

The controversy around the diagnosis of selective mutism – a critical analysis of three cases in the light of modern research and diagnostic criteria

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Summary

The position of selective mutism disorder – SM – has been modified in the last edition of the classification of mental disorders DSM-5. It was removed from “Disorders of childhood and adolescence” and placed in “Anxiety disorders”. This caused two important changes in the interpretation of the symptoms of selective mutism. It highlighted anxious etiology of the disorder and also open the possibility to diagnose selective mutism in adults as a special category of anxiety disorders. The aim of this study was to present three different cases concerning the diagnostic difficulties of selective mutism (the child, the teenager and the persons who became adult during our observation) regarding current views on SM. In this study we presented the current view on the etiology, course and available therapies for selective mutism. Owing to updating the clinical knowledge about SM and describing three cases, we highlighted the controversies around the diagnosis and treatment of this disorder. Selective mutism might be a preliminary diagnosis, often leading to the diagnosis of other disorders of diverse etiology and course. Among the psychiatric aspects of the disorder, the ‘anxiety component’ of SM is crucial. In individuals with selective mutism, developmental disorders, social cognition and neurocognition deficits or dysfunctions of auditory processing often coexist. The severity and the type of comorbidities may determine the future course of the illness and the final effects of the therapy.

Key words: selective mutism, social phobia, ASD – Autism Spectrum Disorder

Introduction

Selective mutism is a psychopathological unit whose etiology and nosological position remains unclear for many years. A comprehensive and uniform theory about the diagnosis and treatment of this disorder does not exist. In the latest edition of the

classification of mental disorders DSM-5, changes concerning the classification of that syndromological unit were made. Selective mutism was moved from “disorders of childhood and adolescence” to “anxiety disorder” category in the DSM-5 [1]. This highlighted and “sanctioned” the concept of primary anxiety etiology of this disorder [1].

Diagnostic criteria for SM were not changed [1]. However, according to the latest edition of the DSM-5, selective mutism can be diagnosed in children and adolescents as well as adults, provided that it is a type of anxiety disorders, not a symptom of psychosis, developmental, neurological or speech disorders. Over the past few years a lot of controversy and confusion raised around SM. Some researchers do not qualify SM as mental or emotional disorders, but consider it to be an effect of speech deficits, or avoiding type of behavioral disorder [2].

Aim and method

The presented paper had two objectives. One was to discuss the current views on the genesis and treatment of selective mutism in relation to the new approach of SM classification. Review of the literature was made via MEDLINE database and based on such key words as “selective mutism”, “selective mutism diagnosis”, “selective mutism treatment”. The papers representative for the analyzed research groups and essential for the diagnosis and treatment of patients with selective mutism were qualified for further analysis. The most important reviews on the analyzed topic published so far were also analyzed.

Another, no less important aim of the study, was a description of controversy around the diagnosis of selective mutism. It was illustrated by three very different cases of patients with SM in which at certain stages of observation selective mutism was diagnosed. Describing these cases we paid attention to the characteristics of clinical symptoms, comorbid mental disorders and the course of the disorder. In two of three presented patients, the diagnosis of selective mutism has been verified.

The nosological position of selective mutism

The etiology and pathogenesis of mental disorders usually raise controversy among scientists and clinicians. Therefore, in order to precisely describe the patients' psychopathology, the concept of comorbidity of psychiatric disorders was created. The extension of that concept is the concept of lifetime comorbidity. According to Wittchen [4] and Jacobi et al. [3], lifetime comorbidity allows to look at the psychopathology of a patient from a developmental perspective. The two most important diagnostic systems used in the psychiatric diagnosis are non-nosological systems.

The classification and diagnostic position of selective mutism evolved from marginally treated “specific behavioral disorders” to the contemporary approaches recognizing SM as an extreme manifestation of social phobia. In 1934, Tramer described the case of an eight-year old child who refused to speak in certain situations and introduced the term elective mutism (EM). This was to emphasize the lack of verbal contact as a conscious choice of people affected by this disorder [5]. Until the 1990s, there were many ambiguous theories about selective mutism in the literature. They emphasized insubordination and stubbornness of people with selective mutism. Also, the manipulative and controlling behavior of overprotective parents (especially mothers) were linked with SM [6].

The term elective mutism and early American diagnostic criteria for the disorder highlighted the importance of conscious will of patients to withdraw from verbal contact. The term was used to define “refusal to talk” to most people and in most social situations, dependent on the patient’s will. The term elective mutism was still present in the DSM-III-R, and have also appeared in Polish psychiatric literature [7]. In the 1980s and 1990s, the withdrawal from verbal contact was interpreted as oppositional defiant features, not as social anxiety manifestation.

In the DSM-IV, DSM-IV-R, and DSM-5 the term elective mutism was replaced by selective mutism. It was done to highlight the phenomenon of mutism, which consists in speech selectivity that applies only to some (selected), not all social environments, and cannot be combined with conscious manipulation of the environment by refusing to speak.

In the ICD-10, selective mutism is listed in “disorders of social functioning with onset specific to childhood and adolescence” [8]. According to the ICD-10, selective mutism is diagnosed when the following conditions are present:

- a) normal or close to normal level of intelligibility;
- b) level of speech competence sufficient for social communication;
- c) clear evidence that the child can speak and speaks normally or almost normally in some situations.

However, in a significant minority of children with selective mutism, delays in speech development or difficulty in articulation are reported.

Thus, according to the ICD-10, selective mutism is still regarded as a health problem limited to childhood and not a form of anxiety disorders that may appear in different periods of life.

Modern social and psychological theories concerning the etiopathogenesis of selective mutism

Most of the scientific findings on selective mutism is based on a very small research groups or case reports only. For this reason epidemiological studies on that diagnostic category may not reflect the real prevalence of this disorder. Epidemiological studies from Western Europe, the United States and Israel show that SM is a relatively rare disorder and is observed in the population with an incidence of 0.03–0.76% [2, 9, 10]. It is observed twice often in girls than in boys [11, 12]. Some authors try to find the sociocultural background of selective mutism, noting that social restraint seems to be more accepted and more often “rewarded” for girls than for boys [13].

As already mentioned, homogenous and applicable theory of selective mutism etiopathogenesis have not been created so far. Among the psychological theories of SM behavioral, psychodynamic and systemic theories are worth mentioning [14].

Behavioral psychologists, using behavioral theory and negative reinforcement in learning process, assume that in new social situations the sympathetic nervous system takes inhibiting control on the ability to speak and on the whole behavior of children with SM. Therefore, children with this disorder in threatening situations (new social situations) behave as if they were “immobile” or “frozen”. On the linguistic level it manifests itself as silence [9, 14, 15].

Psychodynamic theory emphasizes that unresolved oedipal conflict and fixation at the oral or anal stage of psychosexual development are important for the SM etiology [16]. By transposing anger towards the same sex parent, the regression to non-verbal stage of development appears as a defense mechanism in situations which increase fear and exacerbate the internal conflict [16]. Silence becomes a defense mechanism to “punish” parents.

In the systemic theory, the importance of neurotic parental control over the children with selective mutism associated with dependence and ambivalence is highlighted. As a result, children are excessively and interdependently attached to their parents. It causes lack of confidence in the outside world, fear of strangers, fear of assimilating difficulties, and fear of verbal communication. This leads to clinical manifestations of the disorder – silence [9, 17].

Discussing the psychological theories of SM, it is worth to cite the so-called new integrated developmental theory. It stresses the importance of earlier biological and temperamental anxiety predisposition in children with speech development disorders. According to the integrated developmental theory, children with SM remain unaware of their language deficits before starting pre-school education (or school education when a child did not attend kindergarten). However, in new, “demanding” social interaction with their peers with normal development of language functions, children with SM are excluded from that group. Beginning of education can be the

first time when the SM children confront with their disabilities. In children with anxiety response patterns, it may trigger a pattern of avoiding confrontation in the kindergarten/school environment and activate mutistic behavior [2]. The integrated developmental theory also emphasizes the importance of avoiding response pattern in families of patients with SM.

In the 1990s, it was suggested that SM is a kind of strongly expressed social phobia linked with speech and behavioral disorders [18]. This theory is based on the observations that SM is most common in children from families with social phobia history [18]. The symptoms of selective mutism in children may correspond to avoidance of public speaking by adult people with social phobia. In another study, children with SM had higher level of anxiety, more obsessive features and higher percentage of somatic complaints than children in the control group [19].

Oponents of the theory that SM is a part of anxiety disorder spectrum emphasize that the most common period of social anxiety onset is not the same as the onset of SM [17]. The symptoms of SM usually show up in children aged 3–6 years, while the symptoms of social phobia – in adolescents aged 11–13. In a study by Melfsen et al. [17], the severity of anxiety in children with SM symptoms was assessed. The level of social anxiety in studied children was lower as compared with children with a diagnosis of social phobia. Yaganeh et al. [20] confirmed Melfsen's conclusions. The group of children diagnosed with SM had lower level of social anxiety than the subgroup of children with selective mutism symptoms and concomitant social phobia. Moreover, it was observed that children with SM assessed the severity of concomitant social anxiety as lower compared with children with symptoms of social phobia but without the coexisting SM symptoms [20, 21].

The theory of SM being the form of social anxiety is one of the most important theories on this issue in recent years. The study of last two decades caused that SM is not seen as a variation of oppositional defiant behavior any more. This was reflected in the last edition of the DSM-5 by placing SM in anxiety disorder subsection [1]. The following disorders are also listed in that section: separation anxiety, specific phobias, social phobia, panic disorder, agoraphobia, generalized anxiety disorder, unspecified anxiety disorder, substance/medication-induced anxiety disorder, anxiety disorder due to another medical condition.

Descriptive and comparative research on persons with SM have shown that the majority of them also suffered from anxiety disorders [11, 12, 22]. Depending on the study it was 74.1% ($N = 50$) [12], 66% ($N = 100$) [11] or even 100% [22] of examined patients.

Table 1. The prevalence of the anxiety disorders in patients with selective mutism according to the selected descriptive and comparative research

Author	Study construction/ participants	Conclusions
Steinhausen and Juzi 1996 [11]	Descriptive study, N = 100	onset of the disorder – preschool age, more frequent in girls, often preceded by migration or speech fluency or development disorders (38% of responders) mutism most frequently seen at school or to strangers often shyness (85%), internalization disorders and anxiety disorders (66%)
Dummit et al. 1997 [22]	Descriptive study, N = 50	concomitant social phobia in 100% of children with SM other anxiety disorders in 48% of patients
Yegene et al. 2003 [20]	Comparative study, N = 23 children with SM and social phobia and N = 23 children with social phobia	no differences in the level of trait-anxiety and general anxiety level in both groups higher scores in oppositional defiant scales in children with SM as compared with children with social phobia (without SM symptoms)

Audiological and neurobiological theories of selective mutism

The importance of language development deficits, speech fluency and syntax disorders in children with SM are emphasized more often [23, 24]. Studies assessing the development of language function in SM children indicate that despite fluent speech at home, such children, compared to children with social phobia and healthy children, show a number of language deficits in standardized tests evaluating development of speech [23, 24]. Dysfunctions in development of language system in children with SM were similar when observed both at home and in other environment [24]. In a study by Manassis et al. [23], it was proved that older age and better grammatical and syntactical fluency correlate with less severe symptoms of mutism. The results of this study suggest a strong correlation between the severity of language deficits and advancement of applied avoidance patterns manifested by the lack of verbal contact.

Recent studies also indicate efferent auditory processing disorders in children with selective mutism [25]. The processes of hearing and speaking depend on each other. This relationship enables constant monitoring of voice and speech quality, sounds perception and the prevention of overstimulation with excessive vocalization [25, 26].

In humans, two distinct efferent mechanism are involved in monitoring and regulating vocalization:

- 1) MEAR – middle-ear acoustic reflex and
- 2) MOCB – medial olivocochlear bundle reflex.

The MEAR participates in neutralizing loud low-frequency sounds to prevent overstimulation of auditory nerve pathways. When the MEAR is activated by self-vocalization, it is assumed to produce an anti-masking effect by attenuating potential overloading of the cochlea. This allows to maintain a fairly constant level of sensitivity that prevents interference by the speaker's own voice [25, 27]. According to a study by Bar-Haim, the MEAR abnormalities are observed in the majority of introverted people and correlate with their increased auditory sensitivity and preference for more quiet environments [25, 28].

The functional role of the MOCB during vocalization is not fully understood. Probably it protects the ear from noise-induced cochlear damage caused by exposure to moderate sounds intensities (similar to the sounds in various natural environments) [25, 29, 30]. Such a hypothesis is supported by the fact that the MOCB is activated in anticipation of vocalization. Moreover, the MOCB probably plays an anti-masking role for background noise during signal detection/perception [25, 31].

Bar-Haim and Henkin [25] postulate that the deficits in the MOCB and MEAR play an important role in the pathogenesis of selective mutism. In a study in which they compared a group of children with SM and a group of healthy controls, they showed a significant suppression of P50 evoked potential of a subsequent of two auditory stimuli in children with SM [32]. Dysfunctions of the two systems may result in a lack of synchronization in processing the auditory information obtained during simultaneous loud speaking. According to Bar-Haim and Henkin, in children with SM the MEAR and MOCB dysfunction during vocalization can manifest in excessive masking of auditory signals. This causes desensitization of auditory pathways towards processed and analyzed sounds.

According to the concept of Bar-Haim and Henkin, children with SM have difficulty in simultaneous coping with incoming sounds and loud self-vocalization. According to this hypothesis, children with SM adapt to efferent dysfunction of the auditory system through lowering the voice, whispering or total refusal to speak. This is mainly seen in situations that require highly effective processing of auditory stimuli [25, 33]. For this reason it is difficult to point to a consistent pattern of auditory environments and social situations in which they speak or remain silent. The authors of this model emphasize, however, that audiological and neurological dysfunction underlying SM may challenge some children with the dilemma of whether to speak, and thus jeopardize perception of incoming sounds, or listen without active vocal participation [25].

On the basis of the presented model, it can be assumed that home, where dialogues are taken in one-to-one scenarios, is a safe and secure environment for children with SM. In addition, listening to verbal messages selected over the years and unconsciously "adapted" by the environment to the auditory needs of children with SM may allow for optimal compensation of their auditory deficits. This compensation is strengthened

by knowing verbal constructs used by family and by intimacy of such dialogues, when it is unnecessary to synchronize the sounds coming from different sources (e.g., from several people, during a conversation in the classroom, or from strangers, that use other/new language constructs than the closest family).

Description of three clinical cases

In this part of the paper, three cases of patients who showed symptoms of selective mutism at certain stages of clinical observation were described. All presented patients at certain stages of development met the criteria for selective mutism according to the ICD-10. However, in the course of further observation, the diagnosis was verified towards other neurodevelopmental disorders or the diagnosis could not be conclusively determined. The cases were selected in order to highlight the heterogeneity of the disorders that could be hidden under the diagnosis of SM.

Case 1

Patient aged 17, never mentally treated before. He was consulted a few years earlier by a psychologist because of concentration and memory problems observed by parents. Significant cognitive impairment was excluded, but he was offered a relaxation techniques because of anxiety symptoms. Systematic psychotherapy was recommended, but the boy refused to attend and parents could not encourage him.

Pregnancy and delivery period without problems, all developmental milestones on time, no speech developmental disorders were observed. He has no problems with somatic health, no head injuries or seizures were observed.

In family history it was revealed that in first-degree relatives occurred mental disorders with the need of psychiatric intervention. His sister (24 years old) was always very shy in social situations but verbally aggressive at home. In adolescence she self-harmed herself. Because of social phobia and anxiety disorders she was pharmacologically treated with citalopram and attended individual and group psychotherapy a couple of times. The patient's father was also treated with citalopram because of depression.

The first psychiatric consultation took place a few months before he turned 18 years because he refused to return to school after the summer break (second class of comprehensive secondary school). As he was a minor, the visit was in his mother presence. School problems began in the second semester of the first class of comprehensive secondary school, when his grades significantly worsened and he was barely promoted to next class.

The patient always had a very small group of peers and limited relationships. In the last years his social contacts were limited to parents, sister, closest cousin, and

a primary school friend. Few peer contacts have never been initiated by the patient, but always by other people. In the first year of secondary school he did not make peer relations with any of his colleagues.

At school he answered in monosyllables, never developing the subjects. In the last year, prior to described psychiatric treatment, he stopped responding to the questions during lessons. Due to the deepening avoidance of verbal contact with teachers and colleagues, the patient refused to come back to school after the summer break. In the last weeks of vacation his mood worsened. He did not want his best friend to visit him. He spoke only when it was absolutely necessary. The rumination about suicide, without the tendency, started. Verbal contact with their parents remained undisturbed even during the period when the patient completely ceased to communicate in the school environment.

During a psychiatrist visit, which took place a few days after the school year started, he was sitting with his head lowered, with no eye contact, his hair on his face. He did not initiate the conversation, answered reluctantly, in monosyllables, never in sentences. He did not want to describe his mood or mental state. He confirmed slightly depressed mood, anxiety and transient suicidal thoughts, without the tendency. Moderate depressive episode was diagnosed and selective mutism was suspected. Pharmacotherapy with escitalopram was started. Two weeks later, on the next visit, the mood was slightly improved and he denied suicidal thoughts. Still, he refused to return to school. He was offered the individual education (at the school) and gradual inclusion into the previous class. At the same time, the patient was referred for individual and group therapy as well as family consultations to a youth therapy center. However, he was very reluctant about individual therapy. He informed his parents that he had not know what to say during the sessions. The meetings went silent, broken only by the therapist's questions, but the patient did not respond. During one year of psychiatric care, psychoeducational-supporting meetings for parents were taken every three weeks. After 3 months of treatment with escitalopram, improvement of mood was observed and suicidal thoughts resolved. However, he still rationalized his social anxiety, avoidant behavior and symptoms of selective mutism. He explained that he regulated his social contacts by himself and he did not need the presence of other people.

Eventually, he started individual lectures at school after 12 weeks of the school year "because of the order from the school and the threat of a fine". During those lessons he felt a bit more comfortable than during lessons in the classroom because he was not disturbed by the presence of others and forced social interaction. However, on the other hand, teacher's whole attention was focused on him, which caused discomfort. Lessons consisted of teachers' lectures and written tests. During the entire year of treatment, the patient continued individual lessons, talked only with

some teachers and still avoided peers. He was promoted to next class. Within a few months, two consecutive therapists refused conducting the therapy with the patient “due to lack of his cooperation”. During the subsequent psychiatrist visits, he was encouraged to start behavioral training, similar to methods used in the therapy of autism spectrum disorders. During this period, he began to confront with social phobia symptoms (resulting in mutism), which were previously rationalized by him. In the next school year he returned to his previous class. Primary treatment effect at the described stage was the discontinuation of rationalization of mutism symptom, which allowed to take further steps in the treatment and to intensify the social skills training. The diagnosis of selective mutism was verified post factum into the “observation of Asperger syndrome”. The therapy and the observation of the patient were discontinued due to maternity leave of the psychiatrist, which prevented the final diagnosis.

Case 2

A girl aged 8 years, born at 37 weeks of gestation (in hypoxia). Her mother suffered from pregnancy-induced hypertension. Apgar score was 6 (1st minute) and 9 points (3rd minute), (due to decreased muscle tone). Although she was born at 37 weeks of gestation, some prematurity features were observed. Motor rehabilitation and support in speech development started when she was 6 weeks old, and neurologopedical rehabilitation – when she was 2–3 years old. Discrete delays in the neuromotor development were observed. She started to sit when she was 9 months old and to walk when she was 15 months old. Since 6 months of age she has been under the care of speech and language therapist. When she was 2 years old significant delays in speech development were observed (first words after 40th month of age, first two-word sentences at 4th year). She was observed for alalia, dysphasia and aphasia.

Speech development occurred after the implementation of an intensive left-hemisphere functions stimulation program and early reading learning using the method of Professor Jagoda Cieszyńska. The language system developed with delay at 4th and 5th years of age. Since then the girl satisfactorily communicated with the nearest family.

When she was 2.5 years old her neuromotor development was rated as proper, within the broader standards, but inharmonious. Active speech function (according to the scale of development by Brunet-Lezine) was the weakest. It was recommended to enhance the development of the child by playing and to continue speech therapy.

When she was 3.5 years old the emotional and social immaturity, insufficient vocabulary, poor form of speech (two-word sentences or elliptical sentences), low level of manual and graphomotor skills were observed. Her mental development was

inharmonious with non-verbal skills on average level. IQ rated by the Leiter International Performance Scale was $P = 93$. In subsequent examinations, when she was 4 years old and 4 and ½ year old, her emotional and social adjustment was slightly better, vocabulary and forms of expression enriched, but still insufficient in relation to the age standard. Simplification of consonant groups, substitution of rustling sounds by hissing ones and 'r' by 'l' was observed. Her mental development was at average level, still inharmonious. IQ rated by the Terman-Merrill Scale of Intelligence was 94. Level of graphomotor skills was low. Further stimulation of child's development, strengthening her independence and activity, creating the conditions for emotional and social development and social skills training were recommended.

She went to public kindergarten when she was 3 years old. For the first two years in the kindergarten she communicated only through gestures and, in spite of a satisfactory communication at home, in the kindergarten she did not speak. In the last year she had a very close relationship with one of girls from the group, she spoke to and played with her. She performed teachers' commands, benefited from pre-school educational program, but never made any oral answers. Due to the delay of speech development, she developed compensatory mechanism in relations with peers in the form of withdrawals from contacts with strangers or "expansive peers". Symptoms presented by her met the criteria for selective mutism defined by the ICD-10 and DSM-5 [1, 6]. However, that diagnosis has been modified in the course of further observation of the child.

Symptoms of withdrawal from peer relations resulting in refraining from initiating verbal contact become more acute after starting school education. It was probably caused by the need to adapt to the new school and enter completely new peer group. The child started pre-school education at the integrative school. As she was in integrative school, where the intensive therapeutic program with a psychologist, speech therapist, special education teacher, and sensory integration therapist was introduced, she become more open to contacts with peers, but she seldom initiated such contacts. In first grade she functioned satisfactorily, actively participated in lessons and spoke when answered by teachers. Still, she did not initiate contact with her peers. When other children initiated the play, she withdrew, spending time with teachers or the day care room teacher. However, mutism symptoms persisted in new social situations (e.g., during social meetings initiated by parents).

In the first few years of her life, the girl was diagnosed several times by an ENT specialist – a phoniatriest, who did not confirm any significant pathology of the hearing organ. At the age of 9 years, the tests of auditory attention and laterality revealed the reduction of right-ear external attention and the instability of right-ear internal attention. Bilateral discrimination of sounds in the range between 125 Hz and 8000 Hz and the location of sound sources for a frequency between 500 and 1000 Hz were also observed

As we know from parents, in the course of further observation the girl was diagnosed with Asperger syndrome. The behavioral improvement consisting in spontaneous initiation of contacts with people outside the family circle (including peers) was obtained after the completion of the intensive social skills training for children with autism spectrum disorder.

Case 3

A boy, 14 years old, first-grade junior secondary school student. No problems in early childhood development were noticed. He started to walk before he was 1 year old, he started to talk when he was 18 months old. He was shy since early childhood, he has never made verbal contact with strangers, but spoke freely with family members. He went to kindergarten when he was 4 years old, but did not adapt, so after one month the parents resigned from this form of care. He stayed at home with his grandmother. Because of the problems with contact with strangers he has been under the psychologist and psychiatrist care since he was 5 years old. A diagnosis of selective mutism was made, SSRIs were recommended, but pharmacotherapy was abandoned in further observation. An individual therapy was started, but the boy continued to speak only to family members, at the same time functioning well in the school peer group (in integrative class). Only after years of therapy a little improvement was observed – e.g., he whispered into his parents' ear when he was at the store, ordered an ice-cream in the presence of a therapist; he stopped to attenuate his voice when unknown person was around. In addition, till 9 years of age encopresis was observed, but stopped after the single intervention made by his mother (she threatened him that she would not throw him a birthday party). Till 10 years of age he did not want to get dressed by himself (he got dressed by his mother). He ate only selected food products (no fruits and vegetables).

The patient's family was complete, he had two older sisters. His parents were married, father was addicted to alcohol and did not take the treatment. Family history with mental illnesses: the mother's brother suffered from paranoid schizophrenia, he committed suicide; mother's sister – profound mental retardation; paternal grandmother – specific phobias type of anxiety disorders.

No abnormalities were found in brain EEG and CT. The patient was referred to psychiatric ward in order to determine the final diagnosis and to adjust the treatment. The physical examination and laboratory tests (also thyroid hormones and lipids levels) showed no abnormalities.

During the hospitalization, the boy refused verbal communication, he communicated with his doctor by writing. He gave short and precise answers to all the questions. During the examinations by a therapeutic group (several people) he did

not want to get even in written contact. After couple of days of adaptation, during the next examination by the therapeutic team he started to answer to closed questions by head movements. He did not answer many questions (especially those concerning his functioning). Refusal to speak was differentiated with oppositional defiant disorder and social phobia. In the end, it was concluded that the boy's behavior may correspond to those oppositional defiant behavior features which are often observed in children with SM.

He got in good peer contact in the ward quite quickly and he functioned well in this group. He participated in the organized and spontaneous activities (board games, sports activities), however, without initiating verbal communication. He was cheerful, without aggressive or self-aggressive behaviors, but his emotional reactions were poorly modulated. Throughout the 23-day period of hospitalization he did not talk to anybody, also with his parents with whom he corresponded only by SMS. The boy started attending therapeutic meetings based on genogram twice a week, but his parents discharged him from hospital on their demand. They said that patient had signaled them that "he is not going to talk anyway". Individual and family psychotherapy was recommended at discharge. We have no more information about that boy.

Discussion

Many authors emphasize that both the diagnosis and therapy of SM should consist of a comprehensive and multi-modal approach. Pediatricians advise to interview the patient, his/her parents and collect the data on the functioning of the child in the school environment during the diagnosis of selective mutism. Pediatrician, psychiatrist, psychologist, audiologist/phoniatrist, and speech therapist should participate in the final diagnosis, [15].

Due to the fact that SM is included in psychiatric disorders, not disorders on the borderline of neuropsychiatry and audiology, the primary therapeutic approach to the symptoms of SM are determined by the clinical psychiatry perspective. On the other hand, among the modern concepts of the phenomenon of selective mutism, the anxious pathogenesis of SM is predominant, which was highlighted in the DSM-5 [1]. This caused the introduction of the dominant therapeutic approach based on eliminating the consequences of phobic behaviors in children with SM in the recent years.

There are two main trends in therapy: (1) based on cognitive behavioral therapy or (2) based on pharmacotherapy used to treat anxiety disorders, i.e., SSRIs [34–36]. However, there are a few data on the effectiveness of both forms. According to Manassis [34], English-language literature includes approx. 100 cases of the use of SSRIs to treat patients with selective mutism. Based on changes in trends in the diagnosis

of SM initiated by the fifth edition of the DSM, it can also be assumed that the above therapeutic trends will be consolidated and expanded.

The authors of the ICD-10 in the introduction to the chapter “Disorders of social functioning with onset specific to childhood and adolescence”, emphasized heterogeneity of this group of disorders [8]. In the description of “selective mutism”, it is highlighted that “in a significant minority of children with selective mutism delays in speech development or the difficulty in articulation are reported” [8]. However, no classification points to the importance of e.g., auditory processing deficits or neurodevelopmental disorders for the diagnosis of SM. It should be noted that the subtle or severe neurodevelopmental deficits observed in the first years of education often lead to a later diagnosis of autism spectrum disorders. Specific language difficulties in people with Asperger syndrome were described by Tony Attwood as “a significant difference between language knowledge and practice” [37]. According to him, children with Asperger syndrome may show the linguistic efficiency in a formal speech examination, but show considerable difficulties in situations where fast processing of language in real situation, e.g., playing with peers, is needed. Another problem could be listening and understanding speech when noises are in the background [37]. Tony Attwood’s observations seem to be similar to the results of the research of Henkin and Bar-Haim and Henkin et al. [25, 32] on the auditory processing. The presence of such deficits may be important for behavioral techniques, pharmacological treatment or even the way of taking care of a child by his parents.

The presented cases illustrate the clinical difficulties involved in syndromological diagnosis of selective mutism. In Case 2 significant delays in language development were observed, while in two other cases there were no difficulties with speech development. In Case 1 symptoms of selective mutism were accompanied by anxiety disorders, and in Case 3 – by profound developmental disorders, behavioral dysfunctions (nonorganic encopresis) and mental disorders in family history.

Although in all cases the preliminary diagnosis of SM was made, in Case 1 and Case 3 the extreme form of social anxiety disorder, major depressive disorder, Asperger syndrome, oppositional defiant disorder, or first episode of psychosis should be considered during differential diagnosis. In Case 2 the impaired vocal expression was observed in the early development. The symptoms of SM observed at a later stage of her development could be differentiated with autism spectrum disorder, mental retardation and oppositional defiant disorder.

Epidemiological data indicate that children with symptoms of SM frequently meet the criteria for other mental disorders, such as depression, panic disorder, obsessive-compulsive disorder, dissociative disorder or Asperger syndrome (which is observed in up to 7.4% of children with SM) [10, 38].

In all three reported cases no sufficient therapeutic effects were observed. However, the presented clinical material does not allow for unambiguous generalizations

regarding the course of SM symptoms and the effectiveness of various methods used in the treatment of the symptoms of selective mutism.

Many authors also emphasize that there are no proven therapeutic methods or standards of pharmacotherapy in SM. Based on previous studies of people with selective mutism conducted from the perspective of clinical psychiatry, the largest amount of data (but still very limited) were collected for pharmacological methods. Thus, pharmacotherapy is an option worth considering when planning a comprehensive clinical treatment of SM, especially when SM symptoms are severe or when a previous psychosocial interventions did not help. Recently, Manassis described the case of successful treatment with fluoxetine in 8-year-old girl with SM and pointed the value of pharmacotherapy in this disorder [34]. Aspects of SM pharmacotherapy were also described in two systematic reviews of the literature by Carlson et al. [35] and Kaakeh and Stumpf [36]. However, in a study comparing the effects of fluoxetine and placebo ($n = 6$ vs. $n = 9$) in SM therapy, it was proved that SSRI reduced the severity of the symptoms in the majority of children, but did not eliminate all of the symptoms [39].

In the literature there is no long-term observation of patients suffering from SM in childhood or adolescence. Black et al. [39] proved that the symptoms of mutism persist despite the use of pharmacotherapy. In a naturalistic study with 6–8 month observation of the course of SM in children with comorbid social phobia, it was showed that symptoms tended to persist throughout the observation period, despite the use of different therapeutic interventions [40]. In the study consisting in one year observation of children with selective mutism after completing the cycle of behavioral treatment (24 sessions), it was observed that 12 out of 24 children with mutism no longer met the diagnostic criteria for this disorder after 12 months of follow-up [41].

In addition, the extension of this diagnostic category on the population of adults could further complicate the clinical observations regarding the course of the disorder. Observations presented in this paper are also time-limited. This can be an important factor limiting the possibility of a fuller analysis of long-term clinical implications of SM.

In cases presented above the clinical observations were discontinued due to different factors:

- 1) external factor (no possibility of further therapy by the attending physician) in Case 1;
- 2) interfamily factors (the child was discharged from hospital on parents' request) in Case 3;
- 3) partial recovery (however, still not fully satisfying) owing to sociotherapy and speech therapy in Case 2.

In those three cases of SM, the best treatment results were observed in Case 2, where multimodal approach in a child with preschool neurologopedic problems was

used. In this case, an auditory processing rehabilitation based on exercises to improve comprehension of linguistic utterance and on training of auditory attention (the series I listen and learn to speak by Wianecka) was used [42]. Rehabilitation program consisted of hearing exercises, with presenting letters in an extended way. This form of rehabilitation was used as a support for the girl's language system development (between 2 and 4 years of age). In subsequent years, the neurologopedic rehabilitation was no longer intensely focused on auditory attention training. However, in first years of school, her parents noticed lack of concentration of auditory messages. The girl seemed not to hear or not to understand messages addressed to her. Because problems with hearing were excluded by audiologist many times, parents explained such behavior as a temporary lack of interest in situation or the symptoms of negativity. The ability of auditory processing during simultaneous vocalization in the girl has never been evaluated. EEG showed no pathology. It was known, however, that all social situations when more than one or two people appeared in the child's presence caused a significant behavioral problem and the withdrawal from verbal contact. During the follow-up, selective verbal contact was considered to be a characteristic symptom of Asperger syndrome.

No audiological, foniatric or logopedic problems were observed in the boy described in Case 3. The development of his speech was not impaired. Although the boy was sent to the children's psychiatric ward to deepen and/or verify the diagnosis, the verification did not include audiological and logopedical diagnostics. There was no screening for autism spectrum disorder during the diagnostic process in the ward.

In contrast, the boy from Case 1 developed symptoms of SM gradually. From the beginning of his education the symptoms of social phobia were observed. Problems in social contacts with peers increased and reached its peak in comprehensive secondary school when the social requirements have increased significantly (he refused to attend school). In this case, the symptoms of SM could reflect both: the extreme form of social phobia as well as the manifestation of social withdrawal in the course of Asperger syndrome. The lack of early therapeutic intervention may have contributed to the development of symptoms of selective mutism and difficulties with mood control in later development.

According to the life span concept, the majority of mental disorders develops with a linear structure or trajectory, with gradual increase in frequency and severity of symptoms. Usually, it is impossible to distinguish the strict cut-off point when the symptoms of the disorder are unable to identify and when they are already present (according to the current diagnostic criteria) [43]. Taking into account the life span concept, one may suspect that unambiguous categorization of the symptoms of selective mutism among anxiety disorders without considering the neurodevelopmental

and audiological problems as well as the possible coexistence of sensory processing deficits may be a significant simplification.

The study by Kristensen from 2000 revealed that anxiety disorders are not the dominant comorbid pathology observed in patients with selective mutism [44]. This study showed, however, that anxiety disorders in children with SM are observed as often as various developmental deficits (74.1% vs. 68.5%). According to the author of that work, avoiding verbal contact can be a mechanism for “masking” the difficulties arising from various developmental or neurocognitive deficits. In this approach, inadequate SM therapy effects may result from the fact that the most common therapies (pharmacological and behavioral one) influence only anxiety component of the disorder and not include e.g., components associated with auditory processing deficits, neurocognitive deficits or social cognition deficits, which may occur in substantial number of children with SM.

If pathogenesis of SM may be linked to both developmental deficits and spectrum of anxiety disorders, it is extremely difficult (if not impossible) to create a single therapeutic approach to SM symptoms. Complex methods, taking the heterogeneity of symptoms into account should be introduced instead.

According to Henkin and Bar-Haim [25], auditory processing deficits should be taken into consideration in the diagnosis and treatment of SM. These authors propose training to enhance simultaneous auditory processing and loud speaking. Trainings start with exposure to one’s own voice via headphones, then there is subsequent addition of background noise and simulation of speech in social situations precipitating the speech-avoidance behavior. According to the authors, those techniques may facilitate auditory processing during vocalization.

Henkin and Bar-Heim also suggested a possible usefulness of transcranial magnetic stimulation (rTMS) or transcranial direct current stimulation (tDCS), which in combination with behavioral tasks may ameliorate effects of SM therapy, as it has been shown for neuro-rehabilitation of post-stroke aphasia [25, 45].

Conclusions

1. At the current stage of research on the pathogenesis and psychopathology of selective mutism, the concept of SM as social phobia type of anxiety disorder spectrum is most convincing. In the DSM-5, selective mutism is interpreted as an extreme form of social phobia.
2. Selective mutism syndrome is a non-homogeneous group of disorders of diverse etiology and course. Among the psychiatric aspects of disorder the ‘anxiety component’ of SM is first.
3. Comorbid developmental deficits and/or dysfunction of the auditory processing are often observed in individuals with selective mutism.

4. With regard to the life span concept, SM can be a syndrome manifested at an important developmental stages in children with neurocognitive or social cognition deficits.
5. The presence of comorbidities and developmental deficits may determine the future course of the disorder and the final effects of the therapy.

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Table 2. Development of selective mutism diagnostic category in subsequent editions of the DSM, (as cited in [6] with modification)

DSM Edition	Diagnostic category	Classification group	Diagnostic criteria
DSM-I (1952)		-	-
DSM-II (1968)	-	-	-
DSM-III (1980)	Elective mutism		Chronic refusal to speak in most social situations In some children – presence of delays in speech development and articulation disorders
DSM-III-R (1987)	Elective mutism	Other disorders of infancy, childhood or adolescence	Persistent refusal to speak in one or more social situations The refusal to speak is not due to symptoms of social phobia In some children – presence of delays in speech development and articulation disorders
DSM-IV (1994)	Selective mutism	Disorders usually first diagnosed in infancy, childhood, or adolescence	Consistent failure to speak in specific social situations in which there is an expectation for speaking (e.g., school), despite speaking in other situations The disturbance interferes with educational or occupational achievement or with social communication The duration of the disturbance is at least 1 month (not limited to the first month of school) The failure to speak is not attributable to a lack of knowledge of, or comfort with, the spoken language required in the social situation (e.g., children of recent emigrants who do not speak at school until they learn a new language) The disturbance is not better explained by a communication disorder (e.g., childhood-onset fluency disorder), developmental disorder, schizophrenia, or other psychotic disorders
DSM-IV-R (2000)	Selective mutism		
DSM-5 (2013)	Selective mutism	Anxiety disorders	