Selective Mutism: Identification of Subtypes and Implications for Treatment

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Introduction

Selective Mutism (SM) is a rare disorder in which afflicted children will speak normally in one setting (usually home) and will not speak in other environments, such as school. The occurrence in the general population is so low that school psychologists, physicians, and other treating professionals may know little about the disorder or ever have the opportunity to work with children withSM. The criteria and essential feature of SM according to the Diagnostic and Statistical Manual of Mental Disorders-Fifth Edition (DSM-5) (American Psychiatric Association, 2013) is the child's persistent failure to speak in specific social situations where speaking is expected, and at the same time, the child does speak in other situations; therefore, their problem is "selective" and inconsistent across environments, which makes diagnosis challenging. Instead of communicating by standard verbal expression in these selectively mute situations, children with this disorder may communicate by gestures, nodding, or head shaking, or pulling or pushing, or, in some cases, by short monosyllabic or monotone utterances, or in an altered voice. As a result, SM often interferes with educational or occupational achievement and/or with social communication or adjustment. The DSM-5stipulates it must last for at least one month, but not limited to the first month of school. SM should not be diagnosed if the child's failure to speak is due solely to a lack of knowledge, or discomfort with the spoken language required in a social situation (APA, 2013). It is also not diagnosed if the disturbance is better accounted for by embarrassment related to having a language or Communication Disorder, or if it occurs exclusively in the presence of a Pervasive Developmental Disorder, Schizophrenia or other Psychotic Disorder (APA, 2013).

Although SM is a relatively rare disorder, a more recent teacher-identified school-based sample yielded a prevalence of .71% (Bergman, Piacentini, & McKracken, 2002), suggesting that SM may not be as rare as previously reported. SM will typically afflict more girls than boys with a gender ratio of 1.5: 1 to 2.1 (Black & Uhde, 1995; Kristensen, 2000). There is also evidence that SM may be underreported due to families living in social isolation, parents not recognizing SM as a problem in need of intervention, and selective mute behavior existing primarily in the school setting (Hayden, 1980).

The SM diagnosis is often overlooked until the child first attends preschool or kindergarten. This is typically the first time it is brought to the parent's attention that their child will not speak. Often, parents will believe this was a sudden onset, when in reality it may be the first time demands of speaking were placed on the selectively mute child from someone other than their parents or other immediate family members. In some cases, parents may believe the problem lies in the classroom, with the teacher, or school climate because the selectively mute child speaks freely in the home.

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Left untreated, SM can become inadvertently reinforced and subsequently resistant to intervention. The older a child becomes to greater the likelihood the disorder will be more difficult to treat, therefore earlier identification could lead to faster intervention, which could prevent or limit functional impairment (Schwartz, Freedy, & Sheridan, 2006).

Prior research has not explored the multi-dimensionality and factorial nature of SM. This study aims to explore these dimensions. The hypothesis is that not all children with SM are alike and there are distinct SM subtypes that warrant differential diagnosis and treatment strategies. Children with SM have different clinical presentations, and different explanations for the selective withholding of speech. The potential subtypes which will be explored in this paper are: Anxiety Mustim, Expressive Language Mutism, Oppositional Mutism, English Language Learners Mutism, and Sensory Self Regulation Mutism.

The most popular and common theory for the SM condition can be attributed strictly to a high level of anxiety, and closely related to social anxiety disorder (Dow, Sonies, Scheib, Moss, & Leonard, 1995). However, some children may also have difficulty controlling emotional states, which interferes with self-regulation of anxiety (Bronson, 2000). Some children with SM may have neurodevelopmental vulnerabilities that may impact communication and language skills (Viana, Beidel, & Rabian, 2009), and this could be related to second language acquisition in some children with SM (Cohen, Chavira, & Stein, 2006). Lastly, a small number of children with SM may refuse to talk due to a tendency toward controlling, demanding, oppositional and aggressive behaviors (Kumpulainen et al., 1998; Steinhausen & Juzi, 1996). These different presentations lend greater credibility to the potential of distinct subtypes ofSM; and if substantiated through empirical investigation could further advance differential diagnosis, impact treatment strategies, and ultimately improve outcomes for children with SM.

Conceptualizing SM and Potential Subtypes

Selective mutism is etiologically similar to anxiety disorders, it is often comorbid with social anxiety disorder, as children with social anxiety disorder and SM often experience comorbid symptomatology, including but not limited to anxiety (APA, 2013). Therefore, it is natural to delineate anxiety as a suspected specific subtype of SM. However, there are children who do not fit the specific prototype of an anxious child. For example, some children with SM may score in the non-clinical range for anxiety ratings, yet still exhibit full criteria for a SM diagnosis.

Anxiety Mutism

Despite the similarities between social anxiety disorder and SM, there are some major distinguishing factors, such as age of onset. Specifically, the median age of onset for social anxiety disorder is age 13 (APA, 2013) whereas onset for SM is usually before age 5 (APA, 2013; Steinhausen & Juzi, 1996). In addition, many children with SM actually enjoy social situations, so they are not fearful of peer or adult interaction per se. They enjoy going to school, and actively engage in nonverbal communication with peers and adults across a variety of environmental contexts. Some researchers have attributed this to the possibility that children with SM have found a successful avoidance strategy (e.g., selective speaking behavior) in socially anxious situations; hence their anxiety is lessened during these social situations (Yeganeh, Beidel, Turner, Pina, & Silverman, 2003). This type of child would not have a social anxiety disorder, as these children avoid social situations because of the discomfort they cause.

Other researchers believe that SM is a developmental subtype of social anxiety disorder with an earlier onset than other symptoms of the disorder (Bergman et al., 2002). The potential to identify these youngsters with SM may help professionals to intervene earlier with the treatment of social anxiety disorder symptoms (Black & Uhde, 1995; Ford et al., 1998).

There is currently no data available that overwhelmingly demonstrates that children with SM score in the "extreme range" on social anxiety measures. One study found that clinician ratings of social anxiety on the Liebowitz Social Anxiety Scale (Dummit, Klein, Tancer, Asche, Martin & Fairbanks, 1997) were in the moderate range forchildren withSM. In other studies, teacher ratings have indicated that measures of social anxiety were higher for children with SM than the comparison group; their scores were not usually considered in the clinically significant range (Bergman et al., 2002). These findings are inconsistent with the conclusion that selectively mute children are "frozen with fear" (Anstendig, 1998). In actuality, it suggests that while children with SM have higher levels of anxiety, there may be additional cognitive and behavioral factors that are involved in and mediate SM (Mulligan & Christner, 2006). Cognitive factors may include negative internal dialogues surrounding coping, or shame surrounding their behaviors; behavioral factors may include avoidance, especially when expectations to speak are high.

Based on the evidence cited above, a strict reconceptualization of SM and how it relates to social anxiety disorder is warranted. In fact, the lower anxiety scores for large SM samples could reflect the fact that some children with SM have considerable anxiety, while others do not have significant levels. Instead, the author(s) propose that a subtype of SM in which children exhibit primary symptoms of social anxiety, including, fears of speaking, fears that others will make fun of them if they speak, worries of others hearing there voice, and any other anxiety based symptoms might explain these findings.

Expressive Language Mutism

Communication disorders are commonly found among children with SM. Premorbid ICD-10 speech and language disorders were present in 30.3% of one selectively mute sample (Steinhausen, et al., 2006) and in another (Steinhausen & Juzi, 1996), 38.0% of children with SM had speech or language disorders. The most common was in the area of expressive language disorders (28.0%) followed by articulation disorder (20.0%).

An exploratory study comparing narrative abilities of selectively mute children as compared to socially phobic children indicated that children with SM produced significantly shorter narratives than children with social anxiety disorder, despite showing normal nonverbal cognitive and receptive language abilities (Mcinnes, et al., 2004), this suggests that although children with SM and social anxiety disorder have similar presentations, children with SM may exhibit slight expressive language deficits not seen in children with social anxiety disorder, but the small sample size may have limited findings (Viana, et al., 2009). This finding suggests that both anxiety and mild expressive language deficits may be components of SM even when speech and language delay has been ruled out on standardized assessments. Furthermore, the potential academic and social outcomes of a combination of weak or subclinical language deficits coupled with continuing functional impairment from chronic SM symptoms (including anxiety) may be more serious than those associated with weak language skills alone (Mcinnes et al., 2004). Regardless, these findings would be consistent with the hypothesis that some children with SM avoid speaking out of fear of articulation or expressive language errors, which could result in teasing from peers (Standart & Le Couteur, 2003).

Language abilities of children with SM and children with social anxiety disorder were directly compared in an attempt to investigate potential differences between the two groups (Manassis et al., 2003). After reviewing the parents' reports of overall communication ability, phonemic awareness via the Lindamood Auditory Conceptualization Test (Lindamood & Lindamood, 1971), and receptive language using the Peabody Picture Vocabulary Test- III (Dunn & Dunn, 1997) and the concepts and directions subtest of the Clinical Evaluation of Language Fundamentals (Semel, Wiig, & Secord, 1995), selectively mute children scored significantly lower on only one task (discrimination of speech sounds) than socially anxious children.

This suggests the language problems in selectively mute children may not just be expressive. When the authors looked at the group mean of the CELF Total Score, children with SM scored mostly in the average range, but a subgroup (42.9%) of the children with SM scored in the clinical range on at least one of the language measures.

Language problems are only one indication of neurodevelopmental delay. There are many other markers that can signal problems, such as gross or fine motor delays, physical deformities or delays in social and emotional development. However, these problems have been far less researched in children with SM (Viana, et al., 2009). One study which examined broad markers of developmental delay found that children withSM, regardless of comorbid communication disorder, showed higher rates of fine and gross motor delays, higher rates of minor physical abnormalities, and more pre- and perinatal risk factors than controls (Kristensen, 2002). Therefore, neurodevelopmental delay may play a role in the development ofSM (Viana, et al., 2009).

Oppositional Mutism

Some children with SM are reported to display controlling, demanding, oppositional, and aggressive behaviors (Kumpulainen et al., 1998; Steinhausen & Juzi, 1996). Black and Uhde (1995) found that only 10% of children with SM met criteria for oppositional defiant disorder (ODD) and that parent and teacher ratings of oppositional behaviors and immaturity did not correlate with mutism severity. When scrutinized at the item level on the Teacher's Rating Scale (TRS), a comprehensive psychiatric/behavioral rating scale, modified from the Connor's Rating Scale (Gittelman, 1985) scores on the teacher rated items describing oppositional behavior were low and also did not correlate with mutism severity. However, in a more recent study (Manassis et al., 2007), 6.8% of children with SM met criteria for ODD and in another study (Arie, Henkin, Lamy, Tetin-Schneider, Apter, & Sadeh, 2006), 11.1% were comorbid for SM and Attention Deficit/Hyperactivity Disorder (ADHD). Therefore, the comorbidity between SM and disruptive disorders, range anywhere from 6 and 10%, which is somewhat elevated in comparison to rates found in the general child population (Barkley, 2003; Lahey, Miller, Gordon, and Riley, 1999).

Although Ford et al. (1998) found their sample of children with SM had higher Internalizing Problems (e.g. withdrawal, anxiety) than Externalizing Problems (e.g. delinquency, aggression), there was some data to support the presence of oppositionality at least among a sub-sample of children with SM. Oppositional-defiance/aggression behaviors were found among 26% of a sample of German and Swiss children who met criteria for SM (Steinhausen & Juzi, 1996).

Additionally, among a sample of 153 individuals with past or current SM (Ford et al., 1998), the top ranked concerns were avoidance behaviors, shy/withdrawn behavior, toileting problems and strong willed behaviors (e.g., stubborn, controlling). There were many oppositional items that were endorsed, including refusal to talk, stubborn, sullen, or irritable; argues a lot; disobedient in school; whining; and temper tantrums or hot temper. Although other studies dispute the presence of externalizing symptoms in children with SM (Cunningham et al., 2004), the fact there is mixed evidence lends credence to the possibility of a potential Oppositional Mutism subtype, and if present would suggest important treatment considerations when working with this subset of selectively mute children.

English Language Learner (ELL) Mutism

The literature has been quite clear regarding the growing number of immigrant children identified withselective mutism. In fact, the prevalence rate ofSM is nearly 4 times higher for immigrant children than non-immigrant children (Elizur & Perednik, 2003). In addition there are differences in normative social development and neurodevelopmental delays among immigrant children when compared to native children. These differences alone warrant further investigation.

The combined circumstance of immigration and second language learning in children who are extremely shy or anxious elevates risk to develop SM (Bradley & Sloman, 1975; Elizur & Perednik, 2003). A significant number of children who are bilingual or multilingual experience an initial silent period when they first enter an environment that has a different culture and language. This phenomenon is considered normal during second language development (Chitester, 2005). Children engaged in learning a new language need time in this silent period; however, in consideration of SM, one can begin to appreciate how culturally and linguistically diverse (CLD) children can be at an elevated risk to develop SM.

It is common for clinicians to recommend that CLD children practice social communication outside of the school environment, within the community, such as restaurants, stores, the library, etc. However, for some immigrant families there may be limited parental facilitation of exposure tasks and limited compliance with prescribed parent training strategies. This is often because there is minimal social contact outside of the immediate family environment.

Thus, the lack of appropriate modeling by family members in the community and the inability to practice skills learned can negatively impact the immigrant child with SM. When resistance occurs within the family, the clinician must be concerned that as a result the parents are modeling avoidance of outside social interactions (Fisak, et al., 2006). Low acculturation can also contribute if not trigger some social isolation and enhance the severity of SM in the child (Vecchio & Kearney, 2007).

Sensory/Regulation Mutism

The last of the potential SM subtypes may be related to sensory and self-regulation issues. Self-regulation has been described in two important ways. First, it is viewed as the ability to adjust arousal in an appropriate manner in order to attain goals; second, self-regulation is viewed as the ability to direct how emotions are revealed behaviorally in socially adaptive ways (Bronson, 2000). Selectively mute children have difficulties self-regulating their anxiety, and other emotional states. Children with SM have difficulties adjusting their emotions to yield a behaviorally positive or socially adaptive response (Bronson, 2000). These abilities evolve from a complex process involving a dyadic regulatory system. Within this dyadic regulatory system, the cues given by infants and toddlers regarding shifts in their emotional state are acknowledged by their caregivers who ultimately assist in self-regulation (Fonagy, 1999; Sroufe, 1983). In addition, cultural values, societal expectations, physiology, cognition and developing speech and language skills contribute to the organization and the ability to self-regulate (Moldan, 2005).

Children with poor self-regulation may present as shy, timid, and mute in some social situations; however, in more comfortable situations, their behavioral presentation is in stark contrast, loud, impulsive, and hyperactive (Cunningham et al. 2004; Greenspan, 1997). In addition it is hypothesized many of these children also demonstrate sensitivities to touch, noise, and light, and executive function deficits hampering self-control of both sensory processing and motor output demands.

Purpose of the Study

Given the complexity of the SM diagnosis, and the various presenting symptoms and comorbid conditions, it is hypothesized that there are multiple subtypes of SM. The hypothesis is that a cluster analysis of the Selective Mutism Comprehensive Diagnostic Questionnaire (SM-CDQ; Shipon-Blum, 2004) will reveal five different and distinct subtypes of SM. It is predicted that these five subtypes will subsequently be differentiated on parent ratings of behaviors commonly reported in SM populations using multivariate analysis of variance.

Based on the literature summarized above, five subtypes of SM are expected. These Selective Mutism subtypes include: (1) *Anxiety Mutism*, distinguished by a phobia to speak, a fear of others hearing their voice, or a rule out when the SM child does not demonstrate one of the other subtypes and there is a familial history of anxiety. (2) *Expressive Language Mutism*, distinguished by narrative expressive language deficits; (3) *Oppositional Mutism* distinguished by significant challenging behaviors exhibited regardless of the expectation to speak; (4) *ELL Mutism*, distinguishing factors would include selectively mute children whose native language was not English, or who had cultural differences with mutism beyond the "silent period"; (5) *Sensory/Self Regulation Mutism*, distinguishing factors would include selectively mute children with sensory integration dysfunction and/or selectively mute children who demonstrated extreme difficulties with regulation of their emotions during non-mute times; also children who demonstrate significant executive dysfunction would be included in this sub-group.

Method

Participants/Source for Data

Participants were found through the Selective Mutism Anxiety Research and Treatment Center (SM-art-Center) located in Southeastern Pennsylvania. The SM-art-Center data-base included children from other regions of Pennsylvania, multiple states and countries. This was a sample of convenience and archival data was used and approved by IRB. Detailed information regarding the socioeconomic status (SES) of the selected children was not available; however, because of the duration, modality, and the cost of the treatment, SES was likely to be in the middle to upper middle class range.

Table 1 displays the demographic characteristics of the children whose parents completed the SM-CDQ. The sample included 186 children with SM of mostly Caucasian children (81%). There were a large number of girls (61%) in the sample. Although ages ranged from three to 18, many of the children were in pre-kindergarten through first grade (67%). There were a number of children with SM who demonstrated school-related problems. In fact, 10% of the children with SM were retained, while 7% were receiving Americans with Disabilities Act (ADA)/Section 504 services, and another 26% were receiving Individuals with Disabilities Education Act (IDEA) special education services. Although these figures suggest many children with SM had disability diagnoses, a majority of children with SMdid not have any formal services to accommodate their mutism.

Inclusion and Exclusion Criteria

Individuals in the database were excluded if they were out of the age range of the sample desired, if they had a previous diagnosis of autism, if the questionnaire was incomplete or had missing data, or if it was clear they did not meet criteria for SM according to the SM-art-Center staff who conducted the evaluation. In addition, children with known mental retardation or brain injury or medical condition affecting the child's status at evaluation were also excluded.

Instrumentation

Shipon-Blum (2004) developed a comprehensive parent report the measure used in this study entitled the Selective Mutism Comprehensive Diagnostic Questionnaire (SM-CDQ). The SM-CDQ is an assessment tool for children with SM and is an essential tool to help with treatment (Shipon-Blum, 2004). According to Shipon-Blum, the instrument assists in identification of SM symptoms and a SM diagnosis. It also helps the clinician recognize the specific settings the mutism occurs, and the severity and pervasiveness of the symptoms and the child's frequency of non-speaking behavior across situations in which children are expected to speak. This is an important first step at obtaining a deeper understanding of SM.

This questionnaire helps professionals gather data that ranges from basic demographic to detailed diagnostic information about the referred child. There are items about socialization and interactions with friends and classmates, and how the child with SM communicates, if at all with other children. There are items about the child's personality, body language, and behavior, in addition to questions on the school

environment, the home environment, and the community environment.

There is a full developmental history incorporated into the questionnaire, and questions related to the parents' (and extended family) history of anxiety or depression. The SM-CDQ inquires about the child's bladder and bowel control, eating patterns, and any coexisting disorders. It also specifically lists twelve of the most common comorbid disorders with SM and asks the parent to identify whether their child has been diagnosed with any of them. There is also an opportunity to list any alternative disorders that the child may have. There are open-ended response items regarding speech and language history, sensory issues, anxiety, and if the child is multi- or bilingual. The SM-CDQ also addresses issues related to sibling history and the child's current home life. Dr. Shipon-Blum (personal communication, April 21, 2005) reports a common use of the SM-CDQ is to help determine if the presenting child with SM can be categorized into an SM subtype. However, this is the first study that looks statistically if subtypes truly exist.

The SM-CDQ has a list of items at the end of the questionnaire that parents are asked to rate on a scale of 1 to 10, which in this study we named the Mutism Behavior Rating Scale. A low rating on MBRS items indicated the child had few problems, while a high rating would indicate greater problems for that item. Although the SM-CDQ is not fully validated, it has strong content validity and diagnostically was mirrored after DSM-IV TR criteria, and the extant literature on SM, as well as the extensive clinical practice at the Smart Center.

The first subscale is Executive ($M = 41.59$, $SD = 12.56$). It includes items such as *procrastination, distractible, inflexible, impulsive, disruptive, deceitful, concentration, daydreams, hyperactive* and *disorganized*. The means for the individual items in this subscale appeared consistently to have ratings in the low to moderate range, however, *concentration* was the most problematic item on this subscale ($M = 7.50$, $SD = 2.21$). The Executive subscale was highly correlated with the Oppositional, Labile, and Sensory subscales. This may suggest a strong relationship between executive problems and mood/behavior problems. It was also correlated with the Anxiety subscale, suggesting a possible connection between anxious behaviors such as nervousness impacting an executive skill such as concentration.

The next cluster of items comprised the Oppositional subscale ($M = 27.82$, $SD = 8.18$). The items included *stubborn, aggressive, uncooperative, upset by mutism,* and *wants to speak*. On the item level, parents indicated that a majority of the sample of children with Selective Mutism both wanted to speak, and were upset by their mutism. Qualitatively, there was a higher mean for the *stubborn* item, than there was for the *aggressive* or *uncooperative* items, suggesting that although many of the children may be somewhat resistant and/or passive-aggressive, this does not immediately translate into aggression and overt noncompliance. The Oppositional subscale was correlated with the Labile and Sensory subscales.

The Labile subscale ($M = 30.38$, $SD = 11.26$) consisted of the following items, *tantrums, unpredictable behavior, upset easily, labile, feelings intense, dysphoric,* and *self-deprecation*. In this category, the items with the highest means were *upset easily* and *feelings intense*. This suggests that subtypes significant on the Labile scale may be sensitive, reactive, and intense when experiencing feelings. While this could lead to self-deprecation, they appear to be less likely to externalize these feelings negatively. The Labile scale is also highly correlated with the Sensory and Esteem subscales, which may suggest children with SM who have mood problems also have sensory issues and compromised positive self-evaluation.

The Anxiety subscale ($M = 51.35$, $SD = 8.39$) had the following nine items; *positive affect home, positive affect public, shy, enjoys solitary, enjoys playing in groups, worries excessively, nervous, cleanliness preoccupation,* and *order preoccupation*. The items typically characteristic of obsessive-compulsive tendencies were rated as quite low, suggesting these characteristics are not especially problematic for a majority of children withSM. The items with the highest means, suggesting greater problems were related to the child's affect in the home and

shyness. Significant ratings would suggest these children typically prefer to play by themselves than in a group, and although these children tend to be moderately nervous, they are less likely to worry excessively. The Anxiety subscale was strongly correlated with both the Esteem and Academic subscales. It was also correlated with the Sensory subscale.

Only three items make up the Flexible subscale ($M = 15.19$, $SD = 6.25$). These include *adjusts to routines, accepts novelty* and *accepts new situations*. All three of these items had similar means and standard deviations, with ratings suggesting moderate problems in this category. A child who has significant ratings on the Flexible subscale may have more difficulty adjusting to new routines and accepting novelty than accepting new situations. The Flexible subscale was correlated with the Esteem subscale.

The Sensory subscale ($M = 30.90$, $SD = 15.40$) comprises *noise sensitive, light sensitive, crowd sensitive, food sensitive, novel food intolerance, hair sensitive,* and *clothes sensitive* items. The items in this category had low to moderate means with *crowd sensitive* having the highest mean ($M = 5.27$, $SD = 2.95$). This suggests that a subtype significant on this scale is going to have greater difficulty navigating large crowds, or being in large groups, but does not suggest overt agoraphobia.

There were eleven items that made up the Esteem subscale ($M = 72.25$, $SD = 14.68$). The Esteem items include *assertive, expresses emotions, affectionate, empathetic, positive self image, positive relations, positive self care, independent, self view of intelligence, enjoys life,* and *likes self*. They were reverse coded in the data set to indicate higher scores indicated less self-esteem. Overall, at the item level, children in this sample appeared to have difficulty with assertiveness, and expressing affection and empathy. Their self-esteem may be somewhat compromised by problems relating positively to others, feeling like they are not intelligent, or having a general dislike for themselves. The Esteem scale was strongly correlated with the Academic scale, implying that with more positive academic performance comes more positive self-esteem.

Lastly, the items in the Academic subscale ($M = 53.72$, $SD = 10.85$) include *organized, listens well, enjoys art, enjoys music, enjoys math, enjoys reading, focused,* and *abstract thinker*. A child with SM who has significant ratings on this subscale would have global academic problems. A qualitative item analysis suggests that while children in the sample may not have extensive difficulties in their organizational skills, they do have problems listening well. These children most likely have difficulties with math and reading, and may have problems with abstract reasoning and creativity.

Procedure

Variables were broken down into three different groups, descriptive variables (D), characteristics of mutism variables (CM) and a Mutism Behavior Ratings Scale (MBRS). MBRS variables were coded so that higher ratings always indicated greater impairment. The two rating scales within the SM-CDQ were used for inferential analyses. The first set of items included Characteristics of Mutism (CM variables) and the second set of items comprised the Mutism Behavior Rating Scale (MBRS variables). Each of the existing MBRS variables were examined and categorized into more meaningful subscales for this study and included Anxiety, Executive, Oppositional, Labile, Flexible, Sensory, Esteem, and Academic subscales.

Results

The SM population has often been considered homogenous in terms of its clinical presentation, however, more recently research has disputed this, therefore, utilization of cluster analysis can be valuable for discovering the differences in characteristics associated with this possibly not so homogeneous sample ofSM. In this study, cluster analysis was used with the purpose of identifying homogeneous subtypes of children with SM based on subscales from the mutism behavior ratings scale (MBRS).

The hierarchical cluster analysis utilized the Within Groups Linkage method and used a Phi – 4 point correlation for binary data as the distance measure. This method combines clusters so that the average distance between all possible pairs of cases in the resulting cluster is as small as possible, thereby minimizing

within group variability and increasing homogeneity of the cluster. The results of the Within Groups Linkage variant of the UPGMA revealed 6 SM subtypes according to the agglomeration schedule coefficient changes from Step 6 (.373) to Step 5 (.362). Group 5 (*n* = 7) was omitted from further analyses due to small sample size, consistent with the sample size requirements for this study.

Exploring the MBRS means and the descriptive characteristics of the groups of children with SM helped to clarify subtype characteristics in this sample of selectively mute children. Table 2 shows the MBRS means for the five subtypes. Although it is important to note that the scales are not directly comparable, given the number of items per subscale varies, within scale comparisons and examination of item means that comprise that scale helps delineate the subtypes. Using this information, the subtypes were labeled Global Mutism (G), Anxiety/Language Mutism (A/L), Low Functioning Mutism (LF), Sensory Pathology Mutism (SP), and Emotional/Behavioral Mutism (E/B).

Selective Mutism subtype descriptive data are displayed in Table 3. All subtypes were composed of more females than males with the exception of the Sensory subtype, which had an even distribution of males to females. Age was primarily even across the subtypes, although the Low Functioning subtype had the lowest mean age of 5.8. Consistent with age levels, most children were in pre-Kindergarten, Kindergarten, or elementary school.

Most children had a history of ear infections (range 27% to 39%) and language delays (range 4% to 20%), but speech impediments were most common in children in the Anxiety/Language (20%), Sensory/Pathology (14%), and Emotional/Behavioral (18%) subtypes. Children in theSensory/Pathology subtype also appeared to have the most motor problems. Not surprisingly, symptomatology representing difficulties integrating sensory information were evident in this sample with the largest percentage found in the Sensory/Pathology group. Speech and language diagnoses were also common in the Anxiety/Language and Sensory/ Pathology subtypes, while high levels of bilingualism were noted in all subtypes (range 20% to 27%) except those with Emotional/Behavioral Mutism (9%).

Comorbid psychiatric diagnoses are not uncommon in children with mutism, but surprisingly few had comorbid externalizing disorders. Instead, most children evidenced had anxiety, or anxiety related disorders, such as specific phobia and social anxiety disorder; environmental stressors were also endorsed at high rates in several subtypes, with the Anxiety/Language (53%), Sensory/Pathology (46%), and Emotional/Behavioral (36%) showing the highest percentages of cases with an anxiety related comorbidity. A surprisingly large number of children had experienced an environmental stressor in their lives, unfortunately, this was not defined in terms of the severity or duration of the stressor, however, percentages ranged from 13% in the Low Functioning group to 53% in the Anxiety Language group. The latter suggests that there could be a relationship between high environmental stress exposure and rates of anxiety. Finally, comorbid separation anxiety diagnoses were exceedingly high compared to typical samples, with the Global (59%), Anxiety/Language (67%), Low Functioning (65%), Sensory/ Pathology (73%), and Emotional/ Behavioral (82%) groups all showing high rates of this comorbidity.

It appears that across all subtypes, children with SM had mothers and fathers endorse shyness (70% and 58%) and anxiety (39% and 26%) respectively at higher rates than in the general population. Maternal and paternal history of mutism as children were also at higher proportions (9% and 5%) respectively, and lastly maternal and paternal depression rates were also elevated at (21% and 13%) respectively. On all the parent mutism characteristics more mothers suffered from shyness, anxiety, mutism as a child and depression than fathers. However, in the Anxiety/Language Mutism, parental mutism characteristics did not appear to be a factor in how anxious these children are, likely supporting environmental variables also contributing to anxiety.

To further clarify the characteristics of subtypes, each subtype was evaluated separately, and in comparison to each other. Results for the ANOVA with the MBRS subscales as dependent variables are displayed in Table 2. Bonferonni Post Hoc Tests formultiple comparisons were also computed, as were Least Significant Difference (LSD) post hoc tests for discussion purposes only.

There were significant differences between groups on several MBRS subscales. Although Bonferroni post hoc tests revealed that only the Emotional/ Behavioral subtype showed more executive problems than the Global subtype, the Least Significant Difference (LSD) post hoc comparisons revealed executive problems were more prevalent in the Low Functioning subtype and the Emotional/ Behavioral subtype as compared to the Global subtype.

The LSD post hoc tests also showed that the Anxiety/Language, Low Functioning, and Sensory/Pathology subtypes and Emotional/Behavioral subtype had more problems on the Labile scale than the Global subtype. There were also differences on the Oppositional scale with the Sensory/Pathology and Emotional/ Behavioral subtypes having more problems than the Speech Phobic and Low Functioning subtypes. The Global subtype had more problems on the Flexible scale than the Low Functioning subtype, and on the Sensory scale, the Low Functioning, Sensory/ Pathology and Emotional/Behavioral subtypes had more problems than the Global and Anxiety/ Language subtypes. On the Anxiety subscale, the Anxiety/ Language subtype had significantly more problems than the Global subtype. Lastly, on the Academic scale, the Anxiety/ Language, Low Functioning, and Sensory /Pathology subtypes had significantly greater problems than the Global subtype.

The LSD post hoc tests showed that the Anxiety/Language, Low Functioning Sensory/Pathology and Emotional/Behavioral subtypes had more problems on the Oppositional and Labile Scales than the Global subtype. Similarly, the Anxiety Scale, Flexible Scale, and Academic Scale all yield significant results from the LSD post hocs where they did not yield significant results on the Bonferroni post hoc. The Bonferonni is a more conservative post hoc versus the LSD is more liberal, however, the LSD post hoc does not control as well for Type I error.

Global Mutism. This subtype was characterized by the largest number of children with SM (n = 71). The children with SM in this subtype appeared less impaired as compared to the other subtypes. These children may have developmentally moved from a subtype with more significant problems to the Global subtype as their needs, such as anxiety and sensory problems were addressed. Although children in this subtype had problems with self-esteem, and flexibility, they were generally academically capable, and did not exhibit sensory, emotional and behavior problems as some of the other subtypes. The Global subtype had a 2:1 gender ratio in favor of females, and was the only subtype to encompass all racial variables. Most likely the majority of children with SM would fall into the Global subtype, and this may support why some children with SM are not in need of special education services.

Anxiety/Language Mutism. This subtype was characterized by a significantly higher mean score as compared to the other groups for Anxiety based on LSD post hocs. Lability and Academic Success were also problematic for this subtype in comparison to the other subtypes. The Anxiety/Language group also had the largest percent of a comorbid anxiety disorder, and the largest percent of environmental stress exposure. This subtype had the smallest percentage in comparison to the other subtypes to meet speech developmental milestones, and had the largest percent of speech impediments and speech and language diagnoses. This subtype had a 2:1 gender ratio in favor of females, and a slightly elevated number of children with SM with a comorbid learning disability.

Low Functioning Mutism. This Low Functioning subtype has an even gender ratio, with no significant comoribid psychiatric disorders. However, both maternal mutism and depression were high in this subtype, suggesting possible psychopathology in the immediate family. The mean for Positive Academic Success was highest for this subtype, suggesting this subtype has the greatest academic problems. The Positive Academic

Success scale was statistically significant on Bonferroni post hocs, indicating a statistically significant problem on this scale in comparison to the other subtypes. Other characteristics pertinent to this subtype are statistically significant sensory and executive problems as well as lability according to LSD post hocs. These problems are likely contributors to the academic problems as the hallmark of this subtype. The Low Functioning subtype had the highest percent of children with SM in special education, supporting this subtypes academic difficulties.

Sensory/Pathology. The mean for Sensory was highest and statistically significant from the other subtypes according to Bonferroni post hoc analysis. This Sensory/Pathology subtype also had the largest percent of selectively mute children with a comorbid diagnosis of Disorder of Sensory Integration. This subtype also appeared to be characterized by oppositional behavior and lability, as mean scores were significant in these areas according to LSD post hocs. The Sensory/Pathology subtype was the most racially diverse, and had a high rate of bilingual children. This subtype had the largest number of children with separation anxiety problems, and delays in motor skills. Overall, this subtype appeared the most impaired in comparison to the other subtypes. With comorbid diagnoses of learning disabilities, ADHD, ODD, and Depression; this may suggest that this subtype may possibly be the most difficult to treat.

Emotional/Behavioral. This subtype had the highest mean scores for all subtypes in the areas of Executive, Oppositional, and Labile, these mean scores were significant according to Bonferroni and LSD post hocs. This subtype had the second highest mean score for Sensory. This subtype is also noteworthy when looking at gender differences, because the female to male ratio is 10:1, suggesting this subtype is more prevalent in females. Also noteworthy is this subtype does not appear to have problems as compared to the other subtypes academically. This is surprising considering the statistically significant mean score on the Executive scale, and typically children with executive problems perform less well academically.

Discussion

Selective Mutism has often been thought of as a homogeneous disorder, with the primary characteristic of withholding speech in certain settings and not others being the one unifying feature of the disorder. This study provided evidence that there are characteristics that are distinct to some children with SM and not others, or at least to varying degrees. Not only does this research support recognition of SM subtypes for better understanding of the differences among children with SM, it could also have significant implications for best practices in SM treatment for each subtype.

In addition to examining the core features ofSM, it is also important to recognize the functional impact of SM on these children, consistent with Bergman, et al. (2002) noting that children with SM were significantly more impaired than healthy comparison children, particularly in the areas of academics and social functioning. Results from a longer term follow up study also suggest that significant functional impairment remains over time (Remschmidt, et al., 2001), which likely affects the treatment of the core symptoms of SM.

It is clear that researchers need to conduct studies that provide best treatment options for children with SM. This suggests it is relevant to explore possible subtypes of SM, so children afflicted with this condition can make faster, more efficacious, treatment gains.

Subtype Differentiation and Clinical Implications

The cluster analyses revealed six subtypes, however, one of the subtypes did not meet minimum sample size criteria, and as a result, the results for this subtype were not examined. However, for the remaining participants, there were meaningful subtype differences that emerged in this study. Not

surprisingly, most of the subtypes have similar, overlapping characteristics, but there were some unique characteristics worth noting. Shipon-Blum (2010) notes these subtypes are not necessarily pure, and that all subtypes will have some related features. For example, as previously hypothesized by the first author, at least five SM subtypes were predicted to emerge, including Anxiety, Expressive Language, Oppositional, English Language Learner, and Sensory/Self Regulation SM subtypes. Although these specific subtypes did not emerge as clearly as originally predicted, five subtypes did emerge with some primary distinguishing features, coupled with secondary characteristics which also made each subtype unique.

The literature reports that the gender ratio is 1.5: 1 to 2.1, favoring females (Black & Uhde, 1995; Kristensen, 2000). However, in this study gender ratios were sometimes as high as 10:1 in favor of female children. This is a significant finding in relation to the Emotional/Behavior subtype. It has been reported that some children with SM have difficulties self-regulating their anxiety, and other emotional states. Children with SM often experience difficulties adjusting their emotions to yield a behaviorally positive or socially adaptive response (Bronson, 2000). The Emotional/Behavioral subtype found in this study included many similar characteristics. The children in the Emotional/Behavioral subtype not only experienced sensory problems, but had more MBRS Executive, Oppositional, and Labile subscale problems than other subtypes.

Interestingly, this subtype does not appear to experience academic problems relative to the other subtypes. This may suggest that parents are inaccurately rating their children on the MBRS, or that environmental cognitive and/or behavioral factors are indeed maintaining (Mulligan & Christner, 2006) and conditioning SM (Shipon-Blum, 2010). Also noteworthy, is this subtype had the highest rate of maternal shyness, maternal anxiety and fathers of children with SM who were mute when they were children, suggesting possible further exploration into environmental and/or genetic factors is warranted. Additionally, the Emotional/Behavioral subtype afflicts more preschool, female youngsters that may imply this is a newly discovered disorder, and teachers are making attempts to compel the child to speak. In addition, young children will typically shut down more often than older children (Shipon-Blum, 2010). It is during this time many young children with SM may demonstrate oppositional and labile behaviors.

The Sensory/Pathology subtype had a fairly equal gender ratio. In consideration and examination of other significant characteristics in this subtype, overall, children appear to experience the most pathology with comborid diagnoses of LD, ADHD, ODD, and Depression. This is consistent with literature findings that children with SM may have an associated developmental delay or learning problems nearly as often as an anxiety disorder (Cleater & Hand, 2001; Kolvin & Fundudis, 1981).

Externalizing behaviors in children with SM are not as well documented, however, one study concluded that ADHD and ODD were one to ten times greater in the SM population (Ford, et al., 1998). Also of clinical interest in this subtype are two factors not often explored; the psychopathology of the father of the child withSM. The Sensory/Pathology subtype had the highest percentage of both paternal anxiety and depression. This factor may suggest that paternal psychopathology could negatively impact children withSM, who are already vulnerable for psychopathology themselves. Therefore, as is the case with other subtypes, it may be important to not only address the children with SM during treatment, but family members as well.

There is growing evidence that language disorders, especially in the area of expressive language, are prevalent in some children with SM (McInnes, et al., 2004; Shipon-Blum, 2010). Speech and language problems were especially prevalent in the Anxiety/Language mutism subtype. Children in this subtype had difficulties early on with delays in speech and language as this subtype reportedly did not reach developmental speech milestones on time. Other language markers for this subtype were a high percentage of speech impediments, and the largest percentage of children with SM who received speech and language services.

The Anxiety/Language subtype also had the highest percent of children in comparison to the other

subtypes with a comorbid anxiety disorder. Although some studies report comorbidities with anxiety disorders as high as 74% (Kristensen, 2000), the total percent of children with SM in this study with a comorbid anxiety disorder was only 29%, well under what is often reported in the literature. This supports the notion that not all children with SM are anxious, and that it is only in a subtype of SM that high anxiety will be found.   The Anxiety/Language subtype had the highest mean score for Anxiety, which could exacerbate their language difficulties. However, surprisingly, when examining questions for parents in regard to their own shyness and anxiety, the Anxiety/Language subtype had the lowest percentage of parents reporting these problems. In fact, both mothers and fathers of the Anxiety/Language subtype denied being mute as children themselves. Although parents could be in denial about their own anxiety problems, children with SM in this subtype were reported to have the highest percent of environmental stresses compared to the other subtypes, which could be the source of their anxiety. This implies there may be strong environmental variables whereas; the genetic family history of anxiety may play less of a role in relation to some children with SM (Dummit et al., 1997; Kristensen, 2000).

Often children with SM experience difficulties in school. Sometimes this is social and other times it is academic, or a combination of the two. Therefore it is not surprising that a Low Functioning mutism subtype emerged. Hallmarks of this subtype are academic problems as compared to the other subtypes. These children also experience higher levels of both Sensory and Executive problems, and have the largest percentage of IEP's. This subtype affects just as many females as it does males with an even gender ratio. This subtype also has the largest percentage of mothers who were mute as children and maternal depression. This could be a factor in the low functioning children in this subtype, if the mother is not engaged, attends regular school functions, or will advocate for their child.

Lastly, a Global mutism subtype was found, which most likely would encompass the majority of children with SM. The Global subtype was the largest group (*n* = 71). This suggests that this profile may be more typical than the other subtypes, and that the subtypes above are relatively rare in comparison to this group of children with SM. These children also have the highest percentage of high school children with SM, which may suggest that developmentally these children with SM were in a different subtype as younger children. However, with growth and maturity, problems typical in a different subtype lessened for them, and they transitioned to the Global subtype. Not all children with SM are severely anxious, have speech and language problems, are low academically, or experience sensory emotional/behavioral problems. This study supports that although there are many secondary factors that contribute to each subtype, there are indeed distinctions, and characteristics absent in other subtypes. Findings suggest these children may struggle with their self-esteem, yet, the executive, behavioral, and sensory deficits appear to be largely absent. As a result, this subtype may primarily have deficits in social anxiety and communication.

This subtype appeared to have some problems with flexibility. This is not only a common characteristic of children with SM, but also many children withoutSM. However, it should be noted that this subtype was the least impaired in comparison to the other subtypes in most areas. These children may have an elevated level of anxiety, however, it may not be pervasive in all or even most environments, but rather in social performance situations. This assumption would make sense given anxiety is not a hallmark in this subtype, again suggesting there are a large percentage of children with SM who do not suffer from worry, nervousness, or fear, and instead their problems may be largely situational.

Limitations and Future Research

This study utilized a sample of convenience of archival data. The data consisted of parent report only, and there was no control group to compare ratings on the MBRS, so normative comparisons and inter-rater reliability were not possible. The generalization of this study is limited to children with SM from middle to high socio-economic status, due to the expense of the treatment at the SM-art Center.

The archived data did not provide an adequate sample of non-English speaking immigrant families who had children with SM, therefore, the proposed English Language Learner subtype did not emerge, and the minority of children who were bilingual were found in several subtypes. The sample consisted of differing numbers of males and females in the overall sample and within the subtypes, which was expected. The higher percentage of females within the subtypes may have factored into the results obtained and these results may not generalize to a strictly male population. In addition, many of the subtypes had a small sample size, just marginally making the cutoff n =10 for a subtype which decreases the likelihood that results have clearly-established levels external validity.

This study utilized a behavior rating scale that was not externally validated. However, the scale is considered to demonstrate strong content validity as it aligned well with the DSM-IV-TR, and the DSM-5 and was developed based on clinical practice. The scale consisted of a subjective appraisal of children with SM's emotional, psychological, behavioral, sensory, and academic functioning. The ratings were completed by only one parent, so concurrent validity was not established. There was no other validated measure used in this study, so there was no chance for the comparison of functioning, or determine the validity of the characteristics mutism variables or MBRS subscales. Results suggest this study was exploratory in nature, and worthy of replication. It would be interesting to include a true ELL sample to see what, if any unique characteristics emerge. It would also be of interest to look deeper into the factors that maintain SM, and the role parents and teachers and play in the child's progress. Finally, research is needed to see if different subtypes respond to different treatment methods, as this important treatment validity issue was not explored in the present study.

Regardless of these limitations and research needs, results suggest SM is a socially debilitating childhood disorder that affects children in multiple ways, some of which are unique based on the child's SM subtype. Selective Mutism impairs communication in multiple environments, sometimes due to anxiety, but often times there are other cognitive and behavioral factors entrenched that keep the child mute (Mulligan & Christner, 2006). As treating clinicians the goal is to envision, evaluate, and understand the multiple facets and subtypes of this disorder. This would be a huge accomplishment when conceptualizing and testing out treatment options for children with SM.

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Table 1: Basic Demographic Characteristics of Sample

Variable	n	Percent
Gender		
Males	63	34%
Females	123	66%
Grade		
Pre-K	52	28%
Elementary School	111	60%
Middle School	13	7%
High School	7	4%
Self-Identified Race		
African-American	3	1.6%
Asian-American	7	3.8%
Caucasian-American	143	77%
Latino-American	9	4.8%
Biracial	22	12%

Table 2: ANOVA for MBRS Scales Comparing SM Subtypes

MBRS Subscale	G	A/L	LF	S/P	E/B	F	p
Executive							
M	38.69	43.27	45.70	40.23	49.55[a]	3.18	.016
SD	12.13	13.74	11.41	10.31	9.42		
Oppositional							
M	26.48	29.27	26.48	31.14	33.55	3.24	.014
SD	7.57	7.83	9.06	7.55	7.43		
Labile							
M	27.20	35.00	32.48	33.59	38.09[a]	4.04	.004
SD	11.23	10.49	9.30	10.43	12.08		
Anxiety							
M	50.20	55.93	51.39	51.95	52.82	1.74	.144
SD	8.48	6.94	7.33	7.51	7.45		
Flexibility							
M	16.01	14.47	12.70	14.32	13.73	1.52	.201
SD	6.00	6.49	6.20	5.97	6.15		
Sensory							
M	25.27	20.00	40.57[ab]	44.23[ab]	43.73[ab]	21.14	<.001
SD	12.83	11.41	9.80	11.75	9.07		
Self Esteem							
M	74.65	70.40	72.65	74.36	73.82	.322	.863
SD	14.53	19.49	8.26	13.56	15.36		
Academic							
M	51.61	58.87	59.17[a]	56.27	54.64	3.51	.009
SD	9.94	10.17	7.47	13.42	9.01		

Note. [a]Greater than G Subtype.[b]Greater than A/LSubtype . [c] Greater than LF Subtype.
[d] Greater than S/P Subtype. [e] Greater than E/B Subtype.

Table 3: Participant Characteristics on Descriptive Variables by SM subtypes

			Cluster		
	G	A/L	LF	S/P	E/B
n	71	15	23	22	11
Gender (%)					
Female	66	67	52	50	91
Male	24	33	48	50	9
Grade (%)					
Pre-K	41	20	48	9	27
Elementary	48	73	52	82	73
Middle	.07	7	0	9	0
High	.03	0	0	0	0
Race (%)					
African-Amer.	3	0	0	0	0
Asian-Amer.	1	7	4	9	0
Cauc-Amer.	76	67	83	64	82
Latino-Amer.	4	7	4	9	9
Biracial	14	20	9	18	9
ADA/504 (%)					
Yes	6	13	9	5	0
No	93	87	83	96	100
IEP (%)					
Yes	18	27	30	27	27
No	80	73	61	73	73
Ear Infections (%)					
Yes	32	27	39	32	27
No	68	73	61	68	73
Speech Impediment (%)					
Yes	3	20	8	14	18
No	67	80	87	86	82
Age First Talk (%)					
12 m & Below	51	40	30	36	27
13 m +	42	40	65	64	64
Delay Speech Milestones (%)					
Yes	94	73	87	82	100
No	6	4	13	18	0
Delay Motor Skills (%)					
Yes	9	7	9	18	0
No	92	93	91	82	100
Bilingual (%)					
Yes	20	27	22	27	9
No	80	73	78	73	91
Speech/Language Diagnoses (%)					
Yes	4	20	9	18	9
No	67	80	91	82	91
Sensory Integration Diagnosis (%)					
Yes	6	13	13	22	18
No	97	87	87	77	82

Table 3 Continued

	G	A/L	LF	S/P	E/B
Anxiety Diagnosis (%)					
Yes	21	53	22	46	36
No	79	47	78	55	64
Trauma (%)					
Yes	30	53	13	23	27
No	69	47	87	77	73
Separation Anxiety Diagnoses (%)					
Yes	59	67	65	73	82
No	31	33	35	27	18

Note. G = Global; A/L = Anxiety/Language; LF = Low Functioning; S/P = Sensory Pathology; E/B = Emotional/Behavioral