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ORIGINAL CONTRIBUTION

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'More than 100 years of silence', elective mutism

A review of the literature

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Abstract Elective mutism is a rare disorder of communication, where the child speaks fluently in familiar situations, such as home, despite lack of speech in less familiar settings, for example school. A variety of temperamental and behaviour characteristics, co-morbid psychiatric conditions, neurodevelopmental delay and family factors have been associated with the disorder. EM children are described as excessively shy, withdrawn, 'slow to warm up', inhibited, often avoid eye contact, fear social embarrassment and experience significant separation anxiety, on separation from their attachment figures. Their behaviour is often perceived by others as controlling and oppositional. Onset of EM is typically in early childhood years. A number of constitutional and environmental factors have been considered in its onset, progression and response to intervention. Treatment is generally considered to be multimodal, and occurs in a variety of settings, including home and school. Longterm studies suggest that communication difficulties may extend into adulthood. In addition, outcome studies showing a high rate of phobic disorders suggest that EM may be a developmental precursor of adult social phobia. This article reviews the literature on EM, its presentation, aetiology, epidemiology and the various evidence based biopsychosocial treatments.

Key words elective mutism – social phobia – psychosocial interventions – medication

Introduction

Elective mutism (EM) is a rare disorder of communication, characterised by a persistent failure to speak in specific social situations, where speech is typically expected, (e.g. school), despite fluent speech in other more familiar situations, (e.g. home) [6].

EM was first described by Kussmaul in 1877 and termed 'aphasia voluntaria' [54, 58]. Tramer [86] coined the term 'elective mutism', to suggest that the child elected to talk to certain people, but not to others [54]. In 1994 the DSM-IV changed the terminology to selective mutism [6], emphasizing the fact that the child is only mute in select environmental contexts [24, 28, 81], and suggested five criteria for the diagnosis of the disorder.

1. Consistent failure to speak in specific situations (at which there is an expectation for speaking, e.g. at school), despite speaking in other situations.
2. The disturbance interferes with educational or with occupational achievement and with social communication.

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3. The duration of the disturbance is at least 1 month (not limited to the first month in school).
4. The failure to speak is not due to a lack of knowledge of or comfort with the spoken language required in the social situation.
5. The disturbance is not better accounted for by a communication disorder (e.g., stuttering) and does not occur exclusively during the course of a developmental disorder, schizophrenia or other psychotic disorder.

Elective mutism remains the diagnostic label in ICD-10 [90]. The criteria for diagnosis are similar to DSM-IV, but do not include impairment criteria. For the purpose of this review the term elective mutism (EM) will be used.

Elective mutism: the situational context of the disorder

Patterns of communication vary from children to children, with some children mute in all situations total mutism, and in rare situations children may speak at school, but not at home [89].

The school environment is the most frequent social context where the electively mute child manifests symptoms of the disorder [14, 83]. The teacher is usually the person the child is least likely to speak to [14, 55], and mutism is more commonly presented in the classroom environment than the playground [55]. Some EM children speak to no peers in the school setting, others speak to a select few [83]. The family is the context where children with EM are less symptomatic, and in cases where the child withholds speech at home, the father is the most likely person who is not spoken to [83].

Sudden loss of speech in all settings is called hysterical mutism rather than selective mutism. It usually supersedes a traumatic event, is short lived and not preceded by shy inhibited behaviour.

Epidemiology

Elective mutism is generally considered to be a rare disorder with reported prevalence rates between 0.3 and 7.1 per 1,000 children [11, 17, 19, 31, 47, 48, 55]. This figure is based on limited evidence and varies between studies depending on the population sampled and the definition used. Community based studies yield consistently higher results, as do those using less strict criteria than DSM-IV, and those sampling children within a narrow age band (4–6 years) when EM is more likely to present. EM is up to four times more prevalent in immigrant versus native populations [29].

It is more common in girls than boys, with a female to male ratio ranging from 1.2–1 [83] to 2:1 [40, 88, 89]. It is usually first identified in the early school years, between ages 6–8 years [40, 47, 83, 88, 89], although retrospective studies would indicate that symptoms have been present since the age of 3 [73, 75]. Despite this the average rate of referral to specialist services varies between 6.5 and 9 years of age [30, 55, 81], indicating the long time lag between initial identification to referral.

Aetiology

Although the cause of EM is largely unknown, it is thought that both genetic and environmental factors contribute to the aetiology, presentation and response to treatment. Current conceptualization of the disorder links EM to social anxiety disorders [64], in particular social phobia (SP) [8, 13, 28, 50, 93]. Children with EM have been described as slow-to-warm-up or behaviourally inhibited in infancy and early childhood years [14, 28, 30]. In school they have expressed a fear of being judged, together with physical symptoms of anxiety in social situations [81, 87], and avoidance type behaviour when confronted with the feared situation. Diagnostic studies of children with EM indicate that up to 97% meet criteria for social phobia defined by the DSM-IV [5, 8, 14, 28, 50, 93]. In addition a family history of SP and EM has been reported in 70% and 37% of first degree relatives studied [12]. In deed these authors have suggested that EM should be viewed as a subtype or early developmental expression of social phobia rather than a separate diagnostic entity.

EM has being associated with a variety of additional child psychiatric conditions. Several studies report an association between EM and enuresis and encoperesis [47, 50]. Other symptoms linked with EM include, separation anxiety, obsessive-compulsive symptoms and somatic complaints, [26, 30, 50, 83]. Gillberg and Gillberg [34] report a higher than chance association of EM with autistic spectrum disorders.

A delay in language development, or speech disturbances are factors which may contribute to the development of the disorder. Up to 68% of children with EM have speech and language delay [23, 50], and approximately 68.5% of clinic referred samples meet criteria for neuro-developmental delay [50]. Although cognitive functioning is usually average or above average [40, 42], some studies have reported EM in children with learning disability [56, 71].

Families of EM children have been described as isolated and shy [14, 51, 82]. A number of studies

have reported a very strong bond between mother and child [88, 89], causing difficulties in separation and individuation. Hayden [40] coined the term 'symbiotic mutism' to describe the parent–child relationships of his case series, and recommended family therapy as a treatment intervention.

Some studies have reported the development of EM secondary to a traumatic event 402, [83], or hospitalization in early childhood [92]. Andersson and Thomsen [7] reported a traumatic event during the crucial years of speech development in one third of a clinical sample of EM children.

The aetiological nature of the aforementioned predisposing and precipitating factors remain unclear. The increased prevalence of communication deficits in families with EM [82], the close relationship between EM and SP and high rates of developmental delay suggests that a genetic vulnerability may play a role in the development of the disorder which may be compounded by the environment in which these children inhabit, often characterised by geographical or social isolation and modelling by anxious and over-protective parents.

Cohan et al. [25] suggest a developmental trajectory where a child who experiences a high level of anxiety (either because of a high genetic loading for anxiety, behaviourally inhibited temperament or unstable home environment), may have a heightened sensitivity to verbal interactions with others (due to developmental immaturity, a communication disorder or immigrant status) [29, 83]. This may be triggered by an environmental stressor (interpersonal trauma or school entry), leading to a failure to speak in a given setting, despite the ability to do so.

Assessment and diagnosis of elective mutism

The diagnostic assessment of elective mutism requires a multidisciplinary assessment across a variety of settings. As EM is typically not diagnosed until the child commences school, following a period of engaging in normal conversation with family members in the home setting, the teacher is an important informant as to the child's ability to communicate in the classroom.

Many children with EM have pre-morbid speech and language difficulties [83], and speech and language delays are thought to predispose to the development and maintenance of the disorder. Speech and language functioning is assessed by a speech and language therapist [85]. A detailed history from parents, observation of communication (verbal and non-verbal) in a variety of settings, and the use of audio and video-tapes can assist in assessing speech and language ability.

Although children with EM are generally considered to be of average academic ability, EM can occur with learning disability, and a psychologist is best placed to determine level of cognitive and adaptive functioning.

Baseline rating scales useful in assisting the diagnosis, include Selective Mutism Questionnaire [10], a parent self-report measure of SM behaviours and associated impairment, and the School Speech Questionnaire (SSQ) [10] a modified version of the SMQ specifically focused on speaking behaviours at school.

The clinician's global assessment scale (CGAS) [76] is a useful measure of the child's functional impairment as a result of EM.

As up to 100% of children with EM have an additional anxiety disorder [26, 28, 50, 83], in particular social phobia, the social anxiety scale for children revised (SASC-R) [59] is a useful self-report measure of social anxiety, or the Spence Anxiety Rating Scale (SCAS) [80], completed by either parent or child depending on age.

Other rating scales reported to be of use in the school environment in assessing the EM child include the Teacher's report form (TRF) [1], the Children's Global Assessment Scale-Non-Clinician Version (CGAS-NC) [76] and the Strengths and Difficulties Questionnaire [10, 38].

Psychopathology and comorbidity

Children with elective mutism are described in the literature with a wide array of different symptoms (shy, anxious, dependent, clingy, fearful, sensitive, timid, depressive, 'frozen with anxiety', stubborn, disobedient, controlling, demanding and oppositional) [42, 47, 49, 53, 89]. In clinical practice, they present as shy, inhibited, often appear to freeze when spoken to, blush, avoid eye contact, cling to their parents and resist parental separation [42, 60]. Recent reports using validated rating scales [51, 83], and relatively large samples of EM children identify anxiety as a hallmark of EM [87], and suggest that it may be an early expression of social phobia [8, 27, 50, 64].

Although EM is associated with high rates of comorbidity, mutism itself impacts on social adjustment at a crucial stage of development, which if it persists may cause long-term difficulties in peer interaction and academic achievement. As mentioned previously, many children with EM have premorbid speech and language difficulties [23, 50, 83], and few studies report a neurobiological immaturity in cognitive and motor function [52]. In addition, enuresis and encoperesis has been reported in up to 42% of children sampled [7, 50, 83], reflecting a link between

EM and neurodevelopmental delay. Other authors have reported an excess of obsessive compulsive behaviours [40, 50, 88].

There have been a number of case reports of EM in children with chromosomal abnormalities in particular Fragile × syndrome [39] chromosome 18 abnormality [78] and learning disability. Interestingly, autistic spectrum disorder has been found to occur more frequently in EM families [34].

Family characteristics

Families of EM children have been described as shy and reticent [19, 82, 86, 88]. EM has been identified in other family members in up to 50% [12, 50, 62, 68, 82], and social phobia in 70% [12, 50]. Taciturnity in either parent has also been reported in up to 50% [33, 82]. Sharkey and Mc Nicholas [77] described a case of female monozygotic twins in which EM was present in three generations of the family. Parental internalizing disorders and personality disorders have been shown to be an important correlate [14, 51] and outcome predictor [47] among children with EM. Kolvin and Fundudis [47] reported depression in 21% of fathers and 17% of mothers in their study.

A number of case reports in the literature have suggested a link between elective mutism and family's concern about the disclosure of a family secret [42, 60, 61]. In extreme cases EM has appeared to post-date child sexual abuse [3, 40]. While this association is rare it needs to be considered if there are concerns about the child's inappropriate behaviour or parent's inability to care for the child.

Case reports have suggested a link between EM and family dysfunction [35, 91]. Goll [35] described a case series of 10 EM families in which a child presents with EM, and identified individuals in these families playing four specific roles (a) 'the elective mutest'; (b) 'the mutest model or (models)', (c) 'the symbiotic partner' and (4) 'the leader of the ghetto family'. The 'symbiotic partner' (usually the mother) forms a coalition with the 'elective mutest', and this 'close relationship is directly related to the hostile and unsatisfying relationship between mother and father' [20]. Both feel that they cannot survive without the other [65], and the child has great difficulties separating to commence school [70, 92], and individuating to become a separate entity. Mothers are often described as overprotective and over-involved, and fathers as detached. Sibling rivalry is very common [92]. Some parents link the onset of mutism to the birth of the younger sibling [2, 4]. The incidence of EM is increased among siblings, and this may reflect a genetic transmission [82], modelling or vying for the increased attention and affection that is often given to the sibling with established EM. Goll saw the main problem as the family's interaction with society and wondered 'if this constellation of roles was caused by the existence of EM in the family, or whether the family caused the EM's symptoms' ([35], p. 63).

Cline and Baldwin observed that families of EM children are typically characterised by strong tensions and marital discord [89, 91]. Communication is often terse, restricted and unsatisfying. Despite this they usually stay together, but are often unhappy. They often isolate themselves socially and are wary of the outside world and strangers. Parents lack contacts outside their immediate family circle and these contacts are generally very intense. At the clinic they usually deny any relationship difficulties, and it is thought that the child's behaviour serves to divert attention from their own relationship [74]. In summarising the work of ten families Rosenberg and Linblad [74] observed that the atmosphere in the homes was not conducive to expression of feelings, and that open displays of affection between family members were usually absent.

A number of studies have examined the structure of EM families, but taken together the results are inconclusive. It seems that EM depends on subtle dynamic interactions that bear no relationship to family size or birth order.

Management of elective mutism

Most of the literature pertaining to elective mutism consists of single case reports or small case series. Among these reports a wide variety of treatment strategies have been described, including individual, group and family approaches. More recently there have been reports of pharmacological treatment of EM [22, 36, 37, 92]. Treatment outcomes have been inconclusive at best [53, 92]. A persistent message conveyed in the clinical literature, is that EM is difficult to treat [47, 79], and that the optimal intervention strategy may be a long-term, multimodal, multifaceted therapeutic approach [41]. This section describes a review of the literature of the treatment of EM.

Individual psychotherapy

A number of different psychotherapeutic approaches to the treatment of elective mutism have been reported in the literature. For the most part, only case reports of the efficacy of these therapies have been published. The psychotherapeutic methods used have included, psycho-analysis [69, 88], psycho-analytically orientated art therapy and play therapy. More recently, cognitive behavioural therapies [5] have

been adopted, and have been reported to be useful, whereas psychodynamic approaches have been de-emphasised.

Behavioural and cognitive behavioural therapy

Elective mutism has been described as a learned response that the child develops to manage anxiety [72]. Reed used systematic desensitization and stimulus fading to treat a 6-year-old girl with EM [72]. Nolan and Pence [66] described a behavioural programme used to treat a 9-year-old girl with EM. A functional analysis revealed that the whole school reinforced the child's behaviour by making allowances for her. Operant conditioning was used to ignore silence and reward increased communication. Two years post termination of therapy the child was speaking freely in a variety of situations.

Self-modelling has being described as a useful intervention in the treatment of EM [15, 46]. This involves the making of video and audio-tapes that show the child speaking in settings where he or she previously remained mute. These tapes are repeatedly played throughout treatment, with the hope that the child will become accustomed to hearing him or herself speaking in these settings and will begin to believe in his or her ability to do so. Positive results have been reported at 6 and 9 month follow up [45, 46]. Kee et al. [44] described the use of electronic equipment to aid voice production as part of a multimodal treatment programme in the treatment of a 6-year-old boy with EM.

Subsequent to these earlier case reports, a number of other case studies have been reported using operant techniques. Porjes [69] summarised the literature supporting the use of CBT in the treatment of EM. He observed that therapy needed to take place in school and required organized coordination between teachers and psychologists in order to ensure success. He also noted that treatment was more successful in younger children where academic achievement and the development of peer relationships are not compromised.

More recently, Fung et al. described an internet based cognitive behavioural programme for the treatment of EM [32]. This programme includes a web-based child work book and notebook and a downloadable parent–teacher manual which focuses on psycho-education. The treatment which consists of 14 individual weekly sessions, teaches the child to recognize signs of anxious arousal associated with speaking and to use specifically recommended anxiety management strategies. Johnson and Wintgens published a very practical manual based intervention for the assessment and treatment of EM in home and school settings [43].

Calhoun and Koenig [21] described the only controlled study of the use of behavioural therapy in the treatment of EM. In this study eight children were randomly assigned to treatment and control groups and number of words per 30 min period were collected by trained observers at baseline, posttreatment and followup. Treatment consisted of teacher and peer reinforcement of verbal behaviour. Children who received active treatment had significantly more vocalizations than untreated 5 weeks after the start of treatment, but improvement was not significant at 1 year follow-up.

Play therapy

Although no direct reports of the use of play therapy in the treatment of elective mutism have been reported in the literature, several authors have used play therapy techniques as part of a multimodal treatment approach [9, 92].

Group therapy

Group therapy for electively mute children is rare, due to the limited number of cases referred for intervention at any one time. Bozigar and Hansen [16] reported the use of group therapy for four children with EM. The group met weekly for 1 h, over a 2 month to 1 year period, until the children were speaking freely in all settings. The treatment programme involved close liaison with parents and generalisation of treatment gains to the classroom setting. At the end of the treatment period all children transferred to a new school, and were free of symptoms. The children were followed up periodically over 1 year, and remained symptom free in all settings. These authors believed that close cooperation with schools was imperative to the success of therapy.

Barlow et al. [9] described the use of sibling group play therapy as part of a multimodal treatment of a 5-year-old girl with EM. The addition of siblings to the play therapeutic milieu provided a more familiar setting for self-expression and an environment in which the child could model changed behaviour.

Wright et al. [92] used an intensive short-term intervention over a 6-week period to treat three preschoolers with elective mutism. The authors suggest that identification and intervention at an early stage before the behaviour becomes entrenched may optimize treatment outcome and improve long-term prognosis.

The authors conducted a group for 5 children with EM and 3 normal speaking siblings using an eclectic model of behavioural modification, shaping, model-

ling, role playing and relaxation techniques. A parallel group was run for parents. Systematic evaluation of the child's speaking and anxiety significantly reduced as a consequence of treatment as did maternal anxiety and low sense of parenting competence.

Family therapy

Literature to support systemic practice in the treatment of elective mutism is sparse, and this may reflect a reluctance of socially anxious or isolated families to interact with outside agencies. Goll [35] described a case series of ten families with a child with EM. He identified factors, including maternal-child overenmeshment and overprotection as important in the aetiology, and used a systemic approach to treatment.

Pharmacotherapy

Since the 1990s, there have been a number of case reports and a few clinical trials that support the use of medication in children with elective mutism, who do not respond to psychosocial measures. Initial reports suggested the use of monoamine oxidase inhibitors (MAOIs) [36, 37], on the basis, that they are useful in treating adults with social phobia, and their activity on the dopamine receptors was thought to increase talkativeness. More recent reports have advocated the use of selective serotonin reuptake inhibitors (SSRIs), as they are less toxic in overdose and are effective in mood and anxiety disorders [13, 22, 28, 92]. Although, most studies report the rapid resolution in symptoms on medication, some of the authors report unpleasant side-effects including behavioural disinhibition, which can be more problematic than the EM symptoms.

In addition the US food and drug administration (FDA), issued black box warnings about SSRIs in 2004, on the basis that they have been reported to cause suicidal thinking in children and adolescents with major depression, in short term studies, although not contraindicating their use. This warning changed prescribing practice in the United States and Europe. Recent evidence suggests that relative to placebo SSRIs are effective in padiatric depressive disorder, obsessive compulsive disorder and non-OCD anxiety disorders including EM. The effects are strongest for non-OCD anxiety disorders, with benefits much greater than risk of suicidal ideation or attempt across all indications [18]. Best practice recommends the use of fluoxetine in children resistant to psychosocial treatment, starting at a low dose and titrating according to treatment response and the emergence of side-effects. In addition clinicians are advised to monitor the child closely, advising parents of possible adverse events, and how to manage them.

Prognosis

There are very few long-term studies in children with elective mutism, and the age at follow-up in most has been younger than 18 years. Most studies include sample sizes less than ten and reveal remission rates of between 39% [73] and 100% [71, 88]. Variable rates are possibly due to differences in the definition of EM, and duration of time to follow-up. Better remission rates are obtained if follow-up is longer than 10 years, possibly reflecting the fact that in most cases the duration of the disorder exceeds 5 years [67]. Remschmidt et al. [73] interviewed 45 patients with elective mutism 12 years after their initial referral. Thirty-nine percent of this sample was in complete remission, and 61% continued to have some communication problems on validated rating scales. The formerly mute patients described themselves as less independent, less motivated with regard to academic achievement, less confidant and less mature in comparison to a normal reference group. Steinhausen et al. [84] found a remission rate of 58% in a controlled study of children followed up approximately 13 years after initial diagnosis. The high rate of phobic disorders detected in their sample at follow up concurs with previous suggestions that EM is a developmental precursor of social phobia.

The results of follow-up studies confirm the fact that EM is a very persistent disorder with a general tendency of poor outcome [47, 73, 84]. Predictors of poor outcome, include difficult family conditions, uncooperative or psychiatrically disturbed parents [33, 47, 63, 79], intellectual impairment of the patient [57, 62, 91], mutism within the core family [73, 84] and cerebral dysfunction [33].

Conclusion

This article provides an up to date review of the literature pertaining to elective mutism.

EM is portrayed as a clinically heterogenous disorder, closely linked to anxiety disorders in particular social phobia. It is possible that an underlying neurodevelopmental vulnerability predisposes the child to the development of the disorder that is expressed in the context of a family environment marked by unusual patterns of interaction, both within the family and with outside agencies. Current therapy combines, behavioural modification, family participation, liaison with school and in some cases medication. The long-term outcome of elective mutism remains unclear.

Literature suggests that these children are at risk for phobic disorder in adulthood. More extensive studies looking at underlying vulnerability factors and long-term outcome of this rare, but handicapping disorder are clearly warranted.

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