

Selective mutism – an overview of the condition and etiology: is the absence of speech just the tip of the iceberg?

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Summary

The inability to speak in certain situations, as one may briefly characterize selective mutism (SM), according to the most recent classifications (DSM-5, ICD-11) belongs to the anxiety disorder spectrum. The onset of mutism in early childhood may impair further development and adversely affect educational achievements. It is essential that psychiatrists, as well as other physicians, speech therapists, nurses and teachers are familiar with this disorder, since the early start of treatment is associated with better prognosis.

This literature review aims to present the contemporary view of this relatively rare psychopathological syndrome. In light of most recent studies on the etiology of SM, the sole symptom of mutism appears to represent an underlying heterogenic group of disorders. Based on the developmental psychopathology, the interrelations between overlapping abnormalities favor SM manifestation in some crucial moment in an individual's life. The etiologic complexity strongly suggests multimodal approach in the diagnostic and treatment process, which has been postulated by many authors.

Key words: selective mutism – SM, anxiety disorder, autism spectrum disorder – ASD

Introduction

SM was first described by Kussmaul in 1877 as *aphasia voluntaria*. Nearly sixty years later, in 1934, Tramer coined the term *elective mutism*. Both these names emphasized the presumably voluntary refusal to communicate verbally with most people, explained by first authors as attempts of manipulation. Even as late as at the end of the twentieth century, SM was considered a type of oppositional defiant disorder (ODD) [1]. Only with the update of the DSM by the American Psychiatric Association in 1994 (DSM-IV) this concept changed, as illustrated by the replacement of the word 'elec-

tive' with term 'selective'. Since then, the absence of speech in SM was not perceived as a voluntary intent and it was recognized that mutism only presents under certain circumstances (e.g., at school) [2].

The last 20 years in the studies of SM has brought, in particular, clear evidence that anxiety is the most prominent feature of SM [3]. This is also illustrated in the most recent version of DSM [4] and in ICD-11 [5], planned for implementation, where SM is included in the anxiety disorder spectrum. Multiple examples from the literature point to the relationship between SM and social phobia (SP), however, only in some comparative studies all children diagnosed with SM met also the diagnostic criteria for SP [3]. In recent years, a theory based on developmental psychopathology that emphasizes the role of multiple factors in the etiology of SM has emerged as the leading etiological theory [3, 6]. These factors (e.g., behavioral inhibition, coexisting developmental disorders) are often fundamental for the development of anxiety, enhancing the significance of the anxiety component of SM [3]. It is unclear, though, whether SM is a primary anxiety disorder, or whether it is a form of learned avoidance behavior [3, 7]. The most effective methods of SM treatment are those utilized in other childhood anxiety disorders [8].

Aim and structure of the study

The aim of this literature review is to present the contemporary (based on the most recent literature) view of selective mutism that postulates the manifestation of this disorder as a result of mutual influence and accumulation of specific predispositions associated with the etiopathology of anxiety. For more clarity, the article is divided into thematic sections: symptoms and diagnosis, comorbidities, epidemiology, etiology and treatment. At the end of the review we present a summary and a list of factors that may increase the risk of selective mutism in early childhood.

Material and methods

The search was conducted in October 2018 using MEDLINE and Web of Science databases, with keywords related to the modern nomenclature of the condition (selective mutism). We analyzed publications related to the issues of diagnosis, etiology, epidemiology, and treatment of patients with SM. Representative studies with regard to the study population and the most important review studies were included in this review.

Symptoms and diagnosis

The term 'mutism' (Latin: *mutismus*) designates the lack of verbal contact in the absence of damage to the speech center [9]. 'Selective' in this setting denotes that the inability to speak occurs only in certain social settings (e.g., at school) or towards certain

people, while in other settings (usually: at home, towards closest family members) the patient does not experience such inability and does not show symptoms of mutism [4].

According to DSM-5, this psychopathological syndrome may be diagnosed at any developmental stage, if it is impossible to better explain the identified symptoms with a communication disorder (language and/or speech disorder). SM should also not be a symptom of an overall developmental distortion (e.g., autism) or other psychiatric disorders (e.g., schizophrenia). SM most commonly manifests during early childhood, often coinciding with the beginning of preschool or primary education. This is related to the emergence of the first big social and educational difficulties that the child faces. For many healthy children, first contact with the school environment may turn out to be stressful enough that they temporarily present characteristic features of SM. For this reason, to ascertain that a pathology is present, the diagnostic criteria require that the selective absence of speech should persist for at least one month. An absence of speech resulting from not knowing a certain language and elapsing after time required for learning this language is excluded from this definition (e.g., in immigrants) [4].

The absence of speech impairs learning ability, influencing educational achievements of the child [10]. Selectively mute children may additionally meet the diagnostic criteria for communication disorders (up to 50% in one Norwegian study, as compared to 11.5% of controls) [11]. In specific situations, the patients, instead of using speech, may try to communicate nonverbally (e.g., by nodding or gesticulating). It is not, however, characteristic for all children with SM [12]. It must be noted that the inappropriate behavior patterns often persist, even though the symptoms of mutism diminish with age [13, 14].

Comorbidities

SM often coincides with other anxiety disorders and developmental disorder or delay. As an example, in a comparative study by Kristensen [11], evaluating 54 Norwegian children diagnosed with SM (mean age: 9 years), 46.3% of patients also met diagnostic criteria for anxiety disorder and developmental disorder or delay, as compared to as little as 0.9% in the control group. According to DSM-IV criteria (diagnostic criteria for SM in DSM-5 are the same): 68% of SM children were also diagnosed with social phobia, 32% with separation anxiety, 13% with generalized anxiety disorder, and 13% were diagnosed with specific phobia [11]. In a study by Chavira et al. [15], evaluating 70 SM-diagnosed patients (mean age: 6.4 years), all children met the diagnostic criteria for SP and 40% also met the criteria for separation anxiety disorder. In both studies, the percentage of children with SM that met diagnostic criteria for anxiety disorder differed significantly from healthy controls (except for specific phobia in the study by Kristensen et al.) [11, 15]. The dominant developmental disorder in the study of this author were communication disorders, which affected up to 50% of children with SM [11]. Other studies, by Ford et al. [16] and by Steinhausen and Juzi [17], confirmed

the coincidence of these disorders in patients diagnosed with SM (11% and 38% of cases, respectively).

SM may also co-occur with generalized developmental disorders, i.e., autism spectrum disorder. In 2018, in a retrospective study, Steffenburg et al. [18] aimed to evaluate the coincidence of autism spectrum disorder in a group of 97 children diagnosed with SM. This study was conducted in a center specialized in neurodevelopmental disorders. As many as 63% of children with SM met the diagnostic criteria for an autism spectrum disorder (29% – autism, 30% – atypical autism/pervasive developmental disorder not otherwise specified, and 4% – Asperger syndrome). The next 20% of patients only presented with subclinical symptoms of autism, “which, nonetheless, had an impact on their everyday life” [18, p. 1165]. Co-occurrence of Asperger syndrome in SM children was also noted by other researchers: Kopp and Gillberg (7.4%) [19], Kristensen (7.4%) [11], Anderson and Thomsen (10%) [20]. This mild autism spectrum disorder occurs in about 0.3% of individuals in the general population – it is therefore much more frequent in children with SM [21]. In the study by Steffenburg et al. [18], SM children were divided into two groups. The first group consisted of children that met the diagnostic criteria for ASD (SM + ASD); the other comprised only children not meeting these criteria (SM – ASD). Children from the first group were characterized by a later onset of SM symptoms (on average: 5 vs. 3.6 years) and were also diagnosed after longer time. Authors suggest that children with SM may present a lower level of cognitive abilities as compared to healthy children; however, this conclusion may be biased by how these abilities are measured. To make such measurement comparable, it should be performed by persons towards which the child does not present symptoms of mutism [18].

Other psychiatric disorders, such as: obsessive-compulsive disorder (OCD) [22], dissociative disorders, depression, panic disorder [1], attention deficit hyperactivity disorder (ADHD) [6] or Fragile X syndrome [23], also occur more often among children with SM.

Epidemiology

SM is a relatively rare anxiety disorder, however, it is difficult to accurately assess its prevalence due to the inconsistent diagnostic criteria and low numbers of patients reported in the studies. Since children become selectively mute in school environment, the most reliable epidemiologic studies seem those that are based on teachers' reports [24]. Using this method and DSM-IV diagnostic criteria, children from Sweden (7–15 years) [19], the USA (5–6 years) [25] and Israel (4–6 years) [26] were examined. In these two last studies, SM prevalence was estimated at 0.71% and 0.76%, respectively, compared to only 0.18% in the study involving Swedish children. In contrast, the occurrence of SP (the most common anxiety disorder associated with SM) [3] in the US is estimated at about 9.1% in childhood and adolescence [24].

Such low prevalence of SM (Sweden) may stem from the adopted age criteria that implied the inclusion of children older than those included in the other two studies, since SM typically impacts children aged 2 to 5 years and mutism typically resolves after 8 years since diagnosis, while social disabilities typically persist in children who did not receive an appropriate therapy [13].

There are also data on family occurrence of SM [27]. It was also observed that it occurs more often (even twice) in girls [14] and in bilingual persons [28].

Etiology

Genetic and environmental factors

Frequent familial occurrence of SM [27, 29] prompted the researchers to identify genes associated with this disorder. Stein et al. [30] showed that CNTNAP2 (rs2710102) gene polymorphism is associated with greater risk of SM manifestation during childhood and with greater risk of experiencing increased social anxiety during adulthood [30]. CNTNAP2 (contactin-associated protein-like 2) is a gene encoding a protein from the neurexin family that mediates interactions between neurons and glia in the developing brain. Polymorphisms in this gene may exert pleiotropic effect, predisposing to various neurodevelopmental disorders [30]. In studies in which patients were compared with healthy controls, parents of SM children presented more shyness and a higher level of social anxiety and presented an overall tendency towards avoidance behaviors and social isolation [31–33].

The influence that the parent exerts over the child may also predispose towards SM. If a child mimics the pattern of avoidance behavior in parents, it might induce or enhance their avoidance behavior, increasing the probability of mutism manifestation or exacerbating the symptoms in a child already diagnosed with SM [6]. Moreover, in families in which anxiety disorders are present, the children are often overly attached to their parents (separation anxiety disorder frequently co-occurs with SM) [15] and subjected to excessive parental control. In a study by Edison et al., the researchers observed that parents of SM children were overprotective and more controlling as compared to parents of children from control groups [34]. It should be emphasized that in family systems theory of selective mutism, neurotic parental control prompts excessive attachment of child to the parent. This results in the development of interdependence in the parent-child relation and manifests as lack of trust towards extrafamilial environment. The child experiences anxiety, including fear of verbal communication, which manifests as mutism [35].

It is also worth mentioning that some factors may predispose to SM manifestation upon first contact with school environment. Children diagnosed with SM are often qualified to special educational programs due to communication problems and a delayed language development [16]. It can be therefore deduced that communicational deficits

result in learning difficulties and, with time, in educational deficits. Due to speech avoidance manifested at school these deficits are not disclosed and do not expose the child to negative emotions, mainly – anxiety [11]. Children with SM are perceived as generally more uneasy during contact with peers, as compared to controls, and tend to have problems with making friends or joining social groups [29, 33]. Such behaviors promote rejection and bullying towards children with SM by their peers [36]. This may further worsen the condition of the child with SM, by increasing the level of experienced anxiety.

Another seemingly important issue is the immigrant status. Epidemiologic studies in Israel showed that SM occurred in immigrant over four times more often than in native families (2.2% vs. 0.5%) [26]. The cause may be cultural differences and discrimination towards immigrants, which is a frequent source of anxiety in children in ethnic minority societies [37].

Temperamental characteristics

Children with SM present inhibited temperament, which in the case of SM means behavior style similar to behavioral inhibition [16, 32], which means that new stimuli (places, people, objects) induce excessive caution and fear, resulting in excessive avoidance of them [38]. During the preschool period this may manifest as avoiding verbal contact with strangers, closely resembling the behavior of children with SM [39]. Shyness, i.e., tendency towards feeling uneasy and tense during meetings, especially with unknown individuals, may be viewed as a social aspect of behavioral inhibition. Shyness is found in as many as 85% of children with SM [17]. In 2016, a retrospective comparative study that included children diagnosed with SM or SP, children with internalizing behaviors and healthy controls was published. Gensthaler et al. [40], based on the RIBI (*parent-rated Retrospective Infant Behavioral Inhibition*) questionnaire showed that extreme behavioral inhibition diagnosed in infancy and early childhood may preclude the manifestation of SM in older age. Besides, infants diagnosed with SM later in life presented stronger social inhibition compared to infants diagnosed with SP later in life [40], providing a proof for the assumption proposed by some authors, namely – to recognize SM as an extreme form of early childhood social anxiety [7]. Behavioral inhibition in early stages of life increases the risk of manifestation of other anxiety disorders, mainly SP, in the future [41]. Gensthaler et al. suggest to introduce screening tests that will enable early SM prevention in children with extreme behavioral inhibition [40].

Some children with SM present oppositional defiant behaviors (e.g., stubbornness, irritability, contentiousness), but since the elective concept of SM has been ruled out, the behaviors are not considered important in the etiology of this disorder [3].

Neurodevelopmental disorders

As we mentioned in the part of the article concerning comorbidities, in a considerable number of children with SM, communication disorders and/or generalized developmental disorders or developmental delay are present. Some researchers tend to consider these anomalies the source of negative emotions perceived by the children with SM, e.g., during the confrontation with properly developing children [42]. Speechlessness in selected environments may be explained as a manifestation of avoiding expected difficulties, especially in children predisposed to develop some kind of anxiety disorders. Some studies also point to the link between developmental disorders and childhood anxiety disorders [43].

Neurological deficits of auditory processing

During non-verbal communication, an interaction occurs between the processes of speaking and hearing. This enables constant monitoring and regulation of the perception of sounds and speech quality, and prevention from overstimulation of the cochlea by excessive vocalization. The aim is to ensure proper speech quality, at the same time enabling precise perception of auditory information [44, 45]. One of the efferent systems of auditory processing that takes part in the monitoring and regulation of vocalization process is the MEAR (Middle-Ear Acoustic Reflex) system. In response to the feedback for perception of own speech, muscles of the inner ear contract. This mechanism masks own voice during speaking, thus preventing distortions in the reception of auditory information. Without this mechanism, excessive vocalization could lead to overstimulation of auditory pathways. Such overstimulation would result in secondary desensitization, leading to limited reception of stimuli from surroundings during speech [44–46]. Disturbances in the MEAR system are often associated with increased auditory sensitivity that could result in avoidance of loud environments by people with such disorders [47].

According to the hypothesis of Henkin and Bar-Haim [46], disturbances in the efferent auditory processing in children with SM cause adaptation, i.e., whispering or refraining from speech, so they are able to experience undisturbed reception of auditory information. Because of the auditory hypersensitivity to own speech (e.g., disturbances in MEAR), vocalization would lead to secondary desensitization and masking external auditory stimuli [46]. Muchnik et al. [48] identified abnormalities in efferent auditory processing pathways in as many as 71% of selectively mute children, compared with 16% of controls.

The selectivity of mutism in this context may be explained as follows: while in the family environment, over time, the child learns verbal constructs characteristic for every person from the family. This way, the child with SM can better understand them, despite disturbed reception of acoustic stimuli during conversation [35]. This

would also mean that the child with SM presents the symptoms of mutism especially around these strangers whose verbal constructs most significantly differ from those known from the family environment. Under that reasoning, if the child with SM takes part in discussion with several other individuals (e.g., in school environment) they would only participate as a listener, due to the multitude of perceived language constructs. The increased difficulty stemming from the concomitant processing of diverse, unknown language constructs would prevent joining the discussion on verbal level while preserving a satisfactory quality of hearing. Muris and Ollendick [3] conclude that it is not very probable that isolated neurologic deficits of auditory processing may condition the manifestation of SM alone without other predisposing factors [3].

Selective mutism and social phobia

Based on the available literature, it can be assumed that children with SM without any concomitant anxiety disorder are in minority [3]. The most frequently co-occurring anxiety disorder is social phobia. Considering studies representative with respect to group sizes, SP co-occurred with SM in 61 to 100% of cases [11, 15, 49, 50]. The common features often identified both in children with SM and in children with SP include shyness, behavioral inhibition and separation anxiety [51]. Therefore, some researchers propose to treat SM as an early childhood variant of SP or as its extreme manifestation [25, 52].

In favor of the early-childhood SP stage hypothesis, mutism tends to disappear, on average, after 8 years since diagnosis. Meanwhile, the established behaviors associated with SM symptomatology increases the possibility of SP occurrence in the future [13, 14]. SM typically manifests in children aged 2–5 years, while SP presents later, in 8 – to 15-year-olds [53].

In favor of the hypothesis that considers SM an extreme variant of SP, in comparative studies, children with SM presented more pronounced symptoms of social phobia than children with SP but without mutism. For example, Yeganeh et al. [54] compared 23 children meeting the criteria for both SM and SP with equally numbered group of children meeting the criteria for isolated SP. Children from the first group were perceived by the observers as presenting more pronounced symptoms of social anxiety, compared to the second group. In a study by Manassis et al. [49], the level of perceived social anxiety reported by children with SM (61% of whom met also the criteria for SP) was compared with anxiety experienced by other children with anxiety disorders (including social phobia). Children that met the diagnostic criteria for SM presented highest levels of perceived social anxiety [49].

It should be emphasized that not all children meeting the criteria for SM meet also the criteria for SP [3]. For this reason, hypotheses that consider SM a variant of SP do not seem to fully capture the complexity of this disorder, only focusing on its

primarily anxiety etiology. The approach based solely on the anxiety component of this disorder may result in failure in the therapeutic process [35].

Besides similarities, some differences exist also between children with SM and children with SP or other anxiety spectrum disorders. Children with SM more often meet the criteria for communication or developmental disorders [51]. In contrast to children with SP, children with SM tend to willingly interact with peers, are willing to play with friends and share their ideas [10]. Friendships made by children with SM are often based on non-verbal communication [51].

Selective mutism as a type of avoidance behavior

In the previously mentioned study by Kristensen [11], due to frequent coexistence of developmental disorders in children with SM, the author proposed to consider the absence of speech as a form of behavior that masks some concomitant disorders. Five years later, Moldan [55] tried to depict the symptom of mutism as a strategy of regulating negative emotions (such as anxiety or embarrassment). In 2011, Scott and Beidel [42], on the basis of literature analysis, described mutism as a specific type of avoidance behavior that appears in small children with yet undeveloped strategies of regulating emotions that are somehow predisposed to develop anxiety disorders.

These authors emphasize the psychopathology of human development, where childhood is typically associated with compulsory education at school. The child, not being able to avoid school, tries to evade stressful situations, which turns out to be facilitated by the absence of speech [42]. This refers to the probable origin of anxiety and other negative emotions that the child experiences at school – where mutism most frequently manifests. Mutism can be thus compared with school refusal [7, 42], which may originate either from anxiety (e.g., frequently observed in children with SM social phobia or separation anxiety) [15], or from other causes associated with negative emotions. Other putative primary causes of avoidance behavior associated with mutism (however, they secondarily generate anxiety and other negative emotions) are developmental disorders that may be the reason behind difficulties that the child experiences in learning [3, 6, 11]. Similarly, a child with neurological dysfunctions of auditory processing may be constantly subjected to a dilemma while taking part in a discussion: to listen or to speak [46].

Viana et al. [6], in their study summarizing current knowledge about mutism, emphasized that one should not seek to identify a single determinant for the development of mutism, with respect to its evident multifactorial nature that grows over time, impacting the manifestation of SM. This is in accordance with the life span concept, where most mental disorders manifest in a critical phase of human development, on the ground of gradually developing symptoms [56]. Undoubtedly, such breakthrough event in life is the beginning of early childhood education, e.g., for a child with anxiety predisposition that has yet not appreciated their communication deficits. These

children may employ avoidance behavior in order to avoid being exposed to bullying by their peers that communicate adequately [6, 11]. This leads to the development of a functional strategy to avoid negative emotions, such as the absence of speech in selected situations [42, 57]. Avoidance behavior observed in parents may also play an important role, reinforcing avoidance behavior in children [6].

One yet unresolved issue is whether the phenomenon of selective mutism is a legitimate nosological entity or only a specific type of avoidance behavior secondary to some causes, such as in the case of school refusal [3]. In 2012, Young et al. [57] compared the evaluation of anxiety level associated with social interaction tasks. Although groups in the study were small, the results based on children's and observers' reports were consistent with the results of studies performed in bigger groups [54]. In the study by Young et al. [57], children with SM were perceived by the observers as more anxious compared to children with SP and healthy controls. This is in line with the results from the study by Yeganeh et al., where children that met the diagnostic criteria for SM presented much higher level of social anxiety, according to observers, than children with isolated SP [54]. In both studies, the level of anxiety reported by the children with SM was not significantly different from the level reported by children that did not meet the criteria for SM [54, 57].

The originality of the study by Young et al. [57] based on the measurement of the psychophysiological arousal (e.g., heart rate, electrodermal activity) in the children being examined. While performing social interaction tasks, children with SM showed much smaller degree of arousal as compared to other groups. Based on these results, Young et al. are inclined to recognize the phenomenon of mutism as a type of learned avoidance behavior that allows to lower the level of anxiety experienced in certain situations that require social interaction [57].

In light of the most recent studies from the field of biological psychiatry, avoidance behavior is considered an unconscious defense mechanism [58]. This is consistent with rejection of the concept of electivity of mutism, where the absence of speech was seen as a type of oppositional defiant disorder or a desire to willingly manipulate others to achieve some kind of advantage [1, 2, 4]. Avoidance is a very effective method of reducing symptoms associated with anxiety. This translates to a drive to repeat avoidance behaviors and enhances them in the process of causal learning (the second factor contributing to development of anxiety disorder in Mowrer's theory) [42, 58]. More broadly, when the child learns to abuse the avoidance behaviors, it limits their developmental possibilities. Avoidance predisposes to secondary social and language deficits, impacting broadly understood social functioning, including education and career [10].

It is unclear whether the absence of speech in certain situations is a disease *per se* or a type of avoidance behavior. Factors predisposing to SM often predispose also to SP. Children with SM more often than children with SP meet the diagnostic criteria for developmental disorders. Most selectively mute children meet the diagnostic criteria for SP.

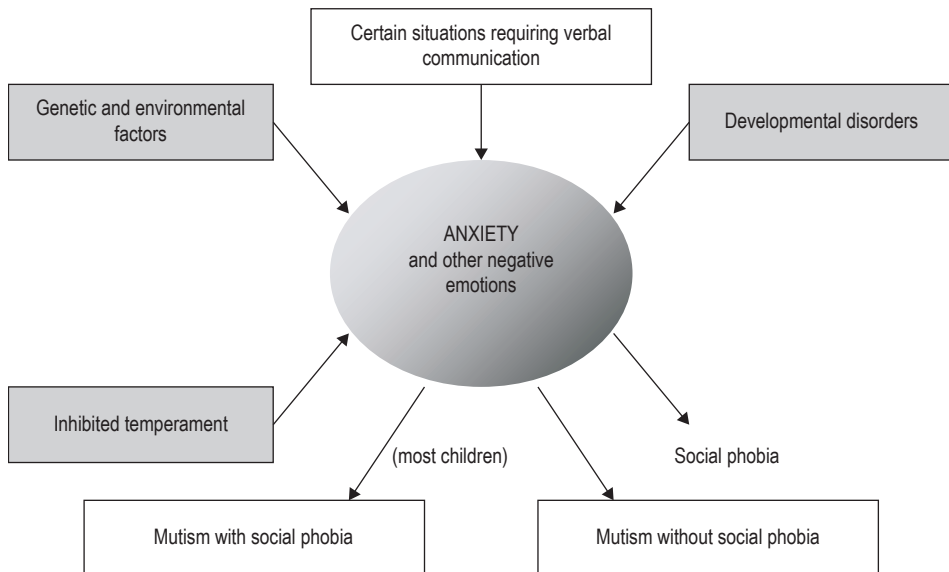


Figure 1. Model explaining the etiology of SM, including developmental psychopathology

Treatment

Recent years have brought evidence that confirmed anxiety etiology of SM, which is reflected in the most recent classifications that incorporate SM into anxiety disorders [4, 5]. For this reason, SM treatment standards resemble the forms of treatment most effective and most frequently used in other childhood anxiety disorders: cognitive behavioral therapy (CBT) and pharmacotherapy using selective serotonin reuptake inhibitors (SSRI) or a combination of both [8]. These therapies are aimed to enable speaking in situations in which the child presents symptoms of mutism [59].

The aim of CBT is to modify behaviors and thinking patterns [53]. Due to the fact that the contact between the therapist and a selectively mute child may be hindered, it is recommended to adequately adapt psychotherapeutic methods in order to ensure that the contact with the patient is the best that is possible [8]. Bergman et al. [60] propose an integrated behavioral therapy (IBT) that engages not only the child in the therapeutic process, but also parents and teachers. Building relation between the therapist and child and the use of behavioral rewards (positive reinforcement) is beneficial on the child engagement in the therapeutic process. It is also helpful to plan the therapeutic sessions so that the first sessions involve both a person towards which the child does not present mutism and a person around which the child does not speak (e.g., parent + therapist). Next sessions are scheduled according to the primarily established plan, i.e., a gradually increasing exposition to situations requiring verbal communication [60].

In 2018, a systematic review synthesizing the results from 15 studies summarized the efficacy of the aforementioned therapies in SM treatment. Symptoms reduction (evaluated based on standardized scales) was most frequently noted in children treated with CBT – 88.3% (53/60). Pharmacotherapy alone was beneficial in 82.1% (55/67) of children and the combined therapy was successful in 85.7% of children; however, in this instance, the group was small (6/7) [8].

Most researchers agree that CBT is becoming the treatment of choice in children with SM [8]. Pharmacotherapy based on SSRI (most frequently fluoxetine) [61] and combined therapy are reserved for more serious cases of SM, e.g., when psychotherapy alone is insufficient [8, 61]. This is associated with possible adverse effects of pharmacotherapy, although SSRI seem to be well tolerated by children with SM [8, 62]. SSRI reduce anxiety symptoms associated with speaking, increasing also the efficacy of CBT [8]. Monoamine oxidase inhibitors (MAOI) are also used in SM, but they are not recommended due to adverse effects and should be used as drugs of last resort [3, 8].

The authors of the aforementioned review conclude that the described therapies (especially CBT) may also reduce symptoms of disorders coexisting with SM (such as SP, separation anxiety, generalized anxiety disorder, obsessive-compulsive disorder, ADHD), but further studies are needed to confirm this observation in a bigger group of children [8]. The treatment (psychotherapy and/or pharmacotherapy) application in younger age is seemingly associated with better clinical outcome – thus it is assumed that early diagnosis is very important for good prognosis [8, 63].

Many children with SM also meet the criteria for other coexisting disorders [3] that may be the source of anxiety and other negative emotions [3, 6, 35]. It can be thus assumed that knowing the source of anxiety and its proper treatment may be crucial for a sustained therapeutic success. For example, Henkin and Bar-Haim [46] propose specialized trainings for children with auditory processing deficits to enhance the synchronization of simultaneous processing of auditory information during speaking.

The authors of studies about SM recommend a multimodal approach to this disorder, that considers the inclusion of psychiatrists, psychologists, audiologists and other specialists in the therapeutic process. The diagnostic process should particularly focus on the symptoms of anxiety disorders, neurodevelopmental disorders (e.g., ASD, speech and language disorders), deficits of auditory processing and other mental disorders [3, 6].

Recapitulation

Selective mutism is closely associated with etiopathology of anxiety. Coexisting developmental disorders (e.g., speech and/or language communication disorders, ASD), a specific temperament (behavioral inhibition), environmental factors (e.g., pattern of

avoidance behavior in the family) are described in the literature as factors predisposing to the development of anxiety disorders. According to the developmental psychopathology, genetic factors and interactions between the aforementioned predispositions may, with time, lead to SM manifestation (the critical period). The multifactorial etiology necessitates a multimodal approach in the diagnostic-therapeutic process that should be undertaken as quick as possible to prevent secondary delay in the cognitive and social development of the child. It can be thus assumed that the prognosis and course of SM depend on the accompanying disorders.

The most effective treatment, similar as in other childhood anxiety disorders, turns out to be cognitive behavioral psychotherapy, recommended as a method of choice. Therapy focused solely on the anxiety component, if other disorders that generate anxiety exist, may be only symptomatic. Further studies should address whether SM is an anxiety disorder or a type of avoidance behavior resulting from experiencing negative emotions, caused by difficulties in communication and education (e.g., at school), because of coexisting disturbances.

Factors that may increase the risk of selective mutism in early childhood:

- female gender;
- CNTNAP2 gene polymorphism;
- parents with tendencies towards avoidance behavior;
- overprotection and excessive parental control;
- educational difficulties and/or difficulties in contact with peers due to: communication disorders and/or developmental delay and/or auditory processing impairment;
- immigrant status;
- shyness;
- behavioral inhibition (especially strongly expressed).

References

1. Sharp WG, Sherman C, Gross AM. *Selective mutism and anxiety: A review of the current conceptualisation of the disorder*. J. Anxiety Disord. 2007; 21(4): 568–579.
2. American Psychological Association (APA). *Diagnostic and Statistical Manual of Mental Disorders*, 4th ed. (DSM-IV). 1994.
3. Muris P, Ollendick TH. *Children who are anxious in silence: A review on selective mutism, the New Anxiety Disorder in DSM-5*. Clin. Child Fam. Psychol. Rev. 2015; 18(2): 151–169.
4. American Psychiatric Association (APA). *Diagnostic and statistical manual of mental disorders: DSM-5*. 2013.
5. Kogan CS, Stein DJ, Maj M, First MB, Emmelkamp PMG, Reed GM. *The classification of anxiety and fear-related disorders in the ICD-11*. Depress. Anxiety. 2016; 33(12): 1141–1154.

6. Viana AG, Beidel DC, Rabian B. *Selective mutism: A review and integration of the last 15 years*. Clin. Psychol. Rev. 2009; 29(1): 57–67.
7. Bogels SM, Alden L, Beidel DC, Clark LA, Pine DS, Stein MB et al. *Social anxiety disorder: Questions and answers for the DSM-V*. Depress. Anxiety. 2010; 27(2): 168–189.
8. Østergaard KR. *Treatment of selective mutism based on cognitive behavioural therapy, psychopharmacology and combination therapy – A systematic review*. Nord. J. Psychiatry. 2018; 72(4): 240–250.
9. Bilikiewicz A., editor. *Psychiatria: podręcznik dla studentów medycyny*, 3rd ed. Warsaw: PZWL Medical Publishing; 2007. P. 706.
10. Smith-Schrandt HL, Ellington E. *Unable to speak: Selective mutism in youth*. J. Psychosoc. Nurs. Ment. Health Serv. 2018; 56(2): 14–18.
11. Kristensen H. *Selective Mutism and Comorbidity With Developmental Disorder/Delay, Anxiety Disorder, and Elimination Disorder*. J. Am. Acad. Child Adolesc. Psychiatry. 2000; 39(2): 249–256.
12. Oerbeck B, Manassis K, Overgaard KR, Kristensen H. *Selective mutism*. In: Rey JM, editor. *IACAPAP e-textbook of child and adolescent mental health*. Geneva: International Association for Child and Adolescent Psychiatry and Allied Professions, Chapter F.5. 2016. P. 1–23.
13. Remschmidt H, Poller M, Herpertz-Dahlmann B, Hennighausen K, Gutenbrunner C. *A follow-up study of 45 patients with elective mutism*. Eur. Arch. Psychiatry Clin. Neurosci. 2001; 251(6): 284–296.
14. Steinhausen H-C, Wachter M, Laimböck K, Metzke CW. *A long-term outcome study of selective mutism in childhood*. J. Child Psychol. Psychiatry. 2006; 47(7): 751–756.
15. Chavira DA, Shipon-Blum E, Hitchcock C, Cohan S, Stein MB. *Selective mutism and social anxiety disorder: All in the family?* J. Am. Acad. Child Adolesc. Psychiatry. 2007; 46(11): 1464–1472.
16. Ford MA, Sladeczek IE, Carlson J, Kratochwill TR. *Selective mutism: Phenomenological characteristics*. Sch. Psychol. Q. 1998; 13(3): 192–227.
17. Steinhausen HC, Juzi C. *Elective mutism: An analysis of 100 cases*. J. Am. Acad. Child Adolesc. Psychiatry. 1996; 35(5): 606–614.
18. SteRenburg H, SteRenburg S, Gillberg C, Billstedt E. *Children with autism spectrum disorders and selective mutism*. Neuropsychiatr. Dis. Treat. 2018; 14: 1163–1169.
19. Kopp S, Gillberg C. *Selective mutism: A population-based study: A research note*. J. Child Psychol. Psychiatry. 1997; 38(2): 257–262.
20. Andersson CB, Thomsen PH. *Electively mute children: An analysis of 37 Danish cases*. Nord. J. Psychiatry. 1998; 52(3): 231–238.
21. Ehlers S, Gillberg C. *The epidemiology of Asperger syndrome*. J. Child Psychol. Psychiatry. 1993; 34(8): 1327–1350.
22. Wong P. *Selective mutism: A review of etiology, comorbidities and treatment*. Psychiatry (Edgemont). 2010; 7(3): 23–31.
23. Sharkey L, McNicholas F. *More than 100 years of silence, elective mutism: A review of the literature*. Eur. Child Adolesc. Psychiatry. 2008; 17(5): 255–263.
24. Merikangas KR, He JP, Burstein M, Swanson SA, Avenevoli S, Lihong C et al. *Lifetime prevalence of mental disorders in U.S. adolescents: Results from the National Comorbidity Survey*

- Replication–Adolescent Supplement (NCS-A)*. J. Am. Acad. Child Adolesc. Psychiatry. 2010; 49(10): 980–989.
25. Bergman RL, Piacentini J, McCracken JT. *Prevalence and description of selective mutism in a school-based sample*. J. Am. Acad. Child Adolesc. Psychiatry. 2002; 41(8): 938–946.
 26. Elizur Y, Perednik R. *Prevalence and description of selective mutism in immigrant and native families: A controlled study*. J. Am. Acad. Child Adolesc. Psychiatry. 2003; 42(12): 1451–1459.
 27. Steinhausen HC, Adamek R. *The family history of children with elective mutism: A research report*. Eur. Child Adolesc. Psychiatry. 1997; 6(2): 107–111.
 28. Cohan SL, Price JM, Stein MB. *Suffering in silence: Why a developmental psychopathology perspective on selective mutism is needed*. J. Dev. Behav. Pediatrics. 2006; 27(4): 341–355.
 29. Cunningham CE, McHolm AE, Boyle MH. *Social phobia, anxiety, oppositional behavior, social skills, and selfconcept in children with specific selective mutism, generalized selective mutism, and community controls*. Eur. Child Adolesc. Psychiatry. 2006; 15(5): 245–255.
 30. Stein MB, Yang B-Z, Chavira DA, Hitchcock CA, Sung SC, Shipon-Blum E et al. *A common genetic variant in the neurexin superfamily member CNTNAP2 is associated with increased risk for selective mutism and social anxiety-related traits*. Biol. Psychiatry. 2011; 69(9): 825–831.
 31. Kristensen H, Torgersen S. *MCMI-II personality traits and symptom traits in parents of children with selective mutism: A case-control study*. J. Abnorm. Psychol. 2001; 110(4): 648–652.
 32. Kristensen H, Torgersen S. *A case-control study of EAS child and parental temperaments in selectively mute children with and without a comorbid communication disorder*. Nord. J. Psychiatry. 2002; 56(5): 347–353.
 33. Cunningham CE, McHolm A, Boyle MH, Patel S. *Behavioral and emotional adjustment, family functioning, academic performance, and social relationships in children with selective mutism*. J. Child Psychol. Psychiatry. 2004; 45(8): 1363–1372.
 34. Edison SC, Evans MA, Mc Holm AE, Cunningham CE, Nowakowski ME, Boyle M et al. *An Investigation of Control Among Parents of Selectively Mute, Anxious, and Non-Anxious Children*. Child. Psychiatry Hum. Dev. 2011; 42(3), 270-290.
 35. Holka-Pokorska J, Pirog-Balcerzak A, Jarema M. *The controversy around the diagnosis of selective mutism – A critical analysis of three cases in the light of modern research and diagnostic criteria*. Psychiatr. Pol. 2018; 52(2): 323–343.
 36. Kumpulainen K, Räsänen E, Raaska H, Somppi V. *Selective mutism among second-graders in elementary school*. Eur. Child Adolesc. Psychiatry. 1998; 7(1): 24–29.
 37. Beidas RS, Suarez L, Simpson D, Read K, Wei C, Connolly S et al. *Contextual factors and anxiety in minority and European American youth presenting for treatment across two urban university clinics*. J. Anxiety Disord. 2012; 26(4): 544–554.
 38. Kagan J, Snidman N, Arcus D, Steven Reznick J. *Galen's prophecy: Temperament in human nature*. New York, NY: Basic Books; 2018.
 39. Coll CG, Kagan J, Reznick JS. *Behavioral inhibition in young children*. Child Dev. 1984; 55(3): 1005–1019.
 40. Gensthaler A, Khalaf S, Ligges M, Kaess M, Freitag CM, Schwenck C. *Selective mutism and temperament: The silence and behavioral inhibition to the unfamiliar*. Eur. Child Adolesc. Psychiatry. 2016; 25(10): 1113–1120.

41. Biederman J, Hirshfeld-Becker DR, Rosenbaum JF, Hérot C, Friedman D, Snidman N et al. *Further evidence of association between behavioural inhibition and social anxiety in children.* Am. J. Psychiatry. 2001; 158(10): 1673–1679.
42. Scott S, Beidel DC. *Selective mutism: An update and suggestions for future research.* Curr. Psychiatry Rep. 2011; 13(4): 251–257.
43. White SW, Oswald D, Ollendick T, Scahill L. *Anxiety in children and adolescents with autism spectrum disorders.* Clin. Psychol. Rev. 2009; 29(3): 216–229.
44. Hoy R. *Tuning in by tuning off.* Nature. 2002; 418: 831–833.
45. Curio G, Neuloh G, Numminen J, Jousmaki V, Hari R. *Speaking modifies voice-evoked activity in the human auditory cortex.* Hum. Brain Mapp. 2000; 9(4): 183–191.
46. Henkin Y, Bar-Haim Y. *An auditory-neuroscience perspective on the development of selective mutism.* Dev. Cogn. Neurosci. 2015; 12: 86–93.
47. Bar-Haim Y. *Introversion and individual differences in acoustic reflex function.* Int. J. Psychophysiol. 2002; 46(1): 1–11.
48. Muchnik C, Ari-Even Roth D, Hildesheimer M, Arie M, Bar-Haim Y, Henkin Y. *Abnormalities in auditory efferent activities in children with selective mutism.* Audiol. Neurootol. 2013; 18(6): 353–361.
49. Manassis K, Tannock R, Garland EJ, Minde K, McInnes A, Clark S. *The sounds of silence: Language, cognition, and anxiety in selective mutism.* J. Am. Acad. Child Adolesc. Psychiatry. 2007; 46(9): 1187–1195.
50. Bergman RL, Keller ML, Piacentini J, Bergman AJ. *The development and psychometric properties of the selective mutism questionnaire.* J. Clin. Child Adolesc. Psychol. 2008; 37(2): 456–464.
51. Diliberto RA, Kearney CA. *Anxiety and oppositional behavior profiles among youth with selective mutism.* J. Commun. Disord. 2016; 59: 16–23.
52. Black B, Uhde TW. *Psychiatric characteristics of children with selective mutism: A pilot study.* J. Am. Acad. Child Adolesc. Psychiatry. 1995; 34(7): 847–856.
53. Hua A, Major N. *Selective mutism.* Curr. Opin. Pediatr. 2016; 28(1): 114–120.
54. Yeganeh R, Beidel DC, Turner SM, Pina AA, Silverman WK. *Clinical distinctions between selective mutism and social phobia: An investigation of childhood psychopathology.* J. Am. Acad. Child Adolesc. Psychiatry. 2003; 42(9): 1069–1075.
55. Moldan MB. *Selective mutism and self-regulation.* Clin. Soc. Work J. 2005; 33(3): 291–307.
56. Woo SM, Keatinge C. *Diagnosis and treatment of mental disorders across the lifespan.* Hoboken, NY: Wiley and Sons, Inc.; 2008.
57. Young BJ, Bunnell BE, Beidel DC. *Evaluation of children with selective mutism and social phobia: A comparison of psychological and psychophysiological arousal.* Behav. Modif. 2012; 36(4): 525–544.
58. LeDoux JE, Moscarello J, Sears R, Campese V. *The birth, death and resurrection of avoidance: A reconceptualization of a troubled paradigm.* Mol. Psychiatry. 2016; 22(1): 24–36.
59. Pionek Stone B, Kratochwill T, Sladeczek I, Serlin RC. *Treatment of selective mutism: A best-evidence synthesis.* Sch. Psychol. Q. 2002; 17(2): 168–190.
60. Bergman RL, Gonzalez A, Piacentini J, Keller ML. *Integrated behavior therapy for selective mutism: A randomized controlled pilot study.* Behav. Res. Ther. 2013; 51(10): 680–689.

61. Carlson J, Kratochwill T, Johnston H. *Prevalence and treatment of selective mutism in clinical practice: A survey of child and adolescent psychiatrists*. J. Child Adolesc. Psychopharmacol. 1994; 4(4): 281–291.
62. Dummit ES, Klein RG, Asche B, Martin J, Tancer NK. *Fluoxetine treatment of children with selective mutism: An open trial*. J. Am. Acad. Child Adolesc. Psychiatry. 1996; 35(5): 615–621.
63. Oerbeck B, Stein MB, Pripp AH, Kristensen H. *Selective mutism: Follow-up study 1 year after end of treatment*. Eur. Child Adolesc. Psychiatry. 2015; 24(7): 757–766.

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