.37)	13.40 (2.95)	$F(2,47) = 5.06, ns, \eta_p^2 = .18$
Task 3: Total talk, responding	3.43 (2.81) _a	17.09 (3.06) _{ab}	27.00 (3.80) _b	$F(2,47) = 11.68, p < 0.001, \eta_p^2 = .33$
Gestures (to clinician or mother) (sec.)				
Task 2: Latency to initiate	149.35 (13.07) _a	77.89 (14.25) _b	27.32 (17.68) _b	$F(2,47) = 14.53, p < 0.001, \eta_p^2 = .38$
Task 3: Latency to respond	30.44 (8.47)	11.22 (9.23)	10.34 (11.45)	$F(2,47) = 1.39, ns, \eta_p^2 = .06$
Gestures (frequency)				
Task 1: Total to mother	3.70 (0.48)	2.25 (0.52)	3.96 (0.65)	$F(2,47) = 2.90, ns, \eta_p^2 = .11$
Task 3: Total to clinician	8.22 (0.77)	6.97 (0.84)	6.87 (1.05)	$F(2,47) = 0.72, ns, \eta_p^2 = .03$
Touch a novel toy (sec.)				
Task 1: Latency, mother's presence	23.90 (10.05)	20.61 (10.96)	25.92 (13.59)	$F(2, 47) = 0.05, ns, \eta_p^2 = .00$
Task 2: Latency, clinician presence	117.2 (16.6)	45.3 (18.1)	30.5 (22.4)	$F(2,47) = 5.76, ns, \eta_p^2 = .18$
Proximity to mother (sec.)				
Task 1	36.42 (8.58)	9.22 (9.35)	2.47 (11.60)	$F(2,47) = 3.20, ns, \eta_p^2 = .12$
Task 2	26.81 (11.28)	15.38 (12.29)	-6.56 (15.25)	$F(2,47) = 1.36, ns, \eta_p^2 = .06$
Task 3	17.17 (6.89)	6.75 (7.51)	-6.28 (15.25)	$F(2,47) = 1.81, ns, \eta_p^2 = .07$

Note : 1=Waiting time, 2=Nonverbal Social Interaction, 3= Verbal social interaction, sec. = seconds.
Bonferroni adjustment for Behavioural observation tasks (15 dependent variables): F value
critical alpha $p \leq 0.003$, pairwise comparisons critical value $p \leq 0.001$.
_{a, b, c.} Significant group difference at the critical alpha.

Table 5: *Behavioural observation tasks (continued).*

Dependent variable	SM	SP	C	F value
	Estimated mean (SE)	Estimated mean (SE)	Estimated mean (SE)	
Bubbles				-
1 Child-rating of anxiety	0.26 (0.11)	0.21 (0.12)	-0.02 (0.14)	
2 Parent-rating of anxiety	1.51 (0.41)	0.84 (0.44)	0.23 (0.51)	
3 Clinician rating of hesitancy	3.56 (0.73)	3.75 (0.78)	2.04 (0.91)	
Stilts				-
1 Child-rating of anxiety	0.32 (0.13)	0.22 (0.14)	0.11 (0.17)	
2 Parent-rating of anxiety	1.84 (0.46)	1.94 (0.48)	1.14 (0.57)	
3 Clinician rating of hesitancy	1.86 (0.52)	1.86 (0.55)	1.31 (0.64)	
Balloon				-
1 Child-rating of anxiety	0.69 (0.21)	0.72 (0.22)	0.88 (0.26)	
2 Parent-rating of anxiety	2.44 (0.58)	3.27 (0.61)	2.49 (0.72)	
3 Clinician rating of hesitancy	3.78 (0.83)	3.30 (0.88)	3.66 (1.04)	

Peer relationships

The MANOVA did not reveal any main effects for group on mother's or father's DP-3 Social-emotional standard scores. Pillai's Trace = 0.09, $F(4,80) = 0.95$, ns , $\eta_p^2 = .05$. A series of Fisher's Exact Tests were then conducted on the ADIS-C/P Interpersonal Relationships variables. Significant differences were revealed between the three groups on number of friends, $X^2(2, N = 57) = 16.81$, $p = 0.001$, difficulties making friends, $X^2(4, N = 57) = 31.80$, $p < 0.001$, and difficulties keeping friends, $X^2(4, N = 57) = 9.74$, $p = 0.02$. Further analyses, initially comparing the SM and C groups, revealed

that children in the SM group had fewer friends, $X^2(2, N = 40) = 12.23, p = 0.001$, greater difficulties forming friendships, $X^2(1, N = 40) = 21.71, p < 0.001$, and greater difficulties keeping friends, $X^2(2, N = 40) = 9.16, p = 0.005$, than children in the C group. A comparison of the SM and SP groups revealed no significant differences between these two groups in number of friends, $X^2(2, N = 42) = 0.84, ns$, difficulties making friends, $X^2(2, N = 42) = 1.58, ns$, or difficulties maintaining friendships, $X^2(2, N = 42) = 0.46, ns$. A MANOVA was then conducted for the group of variables measuring the quality of social relationships (Table 6) and this revealed a significant main effect for group, Pillai's Trace=1.06, $F(26,60) = 2.61, p = 0.001, \eta_p^2 = .53$. In subsequent Univariate analyses, a main effect for group was only found on the mothers' and fathers' responses on the CSPA Shyness subscale. Pairwise comparisons revealed that parents' ratings of shyness for children in the SM and SP groups were significantly higher than for children in the C group, and children in the SM and SP groups did not differ significantly in their shyness scores (Table 6). Univariate analyses did not reveal a significant main effect for group on children's self-ratings of interest in playing with another child or parent's ratings on the CSPA Disinterest subscales. Furthermore, univariate analysis did not find any significant main effect for group on mother-, father- or teacher-reports of prosocial behaviour or peer relational problems (as shown in Table 6).

Table 6: *Quality of peer relationships.*

Theme	Source	Measure	SM Estimated mean (SE)	SP Estimated mean (SE)	C Estimated mean (SE)	Significance
Interest	Child	PCI	6.43	5.62	5.49	$F(2,50) = 0.41, p = ns, \eta_p^2 = .02$
		Child	(0.71)	(0.79)	(0.90)	
		PCI	3.91	2.88	3.55	$F(2,50) = 1.00, p = ns, \eta_p^2 = .04$
	Mother	Alone	(0.48)	(0.54)	(0.61)	
		CSPS	28.25	25.02	13.32	$F(2,49) = 34.68, p < 0.001, \eta_p^2 = .59$
		Shy	(1.09) _a	(1.20) _a	(1.35) _b	
	Father	CSPS	12.40	13.19	10.53	$F(2,49) = 2.08, p = ns, \eta_p^2 = .08$
		Disinterest	(0.78)	(0.86)	(0.97)	
		CSPS	25.50	23.68	15.66	$F(2,45) = 14.94, p < 0.001, \eta_p^2 = .40$
		Shy	(1.12) _a	(1.27) _a	(1.38) _b	
		CSPS	11.13	13.21	10.23	$F(2,45) = 3.13, p = ns, \eta_p^2 = .12$
		Disinterest	(0.74)	(0.85)	(0.92)	
Prosocial	Mother	SDQ	6.99	6.58	8.75	$F(2,49) = 4.19, p = ns, \eta_p^2 = .17$
			(0.42)	(0.47)	(0.52)	
	Father	SDQ	6.46	6.97	7.92	$F(2,45) = 1.09, p = ns, \eta_p^2 = .05$
			(0.59)	(0.67)	(0.73)	
	Teacher	CBS	1.91	1.99	2.53	$F(2,48) = 4.43, p = ns, \eta_p^2 = .16$
			(0.13)	(0.14)	(0.16)	
Peer problems	Mother	SDQ	2.30	2.68	0.57	$F(2,49) = 4.52, p = ns, \eta_p^2 = .16$
			(0.43)	(0.47)	(0.54)	
	Father	SDQ	2.51	3.13	1.38	$F(2,45) = 2.60, p = ns, \eta_p^2 = .10$
			(0.46)	(0.2)	(0.57)	
	Teacher	CBS	1.56	1.66	1.19	$F(2,48) = 2.63, p = ns, \eta_p^2 = .10$
			(0.12)	(0.13)	(0.16)	
		CBS	1.23	1.22	1.12	$F(2,48) = 0.35, p = ns, \eta_p^2 = .01$
			(0.01)	(0.09)	(0.11)	

Note : Bonferroni adjustment for *Quality of Peer relationships* grouping (13 dependent variables):

F value critical alpha $p < 0.004$, pairwise comparisons critical value $p \leq 0.001$.

a, b, c. Significant group difference at the critical alpha.

Discussion

The aim of the present study was to empirically evaluate how children with SM are the same and how they differ from children with SP on the key features of social phobia. Previous findings of social anxiety and avoidance in children with SM (Carbone et al, 2010; McInnes et al., 2004; Mannasis et al., 2003; Yeganeh et al., 2003, 2006; Young et al., 2012) and high comorbidity of SP in children with SM (Bogels et al., 2010) have lead some researchers to question whether SM is a variant of SP rather than a distinct disorder (Bergman et al., 2002; Black & Udhe, 1992 & 1995; Dummit et al., 1997; Muris & Ollendick, 2015). This is the first known study to systematically compare a younger sample of children with a primary diagnosis of SM to children with a primary diagnosis of SP and a nonclinical control group. A fundamental limitation to comparing SM and SP is that the two groups are not mutually exclusive. Consistent with previous findings (Bogels et.al., 2010), the majority of children in the SM group in this sample had a comorbid diagnosis of SP. SM rarely exists in the absence of social anxiety (Bogels et al., 2010) thus, excluding children with a comorbid social phobia diagnosis from the SM sample would create difficulties with recruitment but more importantly such a group would not provide a valid representation of children with SM. Thus, the comparison is actually between children with a primary diagnosis of SM and comorbid social phobia or at the minimum high levels of social anxiety (who will be referred to a children with SM throughout this discussion), and children with a primary diagnosis of Social Phobia who in some instances have a previous history of selective mutism (referred to as children with SP). How these two clinical groups differ on the key features of social phobia remains important to clarifying whether SM is a variant of SP or a distinct disorder, and such a comparison may shed light on why one group of children with social anxiety and avoidance continue to fail to speak in select social

situations and another group of children use their voice. Interestingly, the results of this study pointed to a number of similarities between the two clinical groups, along with a few differences but only in severity.

With respect to speaking behaviour, the contrast was most evident at school. Parent-, teacher- and child-reports all showed that children with SM spoke significantly less than children with SP in the school environment. In turn, however, both clinical groups spoke significantly less than the non-clinical controls at school suggesting that children with SM might differ on speaking behaviour from children with SP largely in degree. This finding is consistent with previous research that identified school as the environment where mutism behaviour is most evident (Kumpulainen et al., 1998; Letamendi et al., 2008; Steinhausen & Juzi, 1996). In community situations with new and less familiar adults and peers, children with SM and SP did not differ significantly, and both groups spoke significantly less than non-clinical controls. Thus, SM and SP appear to share the characteristic of speech restriction (Heiser, Turner, Beidel & Roberson-Nay, 2009; Rao, Beidel, Turner, Ammerman, Crosby, & Sallee, 2007) and it is the consistency and extent of this restriction that identifies children as having SM.

Consistent with Manassis et al. (2003) this study also revealed some restriction of speech among children with SM in their interactions with their mother in context where there was a potential audience or in new situations. According to mothers' reports children with SM spoke significantly less in the home context than non-clinical controls. Children with SP appeared to score between these two groups and they did not differ significantly from either group. Similarly, in the behavioural assessment, children with SM spoke significantly less frequently when alone with their mother than children with SP. One possibility for the disinclination for SM children to speak is the anxiety associated with the fear of potential social scrutiny or the novel environment. An

alternative explanation might be the presence of subtle language difficulties. Consistent with previous studies, the children with SM in the current study had a lower mean receptive vocabulary score than the non-clinical controls (Mannasis et al 2003; Nowakowski, Cunningham, McHolm, Evans, Edison et al, 2009). Similarly, McInnes et al. (2004) observed that children with SM spoke less, used linguistically simpler language and provided fewer details when re-telling a story to their parent than children with SP. Interestingly, the children from the SM group in the current study were more likely to come from homes where English was not the primary language, a finding that has been demonstrated by others (Bergman et al., 2013; Bradley & Sloman, 1975; Elizur & Perednik, 2003; Steinhausen & Juzi, 1996). Combined, these differences point to the possible presence of a lack of comfort or ability with language that may especially characterise children with SM. Such language difficulties may heighten anxiety in situations where speech is anticipated and/or expected (Manassis et al., 2003) and this may be especially true when there is an audience.

Several researchers have questioned whether children with SM experience anxiety in social situations that do not require speech (Yeganeh et al, 2003; Carlson et al. 2007). The DSM-5 guidelines for differential diagnosis of SM and SP note “individuals with selective mutism may fail to speak because of fear of negative evaluation, but do not fear negative evaluation in social situations where no speaking is required (eg. nonverbal play)” (APA, 2014: p. 207). Contrary to this, the findings of this study were that children with SM and SP were both more non-verbally socially anxious according to parent and teacher report than non-clinical controls. Additionally, mothers and fathers rated children with SM as being significantly more fearful of negative evaluation compared to nonclinical controls. No difference was found between children with SM and SP or between children with SP and nonclinical controls on the parent’s

report of the child's fear of negative evaluation. Likewise, parents rated children with SM and SP as significantly shier than non-clinical controls. Children with SM and SP did not differ on shyness, a term commonly used to describe low to moderate symptoms of social phobia (Rapee & Spence, 2004). Furthermore, while no statistical differences emerged on the non-verbal activities in the behavioural observation (i.e. physical tasks, latency to respond with a gesture), the pattern of mean scores suggests that children with SM and SP were more hesitant and anxious than nonclinical controls. The failure to find a statistical difference on the behavioural assessment may be due to limited power (a limitation that is discussed later in this paper). Collectively, these findings indicate that children with SM experience similar, and possibly higher levels of social anxiety (given their higher fear of negative evaluation scores), relative to children with SP.

BI is seen as one of the key risk factors for the later development of social phobia (Clauss & Blackford, 2012). Researchers have postulated that SM shares this underlying risk based on the similarities in the behavioural manifestation of SM and BI (Halpern et al., 1971; Lesser-Katz, 1986), information from clinical interviews (Ford et al., 1998) and a study that assessed temperamental style in children with SM (Kristensen & Torgersen, 2002). This study provides the first behavioural assessment of BI in children with a primary diagnosis of SM. The behavioural tasks included in the observational assessment were used in previous research to measure the underlying temperament style BI. In the present study, children with SM took significantly longer (time in seconds) to initiate and respond with speech, and to initiate a non-verbal gesture of communication in the presence of the assessor, than did children with SP and non-clinical controls. Furthermore, the mean scores for latency in time for children with SM to begin playing with the novel toys in the presence of the unfamiliar assessor

approached significance, and the mean scores for proximity to mother were highest for children with SM. The pattern of scores for latency to play and proximity to mother resembled the findings on the above variables in which a significant result emerged, that is, the mean scores for children with SM were higher than for non-clinical controls, with SP falling in-between. Collectively, these observations point to children with SM being more behaviourally inhibited in novel social-interaction based activities with an unfamiliar person than children with SP and the non-clinical controls.

Further support for the link between SM and BI comes from the mothers and fathers responses on a parent report measure of temperament. Mothers and fathers rated children with SM and children with SP significantly higher on the Approach subscale than nonclinical controls. Children with SM and children with SP displayed a similar tendency to withdraw from novel situations and people. No differences were observed between the three groups on the Inflexibility, Persistence or Rythmicity subscales of the temperament measure. The finding of no difference between SM and SP on questionnaire measure and a difference on the laboratory assessment children may be associated with what is being measured, the observational measures assess inhibition on the first occasion that the child is in the novel situation, whereas the temperament questionnaire examines a pattern of behaviour across a period of time and in a number of different contexts (van Brakel, Muris & Bögels, 2004).

The final question this study investigated was whether the social withdrawal was related to a lack of motivation to seek out social interactions (Coplan & Weeks, 2010) and how children with SM compared with regard to opportunities for social interaction as well as skills in these interactions. On a self-report questionnaire asking children who they would chose to play with in a variety of common play activities (i.e. another child, adult or alone), children with SM chose to play with their peers equally as often as

children with SP and non-clinical controls. Furthermore, no differences were found on maternal or paternal ratings of social disinterest, teacher report of asocial behaviour, or parent and teacher report of prosocial behaviour. This suggests that children with SM and SP, like non-clinical controls, are interested in social interactions. However, based on maternal report, children with SM had fewer friends and they found it harder to make and keep friends than nonclinical controls, and children with SM and SP did not differ on these friendship variables. Previous research has also shown that children with SP had fewer friends and they find it harder to make friends than non-clinical controls and children with generalised anxiety (Scharfstein, Alfano, Beidel & Wong, 2011).

One hypothesis being investigated in the adult SP literature that may provide an explanation for the increased friendship difficulties is that “socially anxious people behave in ways that lead to a negative outcome” (Alden & Taylor, 2004: p. 860). Others perceive the socially reticent behaviour in adults with SP as less warm and outgoing, and as a result they may step back from the interaction, which the adult with SP then interprets as an indication of social scrutiny (Clarke & McManus, 2002). This study did not assess for differences in pro-social non-verbal micro behaviours (eg. smiles, eye contact) but differences were observed in the verbal micro behaviours. Again the pattern was one in which children with SM differed significantly from non-clinical controls, with SP children falling in-between. In the behavioural assessment, the experience of conversing with the unfamiliar person was significantly briefer for children with SM than non-clinical controls. Children spoke less with their mother in the novel environment of the clinic, and previous studies have shown that children with SM conversed less with their mother in situations where there is a potential audience (Manassis et al, 2003; Nowakowski et al, 2011). Thus, there are fewer positive experiences of using voice in situations in which they are socially anxious. This is likely

to contribute to the difficulties with friendships and may contribute to the maintenance of mutism behaviour. Further observational research of socially appropriate behaviour in familiar relationships is warranted.

The primary limitation of the present study, as previously noted, is that SM and SP are not distinct disorders. The majority of children with SM have a comorbid diagnosis of SP, and a proportion of children with SP have a previous history of SM. Group allocation was however, clear-cut as all children with a prolonged period of mutism, the sole symptom of SM, were assigned to the SM group and the inter-rater agreement was high. Further limitations of the present study were that groups were not matched for age and gender, and the small sample size. The small sample size, together with a conservative critical alpha, may mean that some of the non-significant findings are a result of insufficient power. Many of the mean scores showed differences between the clinical and nonclinical groups that failed to reach significance and many variables differed in a consistent pattern of mean scores whereby SM appeared to differ from non-clinical controls, with SP falling between. The lack of significance on many of these variables may reflect type 2 errors and only future research that includes markedly larger samples will be able to determine these relationships. Another potential limitation is sampling bias. The children who formed the SM and SP groups were referred for clinical assessment. The extent to which the present findings generalise to children with SM who do not seek treatment is not clear.

On the other hand, the strengths of this study include the use of widely used and psychometrically sound instruments and multiple informants, in particular, the inclusion of the child's perspective and observational measures of the child's behaviours. The findings of this study have meaningful implications for clinical practice. The research shows that children with a primary diagnosis of SM differ from children with a primary

diagnosis of SP without SM in the severity of social anxiety symptomatology. Recent randomised treatment trials revealed that a proportion of children with SM continued to experience higher levels of social anxiety or to meet criteria for SP post-treatment that focused on increasing the frequency of speech (Bergman et al, 2013; Oerbeck, Stein, Pripp & Kristensen, 2015). Thus, the well-supported techniques used to manage social anxiety in children (i.e. emotion management, thought challenging and parent education about anxiety and parenting an anxious child) (Rapee, Schniering & Hudson, 2009) should be included in the treatment together with the exposure-based practice to speaking (Bergman, 2013; Johnson & Wintgens, 2001; Kearney, 2010). Furthermore, age appropriate games and activities are often used as the medium through which the children practice their exposure steps at school with their peers. Children with SM may benefit from these exposure tasks being extended to include instruction and practice of social skills. Research demonstrates that children with high BI benefit from play-based social skills programs focused on building their socially appropriate behaviours and social skills (Coplan, Schneider, Matheson & Graham, 2010).

In summary, children with SM share the same core features that form the diagnosis of SP. The identified points of difference between children with a primary diagnosis of SM (and in most instances a comorbid diagnosis of SP) and children with SP (without a current persistent lack of speech) are that children with SM displayed more verbal and nonverbal behavioural inhibition in novel social situations, they speak less in select social contexts, and they are possibly more fearful of negative evaluation. Further research with older children who are able to reflect on their internal cognitive experience as well as studies of behavioural inhibition are required to clarify the role of these two variables in the onset and maintenance of SM. Observational studies of play with familiar peers and with family will provide important information about the impact

on friendships. Children with SM are socially anxious, and the higher behavioural inhibition and fear of negative evaluation suggests that they may experience greater social anxiety than children with SP.

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CHAPTER 4

Paper 3

SOCIAL ANXIETY AND PARENTING BEHAVIOUR AMONG PARENTS OF CHILDREN WITH SELECTIVE MUTISM, CHILDREN WITH SOCIAL PHOBIA AND NON-CLINICAL CONTROLS.

Abstract

Parents of children with selective mutism (SM), social phobia (SP) and non-clinical controls participated in a study exploring differences in level of negative affect, social phobia symptomatology and parenting. The study also explored whether there was an association between parental and child anxiety and whether differences in parenting were associated with child anxiety and mutism or parental social anxiety. Fifty-seven mothers and 50 fathers completed self-report measures, and the mother-child dyads were observed while the child completed a challenging puzzle. The study failed to find differences between groups in negative affect, social phobia symptomatology or parenting behaviour. However, it found that parents of children with SM reported more internal thoughts of frustration with their child's reticence than parents of nonclinical controls, while parents of children with SP did not differ from either group. Significant positive associations were found between observed level of encouragement and child anxiety and mutism for the two clinical groups. Furthermore, a positive association was found between maternal social anxiety and internal thoughts of frustration and solicitousness. Future observational studies of the parent-child interaction in social and non-social situations are required to further our understanding of the differences in how the parents think about their child's mutism and anxious behaviour, and to examine differences in verbal parenting behaviour.

Introduction

Selective Mutism (SM) is a childhood anxiety disorder that typically has an onset around the age of three to four years (Bergman, Gonzalez, Piacentini & Keller, 2013; Black & Udhe, 1995; Dummit, Klein, Tancer, Asche, Martin & Fairbanks, 1997; Elizur & Perednik, 2003). Children with SM are capable of spoken language and they speak comfortably at home with their immediate family. They fail to speak with adults or children in select social situations, most commonly (pre)school, in extracurricular activities, and with members of the extended family (Bergman, Piacentini & McCracken, 2002; Kumpulainen, Räsänen, Raaska & Somppi, 1998). A defining criterion for SM is the duration of the mutism. The child must have failed to speak for at least one month, not including the first month in an educational placement (APA, 2014). The anxiety experienced by children with SM is however, not specific to speaking interactions. Research clearly shows a strong overlap between SM and social phobia (Bögels, Beidel, Clark, Pine, Stein & Voncken, 2010; Muris & Ollendick, 2015). Children with SM are nonverbally socially anxious like children with social phobia, and they are more behaviourally inhibited in novel situations and with new people than both children with social phobia and non-clinical controls (Milic & Rapee, 2015). The failure to speak and restricted nonverbal behaviour are also observed in interactions with members of the immediate family when people with whom they do not speak are present (Alyanak, Kiliñaslan, Harmanci, Demirkaya, Yurtbay & Vehid, 2013; Cunningham, McHolm, Boyle & Patel, 2004). Furthermore, a large proportion of children with SM have comorbid social phobia (Bergman et al., 2013; Black & Udhe, 1995, Dummitt et al, 1997) and some children with SM are diagnosed with several concurrent anxiety disorders (Black & Udhe, 1995; Chavira, Shipon-Blum, Hitchcock,

Cohan & Stein, 2007; Dummit et al, 1997; Kristensen, 2000; Levin-Decanini, Connolly, Simpson, Suarez & Jacob, 2013).

Families of children with SM seek treatment on average two to five years post onset (Black & Udhe, 1995; Dummit et al, 1997; Kristensen, 2002; Krohn, Weckstein & Wright, 1992). Thus, there is a significant period of time before treatment starts during which parents play a central role managing the child's mutism and social wariness. The interaction between the parent and the child around the SM is circular, parents and children respond to as well as elicit responses from the other (Rapee & Spence, 2004). When anxious, children move closer to their parents for support. Their mutism and nonverbal wariness are likely to elicit different parenting behaviours than would the behaviours of an "easy-going" child who confidently and comfortably interacts with the outside world. Parents observing their child's discomfort may make a greater effort to engage their child in the conversation or take over to relieve their child's distress (Edison, Evans, McHolm, Cunningham, Nowakowski et al, 2011). Anecdotally parents describe oscillating between being supportive and feeling frustrated at the contrast to home where their child is often very talkative (Hessellman, 1980). Furthermore, in these earlier childhood years, it is parents who organise the activities with peers (e.g. play dates, extra-curricular sports and hobbies, outings). The parent may participate less in social activities (Towe-Goodman, Franz, Copeland, Angold & Egger, 2014) and be less encouraging of developmentally appropriate independent behaviour (Rubin, Nelson, Hastings & Asendorpf, 1999). Additionally, preschool anxiety impacts family functioning. Towe-Goodman et al. (2014) identified the greatest impact from anxiety was on parental wellbeing, as parents worried and were more concerned about their child and their sense of confidence in their parenting was challenged (Towe-Goodman et al. 2014). Parents also find themselves fielding

questions, comments and judgements from adults and children about their child's failure to speak (Roe, 2015; Johnson & Wintgens, 2001).

Given the important role parents play in social development at this early stage, it is vital to understand the potential impact that the interaction between the parent and child may have on the development, maintenance and amelioration of SM. There is a large body of research exploring the role of genetics and environmental factors in the aetiology of childhood anxiety. Factors researched include: parental anxiety and the link between parent anxiety and child anxiety, as well as differences in parenting behaviour and whether differences in parenting are related to the child's anxiety or parent's anxiety. Parallel research in the field of SM is relatively sparse but hints at similarities between families of children with SM and families of children with another clinically diagnosed anxiety disorder.

Parental anxiety: Childhood anxiety research clearly shows that anxiety runs in families, with estimates of heritability ranging from 0.25 and 0.50 (Czajkowski, Røysamb, Reichborn-Kjennerud & Tambs, 2010; Hettame, Neale & Kendler, 2001). In light of the close relationship between SM and social anxiety (Muris & Ollendick, 2015), a closer examination of the SP literature shows that there is a significantly higher rate of Generalised Social Phobia and Avoidant Personality Disorder in the immediate family members of adults with SP than in control groups (Cooper & Eke, 1999; Ollendick & Hirshfeld-Becker, 2002). For children in the preschool years, which is the average age of onset of SM, a recent study by Hudson, Dodd & Bovopoulos (2011a) supported the association between parental and child anxiety. Furthermore, Hudson, Dodd, Lyneham & Bovopoulos (2011b) demonstrated that maternal anxiety when the child was aged 4 years was a significant predictor of clinical anxiety in the child at age 6. Maternal anxiety conferred both a genetic and environmental risk (e.g. modelling of

anxious responses, information processing) for the development of anxiety in later childhood (Hudson et al, 2011b).

With regard to SM, earlier studies identified that parents and siblings of children with SM were more likely to have SP, Avoidant Personality Disorder and SM; be speech avoidant (taciturn) or shy; and have emotional disorders (Black & Udhe, 1995; Brown & Lloyd, 1979; Kolvin & Fundudis, 1989; Kristensen, 2000; Remschmidt, Poller, Herpertz-Dahlmann, Henninghausen & Gutenbrunner, 2001; Steinhausen & Adamek, 1997; Wergerland, 1979). However, these studies lacked a comparison group, and the conclusions were based on non-standardised questionnaires and clinical interviews with an adult family member who was not always the family member identified as symptomatic. Later studies that included a clinical or non-clinical comparison group examined the presence of dispositional characteristics that resembled or were suggestive of higher social anxiety. Consistent with the earlier findings, these studies showed significantly higher rates of restricted speech, social avoidance and neuroticism in family members of children with SM than in clinical and nonclinical control groups (Kristensen & Torgersen, 2001, 2002; Steinhausen & Adamek, 1997). Two recent studies have specifically assessed social anxiety and the incidence of social phobia in parents of children with SM. Edison and colleagues (2011) found that primary care-givers (90% were the child's mother) reported low levels of general and social anxiety, and their scores did not differ significantly from the primary care-givers of children with mixed anxiety and no-anxiety. However, the researchers noted that some parents in the study "were observed to be anxious during the laboratory task" (Edison et al, 2011: p. 287), which suggests that they may have underreported their level of anxiety. Chavira et al. (2007) provide the only study that has assessed mothers and fathers separately using a semi-structured interview to diagnose the presence of DSM-

IV Axis I and Axis II disorders. They found that Generalised Social Phobia and Avoidant Personality Disorder were three to four fold more common among SM parents, and child SM severity predicted parental Generalised Social Anxiety. Examination of parents individually revealed that it was the fathers of children with SM who were significantly more likely to have a diagnosis of Generalised Social Phobia and Avoidant Personality Disorder than fathers of control children, and the difference between mothers did not reach significance. Chavira et al (2011) questioned whether the failure to find a difference between mothers was due to low sample size or differences in genetic vulnerability associated with parent gender.

Relationship between the child's anxiety and parenting: While research indicates that genetic factors play a moderate role in the development of childhood anxiety (Hettema et al, 2001; Rapee, Schniering & Hudson, 2009), a meta-analysis of studies of the association of childhood anxiety and parenting reported that parenting accounted for only a modest four to six percent of the variance in childhood anxiety (McLeod, Wood & Weisz, 2007). Examination of the sub-dimensions of parenting revealed that higher levels of parental over-involvement and withdrawal, and lower levels of parental warmth and autonomy-granting were associated with higher levels of child anxiety (McLeod et al., 2007). Furthermore, the association between parenting and child anxiety was stronger for children with a diagnosed anxiety disorder than children with subclinical levels of anxiety (McLeod et al, 2007). Child gender did not moderate the association between childhood anxiety and parenting (McLeod et al, 2007) however, child's age may act as a moderator. McLeod et al. (2007) found no association whereas Hudson and Rapee (2001) and van der Bruggen, Stams & Bögels (2008) showed that parental control (i.e. over-involvement and less autonomy-granting) was positively associated with the age of the child. This is important to consider given the

onset of SM is earlier than that observed in many of the other childhood anxiety disorders (Black & Udhe, 1995; Dummit et al, 1997; Kristensen, 2002).

Of the few studies that had compared parenting of children with SM and non-clinical controls, the majority found parents of children with SM did not differ from controls on questionnaire measures of discipline practices (Alyanak et al., 2013; Cunningham et al., 2004; Yeganeh, Beidel & Turner, 2006), overprotective behaviour (Alyanak et al., 2013; Yeganeh et al., 2006) or parental acceptance (Yeganeh et al. 2006). Furthermore, Yeganeh et al. (2006) found that parents of children with SM and children with SP did not report differences in parent overprotection and acceptance, and Alyanak et al. (2013) found no differences in parenting based on parent gender. The lack of differences might reflect the fact that all of these studies used questionnaire measures, which have shown smaller effects than observational measures (McLeod et al., 2007). Edison and colleagues (2011) provide the only observational study comparing how mothers of children with SM interact with their child in comparison to mothers of children with mixed anxiety and children with no anxiety. Mothers and children participated in a series of tasks including free play, helping the child prepare a speech, and supporting the child to deliver the speech. Edison et al. (2011) found that mothers of SM children displayed more parental control (i.e., granted their children less autonomy and made more high powered remarks such as directives, choice and process questions) in comparison to mothers of children with Anxiety and No-anxiety. Furthermore, parental control increased with higher observed child anxiety and with decreasing child age.

Relationship between parental anxiety and parenting: A meta-analytic review by van der Bruggen and colleagues (2008) identified that the association between parental anxiety and parenting was weak. Murray and colleagues (2012)

showed that the association between parental anxiety and difficulties with parenting emerged when parents were placed in a challenging situation that triggered what they were most anxious about. The differences that did emerge in parenting, namely clinically anxious parents showed lower parental encouragement and warmth and an increase in parental withdrawal, were consistent across parent anxiety disorders (Murray, Arteche, Creswell, Russ, Zoopa et al., 2012). Given the above-mentioned research of a higher incidence of SP in parents of children with SM, one could postulate that having a child continually fail to speak would be an anxiety provoking situation for the parent and may thus lead to an increase in overprotective parenting. The only study that has explored the association between parental anxiety and SM in children (Edison et al. 2011) found that parental anxiety was associated with how much autonomy mothers granted their child in a free play situation but not in a structured speaking task. Edison et al. (2011) hypothesised that increased parental anxiety in the free play task may have been associated with the lack of structure during free-play or the perception that both the parent and the child were being equally observed in the free play relative to the speaking task where the focus was more on the child.

Summary and hypotheses

The SM research provides some initial support for an association between parental anxiety and child anxiety in children with SM. The recent study by Edison et al. (2011) indicates that there are differences in how mothers parent their child with SM, and that there is an association between parenting and the child's anxiety and between parenting and maternal anxiety. The present study sought to contribute to this body of knowledge by exploring: (a) the emotional health, in particular negative emotion and

social phobia symptomatology, of mothers and fathers of children with SM, (b) the parent-child interaction in situations in which the parent was aware their responses may be a focus of attention, and (c) the association between child anxiety, parental anxiety and parenting behaviour. This is the first study to explore the association between the father's anxiety and their parenting behaviour. A decision was also made to include children with SP as the clinical comparison group because of the current important questions in the literature about the relationship between SM and Social Phobia (SP) (Muris & Ollendick, 2015).

Based on the abovementioned research, it was hypothesised that (a) both mothers and fathers of the SM and SP children would report higher rates of social phobia symptomatology than parents of non-clinical children, (b) parents of children with SM and SP would display more overprotective behaviours and less warmth than parents of non-clinical controls, (c) there would be a positive association between the parent's social anxiety and child's level of social anxiety, (d) child mutism and anxiety would be related to parental warmth and over-involvement, and finally (e) parental anxiety would be related to parental warmth and over-involvement.

Method

Participants

Parents of children aged four to eight years who had participated in a broader study (Milić & Rapee, 2015) were the focus of this study. Children were recruited for the broader study via distribution of flyers and emails to child mental health services, child care services and to schools (66.7% of the sample), direct referral from educational and health professionals (29.8%) and a magazine advertisement (3.5%). Three groups were recruited: children with a

primary DSM-IV diagnosis of Selective Mutism (SM group, $n = 25$), children with a primary DSM-IV diagnosis of Social Phobia (SP group, $n = 17$) and children who were identified as “easy-going” and on assessment did not meet any DSM-IV diagnostic criteria (C group, $n = 15$) (see Table 1). DSM-IV criteria were used as this study commenced prior to the publication of DSM-5. For children who met DSM-IV criteria for Selective Mutism, a comorbid diagnosis of Social Phobia was given if the child displayed a clinically significant level of social anxiety and avoidance in situations where use of voice was not the sole expectation of the interaction (e.g. “working or playing with a group”, “sport class”, “meetings (scouts, team sports)”, “musical or athletic performances”, “attending parties, dances or activities”). For children who met DSM-IV criteria for both Selective Mutism and Social Phobia, Selective Mutism was identified as the primary diagnosis as the failure to speak was associated with the greatest impairment in functioning (see Table 2). Children were excluded if they had a diagnosed developmental disorder (e.g. Intellectual Delay, Autism Spectrum Disorder) or their mother’s English reading and comprehension skills were below primary school level English. The ethnic composition of the sample was: 40.3 % Oceanian or North West European (English speaking countries), 19.3% South/Eastern European, 17.5 % East Asian, 10.5% South/Central Asian, 8.8 % Middle Eastern and 3.6% other non-English speaking country.

Fifty-seven mothers (mean age=37.1 years, $SD = 4.6$) and 50 fathers (mean age = 39.4 years, $SD = 5.2$) participated in the study (see Table 1). The majority were married (86%) or in a de-facto (7%) relationship. Most mothers were at home by choice (49.1%) or employed part-time (31.6%), while most fathers (94.1%) were in full-time employment. Ninety-two percent of families (54 of the 57 disclosed) identified middle to above average family income.

Table 1: *Demographic variables.*

Demographic variable	SM (n=25)	SP (n=17)	C (n=15)	Total
Child				
- Mean age (mths) (SD)	68.4 (14.9) _{ab}	80.0 (17.8) _a	65.9 (15.0) _b	71.2 (16.7)
- Gender: Female [% (n)]	80.0% (20)	52.9% (9)	73.3% (11)	70.2% (40)
Mother				
- Number of participants.	25	17	15	57
- Mean age in years (SD)	35.9 (4.6)	38.2 (5.2)	37.9 (3.8)	37.1 (4.6)
- Education (%)				
- High School	28.0%	11.8%	0.0%	15.8%
- Trade	8.0%	5.9%	0.0%	5.3%
- Certificate/Diploma	28.0%	11.8%	13.3%	19.3%
- Bachelor Degree	32.0%	52.9%	33.3%	38.6%
- Postgraduate Degree	4.0%	17.6%	53.3%	21.1%
Father				
- Number of participants.	22	14	14	50
- Mean age in years (SD)	38.8 (4.5)	39.9 (7.0)	39.8 (4.3)	39.4 (5.2)
- Education				
- High School	21.7%	14.3%	14.3%	17.6%
- Trade	17.4%	35.7%	7.1%	19.6%
- Certificate/Diploma	30.4%	7.1%	28.6%	23.5%
- Bachelor Degree	8.7%	28.6%	28.6%	19.6%
- Postgraduate Degree	21.7%	14.3%	21.4%	19.6%

Note: Means with different subscripts are significantly different at $p=0.05$.

Table 2: *Primary and comorbid diagnoses in the two clinical groups, SM and SP.*

	SM (<i>n</i> = 25)	SP (<i>n</i> = 17)
Clinician Severity Rating for Primary Diagnosis [Mean number (<i>SD</i>)]	6.76 (0.97)	6.53 (1.00)
Comorbid Anxiety Disorder [Mean number (<i>SD</i>)]	1.56 (0.92)	0.82 (0.95)
Social Phobia [<i>n</i> (%)]	22 (88%)	-
Separation Anxiety Disorder [<i>n</i> (%)]	6 (24%)	4 (24%)
Generalised Anxiety Disorder [<i>n</i> (%)]	5 (20%)	7 (41%)
Specific Phobia [<i>n</i> (%)]	5 (20%)	3 (18%)
Selective Mutism (Prior History) [<i>n</i> (%)]	-	3 (18%)
Comorbid non-Anxiety Disorder		
Attention-Deficit Hyperactivity Disorder [<i>n</i> (%)]	0 (0%)	1 (6%)
Oppositional Defiant Disorder [<i>n</i> (%)]	1 (4%)	0 (0%)
Sleep Disorder NOS [<i>n</i> (%)]	0 (0%)	1 (6%)

Measures

(1) Parent self-report measures:

The Depression Anxiety Stress Scale-21 (DASS-21) (Lovibond & Lovibond, 1995) is the short form of the DASS, a self-report questionnaire measuring three dimensions of negative emotion: depression, anxiety and stress. Individuals rate how much each statement applied to them over the preceding week, from 0 (did not apply to me at all) to 3 (applied to me very much, or most of the time). To calculate comparable scores with the full DASS, the items within each dimension were summed and doubled. Higher scores indicated more symptomatology. The DASS-21 is highly reliable and possesses adequate construct validity (Henry & Crawford, 2005). Cronbach α 's for the DASS Total score in the current sample are reported in Table 3.

Social Phobia 12 (SP-12) (Peters, Sutherland, Andrews, Rapee & Mattick, 2012) comprises six items from the Social Interaction Anxiety Scale (SIAS), which assesses the more generalised social interaction anxieties, and six items from the Social Phobia Scale (SPS), which assesses the anxiety of being scrutinised during routine activities. The individual rates how true each statement is on a five-point Likert scale from 0 (not at all characteristic or true of me) to 4 (extremely characteristic and true of me). The items within each subscale are summed to provide a SP-12 Total Score. The SIAS-6 and SPS-6 have good convergent validity and they discriminate well between those with and without a diagnosis of social phobia (Peters et al, 2012). In the present sample, Cronbach α 's for the SP-12 were .93 for mothers and .88 for fathers.

Leibowitz Social Anxiety Scale- Self-Report (LSAS-SR) (Baker, Heinrichs, Kim & Hofmann, 2002) is a 24-item self-report questionnaire assessing amount of fear and avoidance experienced in thirteen social interaction situations and eleven performance situations. The individual provides two ratings for each item. The first rating was the amount of fear experienced in the situation from 0 (no fear or anxiety in this situation) to 3 (severe fear or anxiety), and the second rating was the frequency of avoidance of the situation from 0 (never) to 3 (usually avoid this situation, 67 to 100% of the time). The LSAS-SR performance (fear and avoidance) and LSAS-SR social interaction (fear and avoidance) scores were summed to form a LSAS-SR Total score. The LSAS-SR has strong internal consistency, and good construct and predictive validity (Fresco, Coles, Heimberg, Liebowitz, Hami et al., 2001; Rytwinski, Fresco, Heimberg, Coles, Liebowitz et al., 2009). Cronbach α 's for the LSAS-SR Total in the current study were .97 for mothers and .97 for fathers.

(2) Measures about the child:

Child's Diagnosis: A semi-structured diagnostic interview was conducted with the child's mother using the *Anxiety Disorder's Interview Schedule for DSM-IV: Child Version, Parent Interview Schedule (ADIS:C/P)* (Silverman & Albano, 1996). The ADIS:C/P, assesses for the presence of symptoms consistent with the DSM-IV anxiety, mood and externalising disorders experienced by children and adolescents. This was administered either in person (53%) or by telephone (47%). Previous studies have found the ADIS:C/P can be used reliably as a diagnostic tool with pre-schoolers (Hudson et al., 2011b; Kennedy, Rapee & Edwards, 2009) and it can be administered by telephone (Lyneham & Rapee, 2005; Letamendi, Chavira, Hitchcock, Roesch, Shipon-Blum & Stein, 2008). The first author, a registered and experienced clinician, conducted the interviews. The interviews were audio recorded and 18% were coded for inter-rater reliability by a second rater, an experienced clinician trained in the ADIS:C/P and blind to group status. Clinical Severity Ratings (CSRs on a scale of 0-8) were assigned based on number of settings, severity of symptoms and impact on functioning. A diagnosis was considered "clinical" if the CSR was four or greater. Interrater agreement was as follows: primary diagnosis ($\kappa = 1.00$), secondary diagnosis ($\kappa = 1.00$). The intraclass correlations for the clinician severity rating were: primary diagnosis ($ICC = .97$) and secondary diagnosis ($ICC = .75$).

Family demographic questionnaire: comprised 34 questions that collected demographic data on the child and their immediate family. The child's mother, the primary adult participant, completed the questionnaire.

The *Selective Mutism Questionnaire (SMQ)* (Bergman, Keller, Piacentini & Bergman, 2008) is a 23-item parent-report questionnaire that measures frequency of speech in the preceding month across three domains/factors: (i) school, (ii)

home/family, and (iii) public/social. The first 17 items assess frequency of speech from 0 (never) to 3 (always). The mean of the three factors scores (average of items within a subscale) formed the SMQ Total score. The final six items of the SMQ measure level of interference/distress and these items were not included in the factor score nor were they considered in this study. A lower total score indicated less speaking behaviour. Studies showed that the SDQ factor scores had acceptable to high internal consistency, and good construct and incremental validity (Bergman et al., 2008; Letamendi et al., 2008). Cronbach α 's for the SMQ Total and subsequent measures are reported in Table 3.

Social Anxiety Scale for Children – Revised/Parent Version (SASC-R/P) (La Greca, 1999) is a 22-item parent measure of the child's social anxiety in their relationship with peers. Eighteen of the items group to form three subscales: SASC—R/P:New (social avoidance and distress with new and less familiar social people), SASC-R/P:FNE (fear of negative evaluation or response by peers) and SASC-R/P:General (social avoidance and distress generally when interacting with others). The parent rates how true each statement is for their child on a five-point Likert scale from 1 (not at all) to 5 (all the time). Items in the SASC-R/P:New and SASC-R/P:General asked about speaking. As these items could potentially inflate the score for SM children, a decision was made to calculate these subscale scores without speaking items. The SASC-R/P:General subscales could not be calculated as only one item remained in the subscale. Thus, only the SASC-R/P:FNE and SACS-R/P:New (ns) were included in the analyses for this study, and the subscales were summed to form the SASC-R/P Total score. The SASC-R/P has been shown to have good internal consistency (SASC-R/P:New $\alpha = .87$, SASC-R/P:FNE $\alpha = .91$, SASC-R/P:General $\alpha = .78$) and moderate convergent validity with other measures of anxiety (La Greca, 1999).

The *Revised Preschool Anxiety Scale (PAS-R)* (Edwards, Rapee, Kennedy & Spence, 2010) is a 28-item questionnaire that measures the presence of anxiety symptoms in children from age three years. The items load onto four factors consistent with DSM-IV diagnoses: social anxiety, generalised anxiety, separation anxiety and specific fears. Parents rate each item from 0 (not at all true) to 4 (very often true). Given the high frequency of “talking” items in the Social scale, this factor was excluded. The three remaining factors were summed to form a PAS-R Non-social Total Score. The PAS-R has acceptable internal consistency, strong stability over a 12-month period and moderate to high construct validity (Edwards, et al., 2010).

(3) Parent-child interaction:

New Friends Vignette (NFV) (McShane & Hastings, 2009) is a parent-report measuring how parents respond to their child’s initial shy reaction in a novel social interaction. The scale specifically measures displays of overprotective parenting, critical control and appropriate support. The scale contains two hypothetical social situations, and each scenario contains 27 items. The parent rates on a three-point scale, from 0 (no) to 2 (yes), the likelihood they might have specific thoughts, vocalise particular statements or, display certain actions in their interaction with their own child, the other child and the adults. The responses are summed to provide scores on three subscales that measure parenting behaviour (i.e. overprotective, critical control and appropriately supportive); and three subscales that measure parent’s schema regarding the child’s display of reticence (i.e. Parent-centered/frustration, Child-centered/autonomy-extra support and Relationship-centered/solicitous). Psychometric properties are available for the three subscales that measure Parenting behaviour (McShane & Hastings, 2009). The Parenting subscales have been shown to possess good internal consistency and one-year

test-retest reliability, and moderate convergent validity (McShane & Hastings, 2009). The Cronbach α 's for the NFV parenting and parent schema subscales in the present sample are reported in Table 3.

Behavioural Observation of the parent and child's verbal and nonverbal behaviour was conducted in an outpatient hospital setting and video-taped. The child participated in a series of tasks in their parent's presence and these are reported in a broader study (Milić & Rapee, 2015). One task was included in the behavioural observation that was specific to the current study. The task was a challenging puzzle and children were given five minutes to complete the puzzle. The parent was instructed to "just give your child as much help as you think she/he needs" (Hudson & Rapee, 2001). The clinician was not in the room while the child completed the puzzle. Puzzles differing in complexity were used for preschool and school aged children. The mother's behaviour was rated on three global scales, and each scale contained a nine-point continuum ranging from zero to eight, where four represented a neutral point on the scale (Hudson and Rapee, 2005). The three dimensions were: (a) general degree of involvement (0 = very encouraging to 8 = very critical), (b) unsolicited help (0 = no help at all to 8 = very overintrusive), and (c) response to child (0 = very encouraging to 8 = very critical). Latency for the child to solicit assistance from the parent and number of times the child solicited help were also counted. The first author (M.I.M.) rated the videotapes. A second rater blind to group membership rated 30% of the videotapes. Mean intra-class correlations (ICC, two-way mixed) were high, ranging from .98 to 1.0.

Procedure

Ethics approval for this study was granted by: Western Sydney Local Health Network, Macquarie University Human Research Ethics Committee, NSW Department of Education and Communities, and Catholic Education Diocese of Parramatta. The parents provided informed written consent for their own and their child's involvement in the study. Each parent completed a package of questionnaires. The child's mother then participated in the semi-structured diagnostic interview. Following this, the child and their mother attended the clinic for the Behavioural Observation. Parents of children in the SM and SP group received a clinical report with recommendations for support services. Families in the control group received a shopping voucher as reimbursement for their time.

Statistical Analysis

Less than 5% of the data were missing, most often due to item nonresponse. Eight families dropped out of the study (SM = 2, SP = 2, C = 4) because of the time commitment required. The SP-12 Total and LSAS Total score were converted into z-scores and the mean of these z-scores formed the Social Phobia (SP) composite score. Parent responses on the NFV Child-centered schema subscale and father's responses on the NFV Relationship-centered schema subscales were excluded from the analyses because of the extremely low internal inconsistency of these subscales (Cronbach α between .08 to .39). The Fischer's Exact Test and one-way Analyses of Variance (ANOVA) were used to compare the groups on demographic characteristics. This revealed significant between group differences for mother's education and child's age. The analyses were run separately for mothers and fathers. The univariate analyses for

mothers were computed with maternal education and child age as covariates, and for fathers univariate analyses were run with only child's age as a covariate. Mothers' education and the child's age did not have a significant effect on the results for questionnaires, therefore one-way ANOVA was used for these analyses. When ANOVA revealed a significant difference between groups, independent sample t-tests were conducted to identify between which groups the difference lay. Pearson's product-moment correlation coefficient was used to measure the strength of the relationship between parent social phobia symptomatology, children's anxiety and mutism, and parent-child interaction. The correlations were examined separately for each group (i.e. SM, SP and non-clinical controls) and parent. The Bonferroni correction was not applied as this was an exploratory study aimed at identifying possible directions for future investigation.

RESULTS

Demographic variables:

The parent groups did not differ significantly on most demographic variables including: age, marital status, occupation, employment, reading ability, family income and paternal education. There was a significant between-group difference on mother's education, $X^2(8, N=57) = 18.01, p = 0.009$ (see Table 1). Post-hoc comparison revealed a significant difference between mothers in the SM and C group, $X^2(4, N=40) = 15.50, p = 0.001$, with 72% of SM mothers compared to 100% of C mothers having completed further education beyond high school. No statistically significant difference was found between mothers in the SM and SP groups, $X^2(4, N=42) = 5.39, ns$.

Demographic information about the children is reported in Table 1. The children in the three groups were not matched for gender or age. There were more female participants, and this did not differ significantly across groups, $X^2(2, N = 57) = 3.48, ns$. There was a statistically significant difference between groups on child's age, $F(2,56) = 3.84, p = 0.03$.

Parent symptomatology: No significant between group differences were identified on the DASS Total score for either mothers, $F(2,54) = 0.57, ns$, or fathers, $F(2,47) = 0.97, ns$. For the SP-12 composite score, no main effect for group was identified for either mothers, $F(2,54) = 0.69, ns$, or fathers, $F(2,48) = 0.04, ns$.

Parenting behaviour: A statistically significant difference was found between groups on the NFV Parent-centered schema subscale for mothers, $F(2,53) = 3.38, p = 0.04$, and fathers, $F(2,49) = 4.19, p = 0.02$ (see Table 3). Mothers and fathers of children in the SM group reported more NFV Parent-centered thoughts of frustration with their child's reticence than mothers, $t(37) = 2.56, p = 0.02$, and fathers, $t(35) = 3.12, p = 0.004$, of nonclinical controls. Mothers, $t(39) = 0.38, ns$, or fathers, $t(36) = 1.17, ns$, of children in the SM and SP groups did not differ significantly on their responses on the NFV Parent-centered thoughts subscale. Mothers, $t(30) = 1.59, ns$, and fathers, $t(27) = 1.55, ns$, of children in the SP group and non-clinical controls also did not differ significantly in their responses on the NFV Parent-centered schema subscale. Furthermore, no between groups difference was found for mother's responses on the NFV Relationship-centered subscale, $F(2,53) = 0.42, ns$.

Table 3: *Parent's self-report, parenting (NFV) and parent-report measures about the child.*

Measure	Mothers				Fathers			
	Cron- bach α	SM (n=25)	SP (n=17)	C (n=15)	Cron- bach α	SM (n=22)	SP (n=14)	C (n=14)
		Mean (SD)	Mean (SD)	Mean (SD)		Mean (SD)	Mean (SD)	Mean (SD)
DASS Total	.94	25.05 (22.80)	19.94 (18.22)	18.53 (19.44)	.93	16.33 (17.29)	21.43 (21.96)	12.71 (6.60)
SP Composite score	-	0.07 (0.86)	0.10 (1.23)	-0.26 (0.74)	-	-0.02 (0.91)	0.05 (1.11)	-0.04 (0.85)
NFV Parent-centred schema	.77	5.42 (2.92) _a	5.06 (3.11) _{ab}	2.93 (3.03) _b	.84	5.04 (3.05) _a	3.80 (3.44) _{ab}	2.14 (2.11) _b
NFV Child-centred schema	.39	8.13 (2.44)	9.06 (1.68)	9.40 (1.24)	.08	8.00 (1.54)	8.67 (2.06)	8.86 (1.46)
NFV Relationship-centred	.61	5.31 (2.15)	4.65 (3.06)	4.87 (1.64)	.28	5.43 (2.46)	4.80 (1.74)	4.24 (1.43)
NFV Overprotection	.74	13.68 (3.42)	13.00 (5.69)	12.43 (3.96)	.73	13.12 (4.54)	13.33 (4.27)	11.57 (2.74)
NFV Critical Control	.84	6.04 (4.29)	5.07 (4.81)	2.73 (3.94)	.74	6.11 (3.49)	4.73 (4.56)	3.64 (3.27)
NFV Appropriately Supportive	.76	17.37 (5.08)	18.58 (3.95)	18.07 (2.99)	.77	16.63 (4.63)	16.93 (4.86)	15.64 (4.01)
SMQ Total	.96	0.84 (0.33)	1.52 (0.48)	2.37 (0.32)	.97	0.88 (0.38)	1.54 (0.55)	2.34 (0.27)
SASC-R/P: Total	.94	36.42 (9.27)	35.44 (8.51)	22.33 (6.02)	.94	35.55 (9.37)	34.00 (9.20)	21.00 (15.4)
PAS-R Non-social Total	.91	33.18 (17.06)	35.74 (16.65)	23.13 (10.95)	.89	33.27 (16.28)	32.20 (15.25)	21.54 (7.08)

Note: Means with different subscripts are significantly different at $p=0.05$.

With regard to parenting behaviour on the NFV, no between group differences were found for mothers in the level of overprotection, $F(2,53) = 0.40$, *ns*, critical-control, $F(2,53) = 2.68$, *ns*, or appropriately-supportive parenting behaviour, $F(2,53) = 0.41$, *ns*. Differences were also not found between groups on the fathers' responses on the overprotection, $F(2,49) = 0.85$, *ns*, critical-control, $F(2,49) = 1.93$, *ns*, and appropriately-supportive subscales, $F(2,49) = 0.33$, *ns*, of the NFV.

Mother-child interaction: Observation of mother's behaviour during the Puzzle task in the clinic revealed no main effect for group in the mother's affective response to their child, $F(2,46) = 2.79$, *ns*, $\eta_p^2 = .11$, the level of unsolicited help they provided their child, $F(2,46) = 2.45$, *ns*, $\eta_p^2 = .10$, or how involved they became in helping their child complete the puzzle, $F(2,46) = 2.32$, *ns*, $\eta_p^2 = .11$. There was also no main effect for group in the latency of time (in seconds) for the child to seek help from their mother, $F(2,46) = 0.54$, *ns*, $\eta_p^2 = .02$, or the number of times they asked their mother for help, $F(2,46) = 0.03$, *ns*, $\eta_p^2 = .00$ (see Table 4).

Table 4: *Mother-child interaction on the puzzle task.*

Variable		SM (n=25)	SP (n=17)	C (n=15)
		Estimated Mean (SE)	Estimated mean (SE)	Estimated mean (SE)
Mother	(1) Degree of involvement	2.23 (0.50)	3.50 (0.55)	2.01 (0.64)
	(2) Unsolicited help	2.09 (0.51)	3.20 (0.55)	1.36 (0.65)
	(3) Response to child	2.97 (0.30)	2.25 (0.32)	1.81 (0.38)
Child	(1) Latency (in sec.) seek help.	156.23 (31.18)	179.14 (33.82)	211.15 (39.66)
	(2) Solicited help (frequency).	1.62 (0.62)	1.37 (0.67)	1.51 (0.79)

Association of mother's social phobia and parenting with child mutism and anxiety: With regard to the link between maternal anxiety and child anxiety, the mother's SP composite score correlated significantly with the mother's report of their child's social anxiety on SASC-R/P in the SM group ($p = .005$) and C group ($p = 0.02$), and there was a moderate non-significant positive association for the SP group (see Table 5). Mother's SP composite score also correlated significantly with the child's PAS-R Non-social total score for the SM group ($p = .04$), and the correlations with the SP and C group were in the positive direction but non-significant.

With regard to parenting and child anxiety, a significant positive correlation was found between NFV Relationship-centered schema and the SM PAS-R Non-social total score ($p = 0.02$). A significant positive correlation was found between the mother's NFV Overprotection score and the child's SASC-R/P score for only the C group ($p = 0.02$) (see Table 5). In the observational task (see Table 5), a significant positive correlation was found between the mother's affective response to the child during the puzzle and the child's SMQ Total score for the SM ($p = 0.02$) and the SP groups ($p = 0.02$). There was a significant negative correlation between the child's level of social anxiety on the SASC-R/P and the mother's affective response to the child for the SM group ($p = 0.04$). The child's level of social anxiety correlated negatively with unsolicited help from mothers in the SP group ($p = 0.04$).

Finally, with regard to parenting and mother's level of social anxiety, a significant positive correlation was found between NFV Relationship-centered schema and the mothers SP composite score in the SP and C groups ($p = 0.01$ for both groups). For the children in the C group, their mother's SP composite score was also significantly positively correlated with NFV Parent-centered schema ($p = 0.005$) and NFV Critical control ($p = 0.01$).

Association of father's social phobia and parenting with child mutism and anxiety: No significant associations were found between the father's level of social anxiety and the child's mutism and anxiety scores (see table 6). One significant association was found between parenting and child mutism and anxiety (see Table 6). In the SM group, a significant negative association was found between the father's NFV Relationship-centered schema score and the child's SMQ Total score ($r = -.45$, $p = 0.03$). Finally, with regard to parenting and the father's level of social anxiety, a significant positive association was found between the father's report on the NFV for Critical control and their SP composite score in the SM group only ($p = .52$).

Table 5: Association between mother's anxiety, mother's rating of their child's anxiety and mutism, and parenting behaviours.

Measure	SM (n = 25)				SP (n = 17)				C (n = 14)			
	1	2	3	4	1	2	3	4	1	2	3	
Mother SP Composite Score	.19	.56*	.41*	-	-.22	.40	.17	-	-.18	.59*	.44	-
NFV Parent centered	.34	-.23	-.03	.23	-.23	.16	.27	.01	-.07	.44	.41	.69*
NFV Child centered	-.09	.07	.25	.19	.50*	-.11	.04	.03	.19	-.28	-.17	.16
NFV Relationship centered	-.26	.30	.48*	.33	-.14	.16	.29	.58*	.24	.37	.26	.58*
NFV Over-Protection	-.24	.23	.38	.16	.27	.02	-.05	.13	-.19	.58*	.32	.31
NFV Critical Control	.30	-.25	-.21	.34	.10	-.04	.07	-.09	-.41	.27	.09	.64*
NFV Appropriately supportive	.12	.04	.06	.29	.16	.09	.13	-.12	.04	-.41	-.44	-.28
Puzzle Response to child**	.49*	-.47*	-.27	-.31	.55*	-.01	-.37	.13	-.26	.12	-.02	.51
Puzzle Unsolicited Help**	-.30	-.14	-.04	.04	-.05	-.52*	-.31	.11	-.19	-.13	-.04	-.24
Puzzle Degree of involvement**	-.32	-.10	-.07	.08	-.15	-.39	-.19	.06	-.26	-.01	-.02	-.28

1 = SMQ Total score, 2 = SASC-R/P Total score, 3 = PAS-R Non-social Total,

4 = mother's SP composite score

* $p < 0.05$

** n = 21 for SM group, n=13 for C group.

Table 6: Association between father's anxiety, father's rating of their child's anxiety and mutism, and parenting behaviours.

Measure	SM (n = 23)				SP (n = 14)				C (n = 14)			
	1	2	3	4	1	2	3	4	1	2	3	4
Father SP Composite Score	.02	.32	.25	-	.05	-.15	-.21	-	-.27	-.39	-.14	-
NFV Parent-centered Schema	.02	.19	-.07	-.34	-.50	.27	.28	.08	.07	-.14	.01	.12
NFV Child-centered Schema	.11	-.19	-.19	-.06	.15	.17	-.35	-.49	-.29	-.11	-.03	.35
NFV Relationship centered schema	-.45*	.29	.31	-.12	.03	-.18	-.19	.16	-.16	.08	.25	.03
NFV Over-protection	-.27	.09	.32	.11	.17	-.19	.10	.23	.24	-.29	.02	-.29
NFV Critical control	.05	.12	-.03	.52*	-.47	.17	.48	-.27	-.15	.44	-.31	-.18
NFV Appropriately supportive	.15	.43	-.09	.18	.26	-.14	.07	-.14	.45	-.49	-.18	.18

1 = SMQ Total score, 2 = SASC-R/P Total, 3 = PAS-R Non-social Total,

4 = father's SP Composite score

* $p < 0.05$

Discussion

This present study had three objectives: (1) to identify whether parents with children with SM differed in their levels of distress and social phobia symptomatology relative to parents of children with SP and parents of non-clinical controls, (2) to explore whether parents of a child with SM, SP and non-clinical controls differed in the way they parented their child, and (3) to identify if differences in parenting were related to differences in the child's or parent's levels of social phobia symptomatology. Exploring how the two clinical groups differed is complicated as the majority of children with a primary diagnosis of SM in this study had a comorbid diagnosis of SP, a finding that is consistent with previous SM research (Black & Udhe, 1995; Chavira et al. 2007; Dummit et al., 1997; Mannassis et al., 2007; Kristensen, 2000). SM and SP are distinct anxiety disorders in the DSM-5 (APA, 2014), and group status was clear as the sole symptom of SM, the prolonged failure to speak, defined which group children with social anxiety were assigned to. Excluding children with a comorbid diagnosis of SP from a SM sample would not be a valid representation of children with SM. However, the high comorbidity of SP in SM creates a fundamental difficulty in interpreting the findings as the comparison is between children with a primary diagnosis of SM combined with social anxiety and/or a comorbid diagnosis of SP (children with SM) and children with SP without a current comorbid diagnosis of SM (children with SP). The comparison between the two clinical groups and how they compare to non-clinical controls remains important to furthering our understanding of SM, and in particular, understanding why a subset of socially anxious children fail to speak for prolonged periods of time. As predicted, where differences were found in the study, the pattern of

findings for families of SM children were reflective of the findings of previous research on childhood anxiety.

Mothers and fathers of children with SM did not differ significantly from mothers and fathers of children with SP and nonclinical controls in their level of negative emotion or social phobia symptomatology, however, the mean social phobia composite score for mothers was in the direction predicted. The mean group DASS total scores for all groups was within one standard deviation of the mean score for the Australian adult population norms, indicating that level of reported negative emotion was not higher than for the general population (i.e. mean DASS Total score = 16.48; Crawford, Cayley, Lovibond, Wilson and Hartley, 2011).

Consistent with previous research showing that anxiety runs in families (Rapee et al, 2009), the mother's report of her own social phobia was significantly and positively associated with her report of her child's level of social phobia in the SM and the nonclinical control groups, and this association was positive but non-significant in the SP group. The most probable reason for the non-significant association between mother's report of her own social phobia and the child's level of social phobia in the SP group is insufficient power as previous studies with larger samples found a significant positive association (Bögels et al., 2010; Hirshfeld-Becker, 2010; Ollendick & Hirshfeld-Becker, 2002). No significant associations were found between the father's social phobia symptomatology and their child's social anxiety. The pattern of results for fathers in the nonclinical control and SP group (i.e. negative association) were unusual and inconsistent with research about the genetic vulnerability to anxiety. The most likely explanation for this pattern of associations and the failure to find an association is the small subgroup samples (C = 14 and SP = 14 fathers), in which there was a very small scatter of difference in the father's social phobia composite scores and a larger

scatter of difference in the child's anxiety score. The father's scores for their child's social anxiety corresponded with mother's reports of the child's social anxiety.

Unlike Chavira et al. (2007) who found a positive relationship between SM severity and the odds of having a parent with SP, the current research did not find a significant association between the child's level of mutism and the mothers' or fathers' levels of social phobia symptomatology. Speaking interactions are identified by children and adolescents with social phobia as the situation that is most anxiety provoking and the situations they most avoid (Rao, Beidel, Turner, Ammerman, Crosby & Sallee, 2015). One would expect having a child fail to speak to an adult would draw attention to the child and the parent(s) and in clinical practice the parents frequently reflect a fear of being negatively judged because of their child's failure to speak. This together with the genetic overlap for anxiety would suggest a positive association between level of mutism and social anxiety. The small sample size in each subgroup is the most probable explanation at this point for the failure to find an association particularly given Chavira et al (2007) SM sample was three fold larger than the sample in this study.

The second aim of this study was to explore if there were differences in parenting behaviour. Chavira et al. (2007) had revealed that mothers of children with SM made more high power remarks and granted their child less autonomy than mothers of anxious and non-anxious children. In the current study, the NFV questionnaire responses did not reveal any significant group differences between mothers or fathers in the likelihood that they would respond to their child's social reticence with overprotection, critical control, or a positive and appropriate response. The mean score for mothers and fathers on the critical control and overprotection subscales of the NFV were in the direction predicted by Chavira et al. (2007) study, with SM parents scoring

higher. Similarly, in the laboratory observation of the mother-child interaction during a puzzle task, a significant difference was not found between the SM, SP and nonclinical controls on the clinician's ratings of the mother's affective response to the child, amount of unsolicited help or parental involvement during the task. Given the children sought minimal assistance and there was a large latency in their request for help, it is most probable that the task was not challenging enough.

While their parenting behaviour did not differ significantly, a significant difference did emerge in the types of internal thoughts parents had about their child's reticent behaviour. Mothers and fathers of children with SM acknowledged more thoughts of frustration with their child's reticent behaviour (e.g. *"I just want him/her to stop this and play nicely"*, *"I wish he/she would just go play with them"*, *"Now what am I supposed to do?"*) than mothers and fathers of nonclinical controls. Whilst parents of children with SP did not differ significantly from either SM or clinical controls, the mean score for mothers of children with SP was closer to mothers of SM children than non-clinical controls. Thus, there was less warmth in the way that parents of SM children thought about their child's behaviour. It is interesting then that the differences in their parenting behaviour had not reach significance. One could postulate that the awareness they were being observed, and the anecdotal evidence in therapy of parents reporting that firm limits and critical commentary were ineffective in lowering their child's hesitancy, meant that they did not overtly show their frustration. An alternative explanation is that the frustration does show but in more subtle ways (e.g. tone, posture and language) that are difficult to detect on questionnaires measures. This would be important to investigate in future studies as research shows that highly anxious children are attentive to the subtle cues of their parents (Bögels, Stevens & Majdandžić, 2011). It

is also highly probable that the frustration emerges at home in their conversations with their child about the mutism.

The third goal of this study was to explore across the three groups whether differences in parenting were related to the child's anxiety and mutism, and alternatively, whether differences in parenting were associated with the parent's anxiety. Previous findings of a modest negative association between parental warmth and child anxiety (McLeod et al, 2007; Hudson & Rapee, 2001) did not emerge in this study. No significant associations were found between severity of mutism or anxiety and critical control on the parenting measure. On the puzzle task a significant positive association was found between the clinician's rating of parent responsiveness (i.e. how encouraging or negative/critical the parent was towards the child) and the child's level of mutism and anxiety, suggesting an opposite finding to previous research. However, a closer examination of these results revealed 71% of mothers in the SM and SP groups and 85% of mothers in the non-clinical group were given a clinician rating of 3 or lower on the dimension "response to child". Thus, in the majority of cases there was a warm and positive mood between the child and their parent during the puzzle task. The ratings from 0 to 3 on the 'response to child' scale dimension differ on the amount of verbal encouragement the parent provides the child during the puzzle task. This finding of a positive association between level of parental encouragement and child anxiety and mutism is consistent with previous research on childhood anxiety that identified a link between increased parental protection and higher levels of child anxiety.

Some researchers have hypothesised that "parental encouragement of children's autonomy and independence (e.g. in novel contexts) may augment children's perceptions of mastery over the environment, leading to anxiety reduction" (McLeod et al, 2007: p. 156). However, excessive encouragement may inadvertently maintain

anxiety over time, as the child's confidence and perseverance in challenging tasks is associated with the presence and verbal encouragement communicated by the parent. In this study, there was a significant negative association between unsolicited help in the puzzle task by mother's of SP children and child social anxiety, and the remaining associations between child anxiety and maternal unsolicited help or over-involvement were in the negative direction. This suggests that higher child anxiety was associated with fewer protective behaviours by mothers in the puzzle observation. However, a significant positive association was found between verbal encouragement by mothers in the SM and SP groups and the child's severity of mutism, and between maternal encouragement in the SM group and higher social anxiety in the SM children. In contrast, in the non-clinical group, a negative (non-significant) association was found between frequency of maternal encouragement and severity of child mutism. Less encouragement for the non-clinical controls was one or two encouraging words in a 5-minute period and acknowledgement of the child's efforts at the end of the task. Thus, the mothers in the non-clinical group were appropriately protective, and in the clinical group, the link between higher parental overprotection and child anxiety may have emerged through their protective words.

The association between child anxiety and parental overprotection also emerged in the way they thought about their child's behaviour. Higher levels of non-social anxiety in children with SM was significantly associated with SM mothers' responses on the NFV Relationship-centered (solicitous) subscale which included items such as: *"Maybe we should just leave before he gets really upset"*, *"I don't think he is ready to play with a group like this"* and *"She/He just needs a little cuddle from me"*. The central theme is one of protecting the child from distress by considering withdrawal from the experience or offering extra encouragement/comfort. Furthermore, whilst the

remaining results were non-significant they were in the predicted direction: more solicitous thoughts were associated with higher social/non-social anxiety, across all three groups, and with more severe mutism scores only for the SM and SP groups. Thus, parents may differ more in the subtle ways of parenting and it would be important for future research to examine those aspects.

No significant associations were found between parenting by fathers and the child's anxiety or mutism ratings. There are several possible explanations for the failure to find any significant associations. Firstly, the associations found for mothers were predominately in the laboratory assessment and McLeod et al (2007) had identified that questionnaire measures were less able to detect differences in parenting relative to observational assessments. Secondly, as previously noted the small sample size of each subgroup meant there was not adequate power to detect differences that may be there.

Finally, the last aim of this study was to explore whether parental anxiety was associated with parental warmth and over-involvement. An association did emerge between higher parental anxiety and more solicitous thoughts about their child. In the non-clinical control group, a further significant association emerged between parental social anxiety and thoughts of frustration with the child's reticence, and between parental social anxiety and critical control. The associations for the SM group were in the same direction and are consistent with the parent's anecdotal reports of feeling at the same time concerned and frustrated with their child's mutism, and it may be that those times of mixed emotions are associated with more critical parenting which would need to be clarified by future observational studies. The remaining associations between parental anxiety and parenting were predominantly non-significant. One possible explanation for the failure to find an association is that the social situation in the questionnaire, whilst uncomfortable, did not elicit increased anxiety in the parent.

Additionally, it is highly likely that the questionnaire measure was not sensitive enough to detect differences in parenting behaviour that may be present in real life (McLeod et al, 2007) particularly if the differences are more subtle.

As noted, the primary methodological limitation in comparing children with SM to children with SP is the high comorbidity of SP in children with SM. A second limitation was the small sample size and the consequent impact on the power of the analyses to detect statistically significant differences and associations. The results are correlational so it is not possible to draw conclusions about the direction of any significant effects. Thirdly, potential limitations arise from the groups not being matched for age and gender, and sampling bias. All the families in the SM group, and most families in the SP group, were seeking clinical assessment and feedback on how to support their child. Level of parental anxiety may differ in families who have not sought treatment or who delay seeking treatment. Furthermore, the families in the control group were more educated parents who valued research. The fourth limitation is that the puzzle task did not appear to be sufficiently challenging for the child. Future studies would benefit from preliminary research to identify tasks that are challenging yet sensitive enough to detect differences. Finding potential tasks is complicated by the fact that the children will differ in age and ability. Given that research shows that observational measures are more sensitive to detecting differences in parenting (McLeod et al, 2007), and the recent finding that parental anxiety emerges in the “context of a specific, disorder-salient, challenge” (Murray et al, 2012), future studies should include social and non-social tasks. The fifth limitation is that parental anxiety was not assessed in the observational task, nor were the parents asked about their anxiety in the novel situation where their child was reticent. This would be important to include in subsequent studies to clarify if the failure to find a difference was associated

with the context and task. Finally, information about the parents' emotional health and parenting behaviour was predominately based on self-report instruments. Future studies might be enhanced by including more objective semi-structured clinical assessment of the parent's emotional health.

Despite these shortcomings, a strength of this study was the inclusion of an observational measure of parent-child interaction, and the comparison of children with SP in light of the questions in the literature about the commonalities between SM and SP (Muris & Ollendick, 2015). Whilst this study did not reveal any between group differences in negative mood, social phobia symptomatology or parenting behaviour, the study did find that parents of children with SM reported more thoughts of frustration with their child's displays of social reticence. Associations between child anxiety and parenting emerged in the verbal encouragement provided by parents and parent's internal thinking about the child's behaviour. Similarly, associations between parental anxiety and parenting emerged in their thoughts about the behaviour. The internal frustration may be associated with more subtle differences in parenting, which the anxious child may be attuned to. Thus, future observational studies should include more refined measures of verbal overprotective behaviours as well as measures of internal cognition when exploring differences in parenting. In clinical practice, parents may benefit from being given the space in treatment to reflect on their thoughts (e.g. concern, frustration) about their child's social fearfulness and to think through how this impacts the way they interact with their child in the community as well as at home.

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CHAPTER 5

SUMMARY AND CONCLUSIONS

Selective Mutism (SM), a childhood disorder with a mean age of onset in the preschool years, was reclassified as an anxiety disorder in 2014 with the publication of the *Diagnostic and Statistical Manual of Mental Disorders, Fifth edition* (DSM-5) (APA, 2014). This decision was based on the strong body of evidence collected over the preceding 20 years that showed that children with SM are anxious and their anxiety is predominately related to social interactions with adults and children outside of their immediate family (Bögels, Alden, Beidel, Clark, Pine, Stein & Voncken, 2010; Muris & Ollendick, 2015). Many children with SM are diagnosed with comorbid Social Phobia or Avoidant Disorder of Childhood and Adolescence and they are frequently described as shy and inhibited (Anstendig, 1999; Bögels et al. 2010). Furthermore, studies indicate that there is a higher incidence of social anxiety related disorders (i.e. Social Phobia, Avoidant Personality Disorder) (Chavira, Shipon-Blum, Hitchcock, Cohan & Stein, 2007; Black & Udhe, 1995) and personality constructs that are strongly related to social anxiety (i.e. shyness) in family members of children with SM (Bögels et al. 2010; Muris & Ollendick, 2015). The “significant, albeit not perfect, overlap between SM and” SP (Bögels et al. 2010: p.2010) prompted researchers to question whether SM is a distinct anxiety disorder or a variant of SP (Bergman, Piacentini & McCracken, 2002; Black & Udhe, 1992, 1995; Bögels et al. 2010; Chavira et al., 2007; Dummitt, Klein, Tancer, Asche, Martin & Fairbanks, 1997). Evidence to date on this question is relatively scarce. The thesis sought to clarify the relationship between SM and SP by systematically comparing younger children aged four to eight years with a primary diagnosis of SM to children with a primary diagnosis of SP and a non-clinical control group. Information was collected from mother-, father-, teacher- and child-reports, a semi-structured diagnostic interview with the child’s mother, and an observational assessment of the child with their mother present.

The first paper of this thesis reported on the development of the Selective Mutism Questionnaire-Child (SMQ-C), a 20 item self-report measure of frequency of speaking that can be completed by children from age four years. The three-factor structure of the adult measure, the Selective Mutism Questionnaire (Bergman, Keller, Piacentini & Bergman, 2008), was retained. Examination of the psychometric properties revealed that the three-factor structure and the total SMQ-C scale were internally consistent, and the SMQ-C had good convergent and divergent validity. The study showed that young children are aware of their mutism and their description of how much they talk in different social contexts correlated significantly with parent- and teacher-reports of their mutism.

The second paper aimed to clarify the relationship between SM and SP by exploring the commonalities and differences in social phobia symptomatology and behavioural inhibition in children with SM and children with SP. The research showed that children with SM and SP did not differ in non-verbal social anxiety or non-social anxiety and both groups were more anxious than non-clinical controls. Parents rated children with SM as significantly more fearful of negative evaluation than non-clinical controls. On observational tasks, children with SM were more inhibited both verbally and nonverbally than children with SP and non-clinical controls. Both clinical groups had fewer friends than non-clinical children and they experienced difficulties forming friendships. Children with SM also experienced greater difficulties sustaining friendships.

The third and final paper was an exploratory study examining the level of negative affect, social phobia symptomatology and parenting in mothers and fathers of children with SM, SP and non-clinical controls. No significant differences emerged between the groups with one exception. Mothers and fathers of SM children reported

more internal thoughts of frustration with their child's reticent behaviour than non-clinical controls. This third paper also explored the association between parent and child anxiety and whether differences in parenting were associated with child anxiety and mutism or parental social anxiety. Significant associations were found between level of verbal encouragement provided by mothers and child mutism and anxiety in the two clinical groups. Furthermore, associations were found between child anxiety and maternal solicitous thoughts about the child's reticent behaviour, and between maternal social anxiety and internal thoughts of solicitousness and frustration with the child's reticence.

The current research extends our knowledge about the quality of the social anxiety experienced by children with SM and the relationship between SM and SP. In previous research, clinicians had rated children with SM as more socially anxious than children with SP (Yeganeh, Beidel, Turner, Pina & Silverman, 2003; Yeganeh, Beidel & Turner, 2006; Young, Bunnell & Beidel, 2012), while in a majority of studies, parents, teachers and children reported similar levels of social anxiety between children with SM and SP (Carbone, Schmidt, Cunningham, McHolm, Edison, St.Pierre, & Boyle, 2010; McInnes, Fung, Manassis, Fiksenbaum & Tannock, 2004; Manassis, Fung, Tannock, Sloman, Fiksenbaum & McInnes, 2003; Yeganeh et al., 2003, 2006; Young et al., 2012). These studies had not excluded speaking items from the measures of social anxiety prior to analysis. Some researchers had questioned whether adults were inferring higher social anxiety in children with SM (Carlson, Mitchell & Segool, 2008; Krohn, 1993; Yeganeh et al., 2003; Young et al., 2012). Questions were also raised about whether children with SM were underreporting their level of social anxiety (Bergman, Ford, Sladeczek & Carlson, 1998; Yeganeh et al., 2003). With the exclusion of speaking items, this study corroborates previous adult reports that children with SM

and SP experienced similar levels of non-verbal social anxiety. The study also revealed that in a novel social situation with an unfamiliar person, children with SM took significantly longer both to spontaneously speak and gesture relative to children with SP and non-clinical controls, hence pointing to higher levels of overall social anxiety, even in the nonverbal realm, among children with SM. This finding provides the first empirical evidence of a link between SM and Behavioural Inhibition (BI). The finding that children with SM were more non-verbally inhibited could account for previous clinician ratings of higher observed anxiety in children with SM (Young, Bunnell & Beidel, 2012). It also suggests that children are under-reporting their level of anxiety, as restricted speech may provide a behavioural avoidance strategy. Importantly, it questions the DSM-5 claim that children with SM do not experience fear of negative evaluation in non-speaking interactions (APA, 2014) as the higher behavioural inhibition was observed in interactions that did not require speech.

The findings in this thesis raise questions about the classification of SM. The results provide support for the idea that SM is a more severe form of SP observed in younger children, and the mutism “is a more natural form of social avoidance for younger children” (Bögels et al. 2010). Support for this position comes from the finding that children with SM were more behaviourally inhibited in new and less familiar social situations than children with SP and non-clinical controls (Milic & Rapee, 2015). Furthermore, the non-verbal social anxiety experienced by children with SM is at a similar point on the social anxiety continuum as children with SP. The select situations in which the mutism is most evident (i.e. initiating and joining into conversations, speaking with new and less familiar people, asking teacher a question) are the situations that children and adolescents with SP identify as the most anxiety provoking and the situations that they most avoid (Rao, Beidel, Turner, Ammerman, Crosby & Sallee,

2007). Children with SP fail to speak as frequently as children with SM in new and unfamiliar public situations, and speaking in children with SP continues to be restricted in familiar situations (e.g. school) (Milic & Rapee, 2015). Furthermore, based on parent report, children with SM were significantly more fearful of negative evaluation than non-clinical controls and they did not differ significantly from children with SP (Milic & Rapee, 2015).

The research on information processing biases in people with SP provides a way of understanding why some children may continue to avoid speaking. Firstly, the initial silence and wariness draw increased attention to the child. Children with higher shyness have a greater dislike for being the centre of attention (Bishop, Spence & McDonald, 2003). The continued lack of speech frequently results in less positive reactions, and on occasion, overtly negative reactions. Anecdotally children report being asked by peers, “Why don’t you talk?”, and being reprimanded for their failure to respond (Roe, 2015). Parents of children with SM identified internal thoughts of frustration with their child’s reticent behaviour (Milic & Rapee, 2015). Research shows that socially anxious adults “interpret these mildly negative social events in a catastrophic fashion” (Clarke & McManus, 2002). It is thus, highly probable that young children would similarly interpret comments, particularly from adults, as evidence of a significant negative consequences should they talk (e.g. being yelled at, laughed at, getting into trouble for saying the wrong thing). Secondly, the children’s responses on the self-report questionnaire clearly showed that they were very aware of their mutism. Anecdotal clinical evidence reveals children are also aware of how others see them through the comments they hear (e.g. “They don’t talk”, “They can’t talk”), questions they are asked (e.g. “Can you talk?”, “When are you going to talk?”), and the conversations they have with their parents and teachers about needing to talk. Thinking and talking about

speaking behaviour in anticipation of an event or after a social event is likely to lead to greater avoidance behaviours (Clarke & McManus, 2002). Thirdly, some of the fears are associated with real difficulties. Just like children with SP, children with SM had fewer friends and greater difficulties making friends however, children with SM also had difficulties sustaining friendships. Children with SM may interpret the increased friendship difficulties as indicative of negative evaluation by others (e.g. not liked) (Clarke & McManus, 2002). Furthermore, research shows an increased incidence among children with SM of speech and language difficulties, abnormalities in auditory efferent feedback pathways, and a different primary language spoken at home (Anderson & Thomsen, 1998; Bergman, Gonzalez, Piancentini & Keller, 2013; Black & Udhe, 1995; Bradley and Sloman, 1975; Brown and Lloyd, 1975; Elizur and Perednik, 2003; Ford, Sladeczek & Carlson, 1998; Henkin & Bar-Haim, 2015; Klein, Armstrong and Shipon-Blum, 2012; Kristensen, 2000; Kumpulainen et al, 1998; McInnes et al., 2004; Steinhausen & Juzi, 1996). Perceived and/or real differences in the way their voice sounds and language skills may heighten anxiety in situations in which speech is anticipated and/or expected. The heightened anxiety leads to an increase in self-focussed attention and the likelihood of misinterpreting the internal experience “as evidence that they are performing badly” (Clarke & McManus, 2002). Collectively, these enhanced threat appraisals of speaking situations generated by information-processing biases maintain the safe behaviour of not speaking. Given speech is an essential part of the social interaction, the failure to speak will result in a greater functional impairment than restricted speech.

The findings have meaningful implications for treatment of SM. The research shows that SM is clearly a social anxiety based disorder. Therefore, many of the well-supported techniques used to manage social anxiety in children (i.e. emotion

management, thought challenging and parent education about anxiety and parenting an anxious child) (Rapee, Schniering & Hudson, 2009) should be included in the treatment together with the exposure-based practice. Furthermore, children are likely to benefit from the exposure-based practice being extended to include play-based social skills practice (Coplan, Schneider, Matheson & Graham, 2010) to build confidence in applying the social skills required for making and keeping friends.

The limitations of the present study provide guidance and ideas for future research. Multi-site studies of SM would address the continued limitation of small sample sizes, as this study like previous studies found trends or consistent patterns in the scores that hinted at differences that did not reach significance. This could be extended to include a longitudinal study of children commencing preschool who have failed to speak for a defined period of time that is less than the required two months for a diagnosis of SM. Assessment of these children across time will provide new insights into why some continue to fail to speak and others find their voice a little. Furthermore, the finding that children with SM are more fearful of negative evaluation is based on parent report and the hypothesis that information-processing biases may contribute to the maintenance of the mutism is drawn from research on SP in adults. Direct assessment of the cognitions of children with SM and SP in social anxiety provoking situations as well as studies exploring how they interpret external social events would provide the first direct evidence of how children with SM think. Finally, pre- and post-treatment assessment of the children's receptive and expressive language skills would clarify if the observed differences in expressive narrative language were impacted by the heightened levels of social anxiety.

Adults with social phobia have a low help seeking rate (Hirsch & Clarke, 2004). SM is clearly a social anxiety based disorder. The prolonged failure to speak provides a

clear signal of risk. Early identification and referral of children who are selectively mute promotes timely intervention for Social Anxiety Disorder. Early intervention of their social anxiety will enhance their social confidence and comfort, and optimise their long-term functioning in the social, interpersonal, academic and vocational spheres of life.

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APPENDIX A

SELECTIVE MUTISM QUESTIONNAIRE – CHILD (SMQ-C)

SMQ-C PICTORIAL RESPONSE SCALE

SAMPLE INDIVIDUAL ITEMS OF THE SMQ-C

Selective Mutism Questionnaire – Child Report.

(This has been adapted by the Principal Researcher, Maria Milic, from the Selective Mutism Questionnaire and the School Speech Questionnaire. The author of the original questionnaire has given the Principal Researcher permission to develop a child report version).

Instructions:

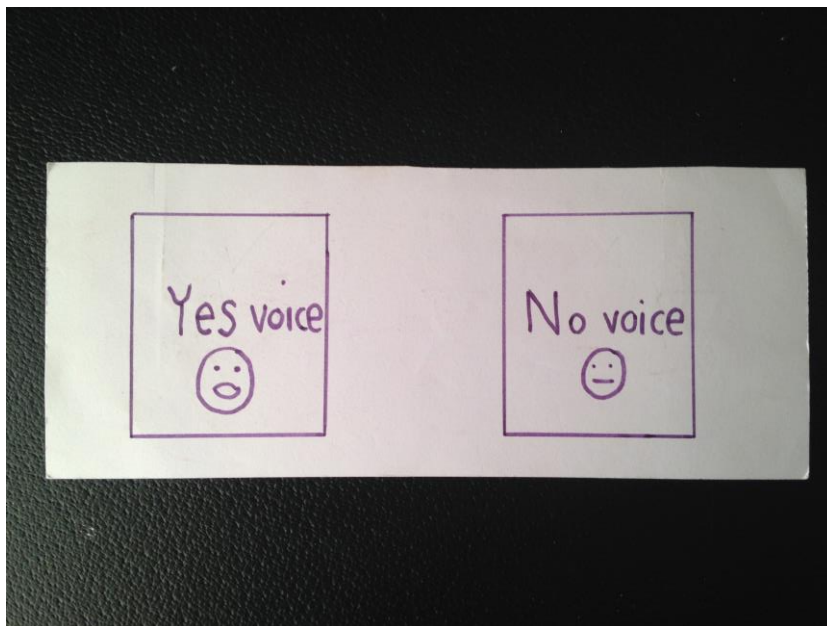
1. Place the rating scale in front of the child and explain to the child the process of responding. Explain that the child can respond by pointing (to the picture on the scale that corresponds), using their head (can gesture yes and no, nod or shake their head), or using their voice.
2. Begin with at least 2 basic questions to practice the process of responding, this will help you ascertain that the child understands what is expected. Pick a simple question to practice with. Eg. Do you/How much do you like spaghetti? Do you/How much do you like watching Dora? Do you/How much do you like the Wiggles? Do you/How much do you like playing in the park?
3. Once the child understands the rating process, begin with Item 1.
4. Each question has two parts,
 - (a) The children are initially asked to respond "yes" or "no".
 - (b) If they answer yes, then ask the second part of the question "how much do you....?".

Item Number	Question	1.		2. (if yes)			
		Do you		How much do you....			
		Yes	No	A little	A lot	All the time	
	AT SCHOOL						
1.talk to most of the children at school? <i>(SMQ: When appropriate, my child talks to most peers at school)</i>						
2. talk to some children, like you friends, at school? <i>(SMQ: When appropriate, my child talks to select peers (his/her friends) at school.</i>						
3.	If you teacher asks you a question,(do you/how much do you)..... use your voice to answer the teacher's question? <i>(SMQ: When my child is asked a question by his/her teacher, she answers)</i>						
4.use your voice to ask you teacher a question when you need to? <i>(SMQ: When appropriate, my child asks his or her teacher questions?)</i>						
5. talk to most of the teachers and adults at school? <i>(SMQ: When appropriate, my child speaks to most teachers and staff at school)</i>						

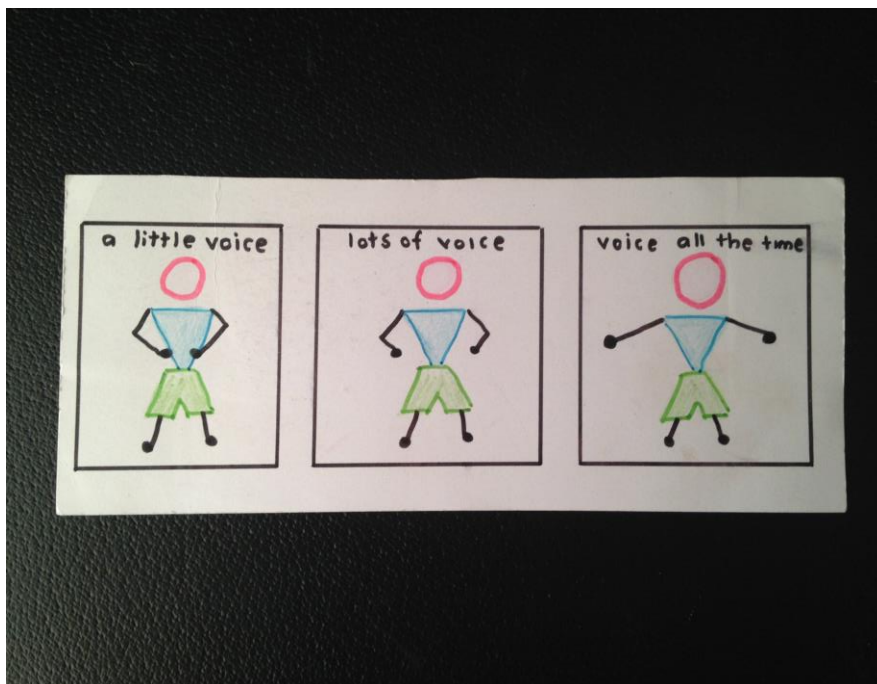
6. talk when you are in a group of children or in front of the class?						
7. ***	<i>(SMQ: When appropriate, my child speaks in groups or in front of the class)</i> In class, when there is an activity where you can join in by talking with your body, like nodding your head or pointing,(do you/how much do you) join in?						
8. ***	<i>(SSQ: Child participates in non-verbal classroom activities)</i> If the teacher asks you an easy question, ... (do you/how much do you)..... answer the question with your body, like nodding your head or by using your hands and pointing.						
9. ***	<i>(SSQ: Child responds non-verbally to simple direct questions)</i> If children in your class are doing an activity in front of the class where they do not have to use their voice like dancing or actions, (do you/how much do you)...join in						
	<i>(SSQ: Child engages in non-verbal classroom performance activities in front of the class)</i>						
	HOME/FAMILY						
10	When other people come to your house to visit.....(do you/how much do you).....talk to your parents and brothers/sisters when they are in the same room as you? <i>visitors</i>						
	<i>(SMQ: When appropriate, my child talks to family members living at home when other people are present?)</i>						
11	When you go to places that you do not know very well with your mother, father and brother/sisters..... (do you/how much do you)..... talk to your parents and brothers/sisters?						
	<i>(SMQ: When appropriate, my child talks to family members while in unfamiliar places)</i>						
12 talk with your grandparents and cousins?						
	<i>(SMQ: When appropriate, my child talks to family members that don't live with him/her (eg. grandparents, cousins))</i>						
13 talk to your parents and brother/sisters on the phone at home?						
	<i>(SMQ: When appropriate, my child talks on the phone to his/her parents and siblings)</i>						
14	Your parents have friends that you see a lot and you know them well. talk to your parents' friends that you know?						
	<i>(SMQ: When appropriate, my child speaks with family friends who are well known to him/her)</i>						

15	Do you sometimes have an adult take care of you at home when your parents are not home eg. like a nanny or baby siter? YES/NO (If yes).....do you/how much do you... talk to them? <i>(SMQ: My child speaks to at least one babysitter)</i>					
IN SOCIAL SITUATIONS (OUTSIDE OF SCHOOL)						
16 talk to children that you do not know? <i>(SMQ: When appropriate, my child speaks with other children who s/he doesn't know)</i>					
17	When you meet your parents friends who you do now know,(do you/how much do you)....talk to them. <i>(SMQ: When appropriate, my child speaks with family friends who s/he doesn't know)</i>					
18 talk to the doctor or dentist when you visit them? <i>(SMQ: When appropriate, my child speaks with his or her doctor and/or dentist?)</i>					
19 talk to the adults who work in the shops and cafes? <i>(SMQ: When appropriate, my child speaks to store clerks and/or waiters.)</i>					
20	Do you play a sport/go to an activity outside of your home? YES/NO (if yes)..... do you/how much do you....talk to other people at the activity? <i>(SMQ: When appropriate, my child talks when in clubs, teams or organised activities outside of school)</i>					
SCORING						
TOTAL of 17 items from the SMQ (ie. excluding items 7-10)			Never	Seldom	Often	Always
TOTAL SCORE						

PICTORIAL RESPONSE SCALE – First side of the response scale



PICTORIAL RESPONSE SCALE – Second side of the response scale.



Individual items of the SMQ-C



APPENDIX B

ETHICS

External approval: Human Research Ethics committee of Macquarie University

(REF 5201100751).

Western Sydney Local Health District Human Ethics Committee

HREC/10/WMEAD/179 SSA/11/WMEAD/190

Department of Education and Communities

SERAP Number 2010175

Catholic Education Diocese of Parramatta (letter dated 24-11-2011).

----- Forwarded message -----

From: Ethics Secretariat <ethics.secretariat@mq.edu.au>
Date: Wed, Sep 7, 2011 at 9:41 AM
Subject: External Approval Noted- Rapee (5201100751)
To: Prof Ron Rapee <ron.rapee@mq.edu.au>
Cc: Miss Maria Ivanka Milic <maria.milic@mq.edu.au>

Dear Prof Rapee

Re: "The Nature of Anxiety in Children with Selective Mutism"

The above application was considered by the Executive of the Human Research Ethics Committee. In accordance with section 5.5 of the National Statement on Ethical Conduct in Human Research (2007) the Executive noted the final approval from the Western Sydney Local Health Network and your right to proceed under their authority.

Please do not hesitate to contact the Ethics Secretariat if you have any questions or concerns.

Please do not hesitate to contact the Ethics Secretariat at the address below, if you require a hard copy letter of the above notification.

Please retain a copy of this email as this is your official notification of external approval being noted.

Yours sincerely

Dr Karolyn White

Director of Research Ethics

Chair, Human Research Ethics Committee



HREC Committee Secretariat:

Professor Stephen Leeder AO
Chair
Professor of Public Health &
Community Medicine

Dr Jim Hazel
Secretary
Medical Graduate -
Endocrinologist

HREC Committee Members:

Sr Patricia Bolster RSM
Catholic Chaplain

Ms Therese Burke
Clinical Trial Coordinator

Mrs Patricia Fa
Clinical Trials Pharmacist

Mr John Fisher
Lawyer

Dr Craig Godfrey
Veterinary Science Graduate

Ms Jillian Gwynne Lewis
Patient Representative

Dr Anthony Harris
Medical Graduate - Psychiatrist

Ms Sheila Holcombe
CEO - GP Network

Ms Jan Kang
Diversity Health Institute

A/Prof Ian Kerridge
Haematologist and Bioethicist

Rev Sarah Plummer
Minister of Religion

Mr John Shaw
Layman

Dr Geoff Shead
Medical Graduate - Surgeon

Dr Howard Smith
Medical Graduate - Endocrinologist

Prof Shih-chang (Ming) Wang
Medical Graduate - Radiologist

Ms Shane Waterton
Laywoman

Ms Christine Wearne
Clinical Psychologist

Our Ref: **HREC2011/3/4.12(3206) AU RED HREC/11/WMEAD/26**

9 May 2011

Ms Maria Milic
Department of Child and Adolescent Psychiatry
Redbank House
Westmead Hospital

Dear Ms Milic

Project title: 'The Nature of Anxiety in Children with Selective Mutism'

Thank you for your letter dated 18 April 2011 addressing the matters raised in the Western Sydney Local Health Network HREC's letter dated 6 April 2011 following single ethical review of the above project at its meeting held on 29 March 2011.

This HREC has been accredited by the NSW Department of Health as a lead HREC to provide the single ethical and scientific review of proposals to conduct research within the NSW public health system. This lead HREC is constituted and operates in accordance with the National Health and Medical Research Council's *National Statement on Ethical Conduct in Human Research* and the *CPMP/ICH Note for Guidance on Good Clinical Practice*.

I am pleased to advise that the HREC has now granted ethical approval of this **single site** research project to be conducted at Department of Child, Adolescent and Family Psychiatry, Redbank House, Westmead Hospital - Chief Investigator Ms Maria Milic.

The following documentation has been reviewed and approved by the HREC:

- NEAF submission code AU/1/2E07011
- Protocol Version 2 dated 10 February 2011
- Revised Participant Information and Consent Form Version 3 dated 18 April 2011
- Participant Information and Consent Form (for Teacher) Version 2 dated 10 May 2011

Please note the following conditions of approval:

- The Chief Investigator will immediately report anything which might warrant review of ethical approval of the project in the specified format, including unforeseen events that might affect continued ethical acceptability of the project.

HUMAN RESEARCH ETHICS COMMITTEE

Research Office, Room 1072, Level 1, Education Block
Westmead Hospital, Hawkesbury Road, Westmead NSW 2145

Telephone: 02 9845 8183
Facsimile: 02 9845 8352
Email: ResearchOffice@swahs.health.nsw.gov.au

Western Sydney Local Health Network
ABN 48 702 394 764

Level 3, Dental School, Westmead Hospital
Darcy Street, Westmead NSW 2145
PO Box 63, Penrith NSW 2751
Tel. (02) 9845 7005 Fax. (02) 9689 2041

Providing health services to the communities of • Auburn • Baulkham Hills • Blacktown • Holroyd • Parramatta

- The Chief Investigator will immediately report any protocol deviation / violation, together with details of the procedure put in place to ensure the deviation / violation does not recur.
- Proposed amendments to the research protocol or conduct of the research which may affect the ethical acceptability of the project, must be provided to the HREC to review in the specific format. Copies of all proposed changes must also be provided to the relevant research governance officer.
- The HREC must be notified, giving reasons, if the project is discontinued at a site before the expected date of completion.
- The Chief Investigator must provide an annual report to the HREC and a final report at completion of the study, in the specified format. HREC approval is valid for 12 months from the date of final approval and continuation of the HREC approval beyond the initial 12 month approval period is contingent upon submission of an annual report each year. A copy of the Annual / Final Research Report Form can be obtained electronically from the Research Office on request.
- It should be noted that compliance with the ethical guidelines is entirely the responsibility of the Chief Investigator.

You are reminded that this letter constitutes *ethical approval only*. This research project must not be commenced at a site until separate authorisation from the Chief Executive or delegate of that site has been obtained. Copies of this letter, together with any approved documents as enumerated above, must be forwarded to all site investigators for submission to the relevant Research Governance Officer.

Should you have any queries about the HREC's Terms of Reference, Standard Operating Procedures or membership, please contact the HREC Executive Officer through the Research Office on 9845 8183 or emailing researchoffice@swahs.health.nsw.gov.au.

In all future correspondence concerning this study, please quote approval number **HREC2011/3/4.12(3206) AU RED HREC/11/WMEAD/26**.

The HREC wishes you every success in your research.

Yours sincerely



Ms Tina Goodenough
HREC Executive Officer
WSLHN Human Research Ethics Committee



Health
Western Sydney
Local Health District

Research Governance Officer
Nepean Blue Mountains Local Health District and
Western Sydney Local Health District
Room 1072, Level 1, Education Block, Westmead Hospital
Hawkesbury Road Westmead NSW 2145

Telephone: (02) 9845 9634

Facsimile: (02) 9845 9636

Email: margaret.piper@swahs.health.nsw.gov.au

22 September 2011

Ms Maria I Milic
Adolescent and Family Unit
Redbank House

Dear Ms Milic

HREC reference number: HREC/10/WMEAD/179
SSA reference number: SSA/11/WMEAD/190
Project title: The Nature of Anxiety in Children with Selective Mutism
Protocol number: version 2 dated 10/2/2011

Thank you for submitting an application for authorisation of this project. I am pleased to inform you that authorisation has been granted for this study to take place at the following site:

- Redbank House - Westmead Hospital

The approved information and consent documents for use at this site are:

- Parent / Guardian - Participant Information Sheet and Consent version 3 dated 18 April 2011
- For Teachers - Participant Information Sheet and Consent version 2 dated 10 May 2011

The following conditions apply to this research project. These are additional to those conditions imposed by the Human Research Ethics Committee that granted ethical approval:

1. Proposed amendments to the research protocol or conduct of the research which may affect the ethical acceptability of the project, and which are submitted to the lead HREC for review, are copied to the research governance officer;
2. Proposed amendments to the research protocol or conduct of the research which may affect the ongoing site acceptability of the project, are to be submitted to the research governance officer.

Yours faithfully


Maggie Piper
NBMLHD and WSLHD
Research Governance Officer

Western Sydney Local Health District
ABN 48 702 394 764
Level 3, Dental School, Westmead Hospital
Darcy Street, Westmead NSW 2145
PO Box 533, Wentworthville NSW 2145
Tel. (02) 9845 7005 Fax. (02) 9689 2041



Education & Communities

Ms Maria Milic
Redbank House Institute Rd
WESTMEAD NSW 2152

DOC 11/222129

Dear Ms Milic

SERAP Number **2010175**

I refer to your application to conduct a research project in New South Wales government schools entitled *The Nature of Anxiety in Children with Selective Mutism*. I am pleased to inform you that your application has been approved. You may now contact the Principals of the nominated schools to seek their participation. **You should include a copy of this letter with the documents you send to schools.**

This approval will remain valid until 09-05-2012.

The following researchers or research assistants have fulfilled the Working with Children screening requirements to interact with or observe children for the purposes of this research for the period indicated:

Name	Approval expires
Maria Milic	09-05-2012

I draw your attention to the following requirements for all researchers in New South Wales government schools:

- School Principals have the right to withdraw the school from the study at any time. The approval of the Principal for the specific method of gathering information for the school must also be sought.
- The privacy of the school and the students is to be protected.
- The participation of teachers and students must be voluntary and must be at the school's convenience.
- Any proposal to publish the outcomes of the study should be discussed with the Research Approvals Officer before publication proceeds.

When your study is completed please forward your report marked to Manager, Schooling Research, Department of Education and Training, Locked Bag 53, Darlinghurst, NSW 2010.
Yours sincerely

Bill Tomlin
R/Senior Manager
Student Engagement and Program Evaluation
5 October 2011

Student Engagement and Program Evaluation Bureau NSW Department of Education and Communities
Level 3, 1 Oxford Street, Darlinghurst NSW 2010 – Locked Bag 53, Darlinghurst NSW 1300 Telephone: 02 9244 5619– Fax: 02 9266 8233 – Email: serap@det.nsw.edu.au



Education &
Communities

Ms Maria Milic
Redbank House
Institute Road
WESTMEAD NSW 2152

DOC13/224593
SERAP Number **2010175**

Dear Ms Milic

I refer to your application for extension/variation of your research project *The Nature of Anxiety in Children with Selective Mutism* in NSW government schools. I am pleased to inform you that your application has been approved.

This approval will remain valid until 20 August 2014.

The following researchers or research assistants have fulfilled the Working with Children Check to interact with or observe children for the purposes of this research for the period indicated:

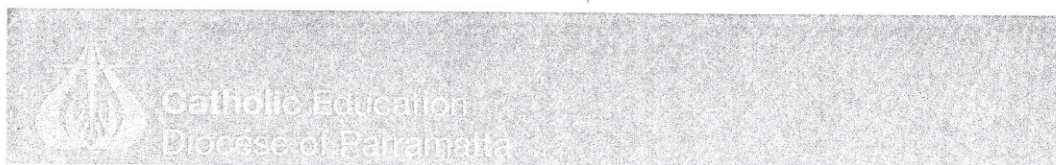
Name	Approval expires
Maria Ivanka Milic	10/09/2014.

When your study is completed please forward your report marked to: Manager, Quality Assurance Systems/Research, Department of Education and Communities, Locked Bag 53, Darlinghurst, NSW 1300.

Yours sincerely

Dr Robert Stevens
Manager, Quality Assurance/Research
27 November 2013

Policy, Planning and Reporting Directorate
NSW Department of Education and Communities
Level 1, 1 Oxford Street, Darlinghurst NSW 2010 – Locked Bag 53, Darlinghurst NSW 1300
Telephone: 02 9244 5060 – Email: serap@det.nsw.edu.au



Ms Maria Ivanka Milic
Redbank House
Institute Road
Westmead NSW 2145

24 August 2011

Dear Maria Ivanka,

Thank you for your Application to Conduct Research in Parramatta Diocese which we received on 7/7/2011. We have now reviewed your ethics approval. I am happy for you to approach the schools in the Parramatta Diocese in order to carry out research on '*The Nature of Anxiety in Children with Selective Mutism*'.

We always stress the following points in relation to research requests:

- It is the school principal, who gives final permission for research to be carried out in his/her school.
- Confidentiality needs to be observed in reporting and must comply with the requirements of the Commonwealth *Privacy Amendment (Private Sector) Act 2000*.
- There should be some feedback to schools and a copy of the findings of the research forwarded to this office.
- This letter of approval should accompany any approach to schools.

I look forward to the results of this study and wish you the best over the coming months. If you would like to discuss any aspect of this research in our diocese, please do not hesitate to contact me on 02 9407 7079 or john.decourcy@parra.catholic.edu.au.

Yours sincerely,

Dr John DeCourcy
Director Strategic Accountabilities Services
Catholic Education Office
Diocese of Parramatta

enabling learning in today's world

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