Selective Mutism in Children: A Literature Review of Cognitive Behavioural and Integrative Psychotherapeutic Schemes

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Abstract

Selective mutism is a rare childhood psychiatric condition, repositioned under the umbrella of anxiety disorders in the DSM-5. A minor proportion of children is affected by selective mutism, being physically unable to elicit speech across unfamiliar social settings. The disorder's origin and nature are largely unspecified, though comorbidity development, and long-term academic, social, and emotional adversities are evident. Optimal treatment relies on limited published research papers placing emphasis on cognitive behavioural and integrative interventions. The present review aimed at evaluating differences amongst these psychosocial configurations regarding their effectiveness and suitability for treating selective mutism in children. PubMed, Science Direct, APA PsycNet, and ERIC databases were thoroughly searched in March 2020 for the identification of research articles published in peer-reviewed journals throughout the previous decade. Six research articles were retrieved, each assessing cognitive behavioural and integrative modalities. Overall, cognitive behavioural approaches appropriately address multiple levels of children's developmental needs while reinforcing their cognitive-behavioural competences via means of cognitive restructuring and behavioural modification. Still, several omissions such as post-treatment enduring symptoms or improper focus upon anxiety-related aspects of children's psychosocial functionality were discernible. Conversely, integrative approaches offer solid foundations for holistic and personalised treatment plans while consistently involving important stakeholders (parents, school). Yet, their limited exploration has not enabled for their absolute effectiveness, and their predominantly manualised nature hardly stipulates for a uniform approach, hindering successful clinical practice. Therefore, the clinical applications of both psychotherapeutic schemes display major differences instead of complementing each other. Future research should conduct further needs analyses, utilise technology sensibly, and consider cultural variations.

Keywords: clinical psychology, developmental needs, quality of life, selective mutism, social anxiety

1. Introduction

The 5th edition of the *Diagnostic and Statistical Manual of Mental Disorders* re-classified Selective Mutism (SM) within the framework of anxiety disorders (American Psychiatric Association, 2013). SM entails a recurrent persistency of an individual's physical inability to articulate thoughts in various social contexts, especially in novel social circumstances (e.g., school), irrespectively of being unobscured in producing speech within more familiar settings (e.g., home). Most frequently, SM diagnosis eventuates in early childhood whilst children's engagement in social interactions is expected, with SM onset ranging from 2.7 to 4.1 years, whereas resembling referral ages expand to 6 to 9 years (Martinez et al., 2015, Viana, Beidel, & Rabian, 2009).

Estimations of SM prevalence primarily emanated from school-based communities, generating range approximations of 0.03 to 0.79%, thus illustrating the rareness of this psychiatric condition (Sharkey & McNicholas, 2012). Yet, such estimates gradually increase, reaching percentages of 1.9% (Zengin-Akkuş et al., 2018), engendering the acknowledgment of previous SM misdiagnoses/underdiagnoses stemming from screening/assessment instruments not being cross-culturally accessible (Pereira et al., 2019), while reflecting a growing awareness of the disorder's substantiality. SM aetiology is moderately unspecified, although certain identified risk factors embody behavioural inhibition of children's temperaments or a clinical record of previous impediments in speech and language expression (Klein, Armstrong, & Shipon-Blum, 2012; Muris, Hendriks, & Bot, 2016). Besides, the controversial nature of SM is voiced by researchers' qualifying the disorder as a consequence of speech deficits or an avoidance strategy stationed in a behavioural disorder (Holka-Pokorska, Piróg-Balcerzak, & Jarema, 2018).

In cases of continuous persistence of SM-related symptoms following successful resolution of SM, long-term adversities such as disproportionate development of social-emotional and linguistic competences (e.g., communication skills), poor school performance, peer relationships disengagement, and children's residual social withdrawal may inevitably occur (Carbone et al., 2010; Bergman, Piacentini, & McCracken, 2002). These may culminate in increased prospects of comorbid problem internalisation, social distress, anxiety intensification, and seclusion from extracurricular activities. In affirmation, one retrospective study explored childhood experiences of 45 adults formerly diagnosed with SM, eventually reporting social issues, dependency issues, and an extensive reduction of confidence (Remschmidt, Poller, Herpertz-Dahlmann, Hennighausen, & Gutenbrunner, 2001). The subsequent self-selected isolation is, in turn, a contributive factor to the emergence of depressive symptoms and somatic complaints, ultimately hindering children's quality of life (Alyanak et al., 2013). In absence of appropriate and early clinical management, chronic repercussions may emerge, impairing several areas of children's personal, academic, and social functionality (Kamani & Monga, 2020). Thus, interventions ought to improve children's current and prospective quality of life, while properly cooperating with teachers and parents, since a vast majority of SM symptoms is exhibited at school (Bergman, Keller, Piacentini, & Bergman, 2008).

Throughout the previous decade, various pharmacological and psychological interventions were introduced to the literature, all with the fundamental objective to mitigate children's non-speaking behaviours. In this respect, Diliberto and Kearny's (2016) study elucidated endorsements of treatment specifications punctuated for different SM subtypes/subgroups (Mulligan, Hale, & Shipon-Blum, 2015). On account of presumably originating from a multifactorial essence, SM interventions diverge; cognitive behavioural and integrative approaches (both deriving from behavioural theoretical frameworks) incorporating wide-ranging behavioural techniques have been broadly utilised by scientific research, with demonstrated effectiveness. The current review aims at presenting, evaluating, and reaching solid conclusions regarding their efficacy, since their distinct differences and heightened popularity within the SM literature render them appropriate for in-depth investigation. In particular, the effectiveness of Integrated Behaviour Therapy for Selective Mutism, Conjoint Consultation, coupled with three less commonly employed variations of Cognitive Behavioural Therapy will be assessed, in order to provide clinicians, psychologists/psychiatrists, and researchers with beneficial information regarding their clinical utility.

2. Method

The present review includes articles published in peer-reviewed journals in the past 10 years (since 2010), written in the English language. These were identified via the National Institutes of Health National Library of Medicine PubMed, ScienceDirect, APA PsycNet, and ERIC literature search databases. Search terms contained the following words: *selective mutism*, *paediatric mutism*, *children*, *interventions*, *treatment*, *developmental needs*, *quality of life*, *cognitive behavioural therapy*, *integrative approaches*. Search was conducted by using all combinations that were possible between terms (e.g., *selective mutism treatment for children*). Exclusion criteria stipulated that all non-English articles, book chapters, monographs, review articles and meta-analyses, abstracts, letters to editors, articles utilising pharmacological interventions, and all non-peer-reviewed, non-data analytic reports not be taken into consideration. Inclusion criteria required that the selected articles involved one psychosocial/behavioural intervention targeting the developmental needs of children with diagnosed SM. Preliminary identification pinpointed 332 unique records. Following an initial screening of keywords, resource types, and titles, 94 duplicates were filtered out due to their unsuitability for inclusion. The remaining articles were further screened and, in cases of unclear aspects of the articles' abstracts, full text was consulted. Eventually, 6 retrieved research articles were eligible for review, all fulfilling inclusion criteria, with an equivalent number of studies for each intervention. All articles were critically inspected in accordance with the Critical Appraisal Skills Programme (CASP) to ensure that the included studies were of high-quality.

3. Cognitive Behavioural Therapy for Selective Mutism

Cognitive Behaviour Therapy portrays a major SM psychotherapeutic standpoint and extensive research endorses mere CBT provision devoid of pharmacological administration such as anxiolytics, selective serotonin reuptake inhibitors, and fluoxetine (Manassis, Oerbeck, & Overgaard, 2015). Most structured and brief CBT interventions place exponential consideration on cognitive restructuring, implying the modification of children's behaviour and thinking patterns, hindered by the behavioural component of SM symptomatology and its early

onset (Hua & Major, 2016). Various modalities have been assimilated into elaborate developmental CBT adaptations, such as Modular CBT and web-based CBT.

3.1. School-Based Cognitive Behavioural Therapy

Most recently, Oerbeck, Overgaard, Stein, Pripp, and Kristensen (2018) scrutinised a school-based CBT intervention by providing 5-year outcome data for 30 children (8-14 years of age) diagnosed with SM, undergoing CBT for approximately 21 weeks. Researchers aimed to investigate whether treatment outcomes could be maintained 5 years after treatment initiation. Pursuant to the practitioner review recommendations (Cohan, Chavira, & Stein, 2006; Keen, Fonseca, & Wintgens, 2008), the intervention conjoined psychoeducational portions, behavioural engineering, and defocused communication, consisting of two weekly sessions initiated at home for guaranteeing a feeling of safety for the children. Gradual increased exposure (stimulus fading), contingency management, and positive rewards for verbal expression were accentuated and applied through play activities included in the Selective Mutism Resource Manual (Johnson & Wintgens, 2017). Early verbal expression before treatment termination stipulated for discontinuation.

Positive amendments were ascertained in the 1-year follow-up, with comorbid conditions (e.g., social phobia, specific phobia, enuresis nocturna, and separation anxiety disorder) establishing a significant diminution. Five years later, improvements were generally sustained, with 70% of the participants being in full remission, 17% in partial remission, 13% still fulfilling SM diagnostic benchmarks, and 50% experiencing ongoing minor verbal difficulties. Taking this into account, the treatment's potential long-term effectiveness can be postulated. Early intervention in younger children played a decisive role in treatment gains, yet this might allegedly constitute school-based CBT a more younger-age centered approach, considering that active cognitive restructuring would be of prominent value for older children as well which was disregarded by this study. Bilingualism and gender, oftentimes deemed vulnerability factors for SM, did not negatively influence treatment outcomes, hence indicating the treatment's efficacy for a wider spectrum of children than previously assumed (Gensthaler et al., 2016). Notwithstanding the foregoing, in comparison to preceding studies (Lang et al., 2015; Klein et al., 2016), practitioners had no proper training for CBT provision. Although findings were of significant effect, this draws questions regarding whether there could be an alteration of the results if trained practitioners were involved.

3.2. Modular Cognitive Behavioural Therapy

Another compelling publication by Lang et al. (2015), a retrospective naturalistic study, presents the 1-year outcome data of a specifically designed Modular Cognitive Behavioural Therapy (MCBT), aiming to improve SM enduring symptoms while reducing psychiatric comorbidities. Sample size amounted to 24 children (5-15 years of age) diagnosed with SM. Treatment duration exhibited a prolongation of grades (from preschool to first grade) as researchers aimed at completely curing SM instead of solely improving/alleviating symptoms. The adopted approach was centered upon separate modules, flexibly employed by clinicians, for the formation of tailored treatment plans for participants. Psychoeducational programs, presupposing that SM is a manifestation of social anxiety, for both parents and children were included, along with physiological, cognitive, and behavioural training. More precisely, muscle

relaxation and breathing techniques, cognitive restructuring and symptom externalisation, and parental training for enhancing children's motivation (thus facilitating the discontinuation of non-speaking behavioural patterns and promoting resilience) were cautiously applied. In contrast to Oerbeck and colleagues' (2018) study, staff working in school and recreational facilities were trained as facilitators of the intervention. All children were entitled to both in-school and parental guidance.

Robust clinical improvements from baseline to follow-up were observed in each situational dimension (at home/with family, at school, in public/social settings) and social anxiety symptoms, maintained for *circa* 2 years after treatment termination. Psychiatric comorbidities (social anxiety disorder, specific phobia, enuresis nocturna) showed a significant decline over time. Still, a minor proportion of children resumed fulfilling diagnostic criteria for social anxiety disorder following SM recovery. Such findings support MCBT's feasibility for children with SM, with promising long-term outcomes. Contrary to previous notions that cognitive techniques cannot be employed in preschoolers due to their cognitive functions being underdeveloped (Schwartz & Shipon-Blum, 2005), researchers applied them successfully, perhaps further addressing various cognitive aspects of SM mentioned in the literature (Capobianco & Cerniglia, 2018). Conceivably though, MCBT cannot focus upon distinct social anxiety disorder facets impertinent to verbal expression (e.g., performance). Underlying reasons for occurring attrition rates in children exhibiting considerably more acute SM symptoms remain uninvestigated; the treatment's partial inefficacy, imprecise adaptation to their needs, or other reasons may have resulted in dropouts. MCBT addresses several SM multidimensional developmental configurations (Reuther et al., 2011), but its effectiveness inclines towards alleviating persuaded symptoms instead of totally curing SM.

3.3. Web-Based Cognitive Behavioural Therapy

The third article in review (Ooi et al., 2016) is a pilot randomised controlled trial discussing the efficiency degree of a 14-week web-based CBT program entitled 'The Meeky Mouse Program', with electronic interface aiming at further developing children's learning and coping strategies, decreasing their anxiety levels, and ameliorating clinician-rated severity of mental illness. Twenty-one children (6-12 years of age) with SM were randomly allocated to either 14 weeks of web-based CBT or to 14 weeks of in-person therapist interaction with simultaneous use of computer games. Eight training sessions were introduced, including anxiety management courses coupled with 6 practice sessions. Social competences training exposure encompassed evaluating one's thinking patterns, developing confidence, and employing the CHAT[1] plan towards improving social communication. Progressively demanding and anxiety provoking tasks entailed children's homework plus parents' active involvement. Children's messages were recorded and replayed in sessions. Through hearing their own voice, desensitisation to speaking occurred.

Intriguingly, in follow-up analyses, significant improvements were noted in the control group regarding pre- and post- treatment total speech frequency. Parent-rated anxiety symptoms revealed non-significant improvements for the Meeky Mouse group, while for the intervention

[1] Check your body's feelings, Having bad thoughts, Attitudes and Actions that can help, Time for a reward

group, clinician-rated mental illness severity scores were improved significantly. These results do not support provision of web-based CBT for children with SM. Although literature has assembled convincing justifications for alternative CBT-based interventions application for anxiety reduction and speech reinforcement (Fung et al., 2002; Reuther et al., 2011), the findings did not verify these prospects. Past research potently encourages parental training (Khanna & Kendall, 2009), howbeit, in this study, an apparent unsuccessful incorporation of parents' psychoeducation component and their scarcity of necessary task-related coaching acquisition is prominent, generating a probable interference with the end results. Admittedly, electronic interface omitted children-therapist physical communication, thus effective parental involvement could beneficially ease their interface experience, thus constructing a more naturalistically sound study.

4. Forwarding Approaches to Treatment: Integrative Interventions

Integrative interventions, although more narrowly explored, deliver treatment via means of multimethod behavioral techniques and ecobehavioural developmental models (Mitchell & Kratochwill, 2013). The principal purpose is to elicit speech across settings (Bergman, 2013) while tenaciously considering child input proceeding from multiple informants such as parents, clinicians, and other adults in the child's surrounding environment, who assemble to formulate treatment plans mainly comprising gradual exposure assignments. Their distinct difference from CBT is that stakeholder involvement is mandatory, acting as the intervention's guiding agents.

4.1. Integrated Behaviour Therapy for Selective Mutism

The first research paper by Bergman, Gonzalez, Piacentini, and Keller (2013) aimed to assess the efficacy, acceptability, and feasibility of Integrated Behaviour Therapy for Selective Mutism (IBTSM) regarding SM symptom reduction and speech dysfunctionality improvement. Twenty-one children (4-8 years of age) with primary SM were randomised to either 24 weeks of IBTSM or 12-week waitlist control. Results were reviewed by blind external evaluators, teacher and parent reports, and structured behavioural measures. Sessions followed a detailed treatment plan, described in the correspondent IBTSM manual (Bergman, 2013), comprising 20 1-hour sessions. The intervention's behavioural component consisted of gradual exposure to feared stimuli, in-session exercises that became progressively demanding for speech promotion in public settings, and behavioural control tools. Parental and teacher/instructor involvement was constantly assimilated. Designated cognitive restructuring principles were occasionally introduced, depending on each child's developmental level.

Significant improvements concerning speaking behaviors were reported by independent evaluators and teachers for children in the IBTSM condition post-treatment compared to waitlist participants, and 67% attained a SM diagnosis removal. Follow-up analyses revealed solid maintenance of treatment gains. Nevertheless, comorbid conditions (social phobia and social anxiety) demonstrated undeviating persistence. Consequently, discernible propitious clinical outcomes strengthen the initial evidence of IBTSM's effectiveness/provision for children with SM. Amalgamating multiple informants, blind evaluators, and school involvement (though perhaps not used sufficiently in terms of resources) enabled for a more holistic and naturalistic approach applied through collaboration between children and their natural settings. Yet, IBTSM

could not address social anxiety, increasing conjectures of a complex relationship between SM and social anxiety/social phobia (Gensthaler et al., 2016). Furthermore, missing elements, namely additional measures for children's academic or social functionality post-treatment, signify concerns regarding the absolute suitability of IBTSM to examine all aspects of children's developmental needs.

4.2. Replicated Single-Case Integrated Behaviour Therapy for Selective Mutism

Siroky, Carlson, & Kotrba (2017) conducted a replicated single-case study to investigate IBTSM effects in two cases of 4-year-old boys diagnosed with SM, aiming at increasing speech and relieving anxiety. A trained and experienced SM practitioner delivered 20 sessions for 6 months using the IBTSM manual. This study opted for a shorter version of IBTSM, as recommended by Bergman et al. (2013), consisting of 45-minute therapy session in a 12-week span, while preserving the original intervention's fundamental elements (Bergman et al., 2013). Parents' role was prominent during the last sessions and they were given resources in case of potential post-treatment relapse.

SM and social anxiety symptom severity significantly declined for both cases, with verbal communication ameliorating. At 3-month follow-up, there was increased and maintained speech frequency for both cases, though one child continued satisfying SM prerequisites. Parental satisfaction was high, characterising IBTSM as effective and feasible. On this account, results partially support short-term IBTSM provision. Authors comment on the treatment duration's inadequacy for the second child to display significant improvements, since his baseline severity was higher. This is supported by research indicating that the greater the symptom severity at baseline, the fewer gains are substantially obtained (Oerbeck et al., 2015). Conversely, in Bergman and colleagues' (2013) study, treatment gains were sufficiently obtained in 12 weeks, leading to implications concerning the optimal treatment duration's tentativeness. Noteworthily, in both studies, IBTSM's manualised nature may have affected optimal clinicians' practice. Accurate instructions with regards to in-school progress monitoring and unceasing communication with gatekeepers are marginally endorsed by the manual, not ensuring children's proper academic development as well (Muris & Ollendick, 2015; Oerbeck et al., 2015).

4.3. Conjoint Consultation

Following similar integrative principles, Mitchell and Kratochwill (2013) investigated an 11-week treatment protocol's (Conjoint Consultation) implementation on four children (5-10 years of age) diagnosed with SM using multimethod behavioural techniques through a replicated, randomised multiple-baseline design, endeavoring to improve participants' speaking behaviours and reduce their anxiety levels. Services were delivered indirectly by consultees (parents, teachers, and other significant adults within a child's life), integrating standardisation and generalisation of the treatment by its initiation in a clinic setting and its subsequent progression to school settings.

Children's anxiety levels did not change significantly over the course of the study. However, positive results with minimal increases in speech for two children were observed, and modest decreases in SM severity for all children, as stated by impartial observers. Treatment generally received positive evaluations by parents, deeming it appropriate. However, the intervention's

plausibility of provision remains debatable. Most notably, not all participants were necessarily present in all sessions due to time constraints, hence the possibility of them not receiving full treatment effects appears as a predominant restriction. Although all children were qualified as treatment responders, researchers commented a descending response rate trend, for which they claim that treatment's progressive demands may be the underlying cause. This, however, is contradicted by the aforementioned studies in which treatment demands did not pose obstacles. Therefore, although profound developmental theoretical background is offered, integrative treatment is not always clinically applied successfully.

5. Discussion

Investigation of current SM treatment modalities spawns several critical conclusions. Generally, there are apparent issues *vis-à-vis* the absence of prospective long-term outcome studies with larger sample sizes, highlighting the requirement of developing and validating more universally accepted SM metrics and adequate interventions for the percentage of children with recurrent SM symptoms (Oerbeck et al., 2018).

CBT interventions address numerous levels of children's needs while reinforcing their cognitive and behavioural competences via means of cognitive restructuring and behavioural modification, indispensable for typical development (Capobianco & Cerniglia, 2018). However, behavioural avoidance facilitating social anxiety persistence and social anxiety dimensions impertinent to verbal expression (e.g., performance) may account for post-treatment enduring symptoms (Lang et al., 2015). Consequently, CBT steers clinical attentions towards certain vital aspects of children's psychosocial functioning (Bergman, Piacentini, & McCracken, 2002) and cannot focus upon anxiety-related ones (developmental difficulties, language, oppositionality) (Muris & Ollendick, 2015). Thereof, an enrichment of CBT's aims/objectives or a singular treatment proposition is strongly recommended to further foster comorbidity diminishment. Moreover, research speculations of SM's relations to a behavioural inhibition's personality trait or taciturnity cannot entirely offer insight to post-treatment incessant verbal obscurities (Gensthaler et al., 2016), potentially inferring that speech anxiety remains unalterable and CBT augmentation and/or prolongation is thus necessitated. Unambiguously though, CBT arrangements *per se* should further consider separate parameters such as parental psychopathology (Koskela et al., 2020) or exposure tasks' inadequate practice/duration (Furr, Sanchez, Hong, & Comer, 2020), so as to be structured more suitably.

Integrative interventions appear as relatively developmentally sensitive while consistently involving important stakeholders, corroborating widespread research attaching importance to active school and parental participation throughout the therapeutic process (Bergman et al., 2008; Bergman, 2013). Regardless of the observed improvements in functionality impairments, social anxiety did not dwindle, advocating an eventual multifaceted correlation between SM and social phobia (Vogel et al., 2019). Put differently, the two share similar attributes that cannot be easily distinguished into one integrated plan, thus requiring different therapeutic regimens. Optimal treatment duration needs to be further re-evaluated to successfully address various baseline severities. Thus far, the deduction that lengthier treatment schemes are more beneficial to children can be considered to target their adaptation to each therapeutic demand. Another raised issue concerns treatment delivery's consistency. In all cases, the use of manuals

hardly stipulated for a uniform approach, thus potentially hindering successful practice, especially when executed by consultees (Siroky et al., 2017; Mitchell & Kratochwill, 2013). An enrichment of the manuals is of paramount significance to tackle a detailed explanation of the exact implementation of behavioural strategies.

SM treatment has been brought under the microscope in recent years, with central focus located on CBT approaches and lesser on integrative ones. The present review offers important considerations over them, adding to the body of literature by identifying that, instead of complementing each other, their clinical applications present major differences. CBT has already been repeatedly employed with other anxiety disorders, and its favourable outcomes on SM are indisputable, while integrative plans enclose solid theoretical foundations not yet effectively applied.

Overall, the conceptual clarity of SM remains controversial, imperiling the purposefulness of the aforesaid interventions. Future recommendations include shedding light on the condition's nature, even reaching a neuroscientific core (Henkin, & Bar-Haim, 2015), and needs analysis assessment should be conducted to allow for an effective intervention planning and recognition of distinctions between SM and social anxiety. The utilisation of technological input to further support individualised treatment plans (Bunnell, Mesa, & Beidel, 2018; Bunnell & Beidel, 2013) needs to be approached with caution in light of controversial findings. It is also advised that cultural discrepancies be considered (Starke, 2018), as they may require adopting different treatment plans catered for children's cultural needs. Lastly, increasing the number of published studies will allow for necessary comparisons amongst novel treatment approaches (e.g., Intensive Group Behavioural treatment, Parent-Child Interaction treatment, Psychomotor treatment) (Cornacchio et al., 2019; Catchpole et al., 2019; Esposito et al., 2017) to determine their efficiency towards the elimination of long-term SM repercussions in children.

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