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Selective Mutism – Prevalence and Key Characteristics: A Population Study

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Abstract 1 [English]

We report the first-ever prevalence study of selective mutism in Poland. Preschool and primary school teachers were surveyed at four locations; parents were followed up if selective mutism was suspected. Our point prevalence rate of selective mutism (per thousand children) was 5.7 for 3-6 years-old preschoolers (95% confidence interval: 3.7-8.8); 2.0 (1.1-3.9) for 1st-3rd graders; and 2.1 (0.9-5.1) for 4th-6th graders. The accuracy of that estimate may be compromised by low response rate. 56% were girls. Less than 20% had been formally diagnosed with the condition, which suggests lack of adequate recognition and support.

Abstract 2 [Polish]

Mutyzm wybiórczy – rozpowszechnienie i podstawowe cechy: Badanie populacji

Przedstawiamy wyniki pierwszych polskich badań nad rozpowszechnieniem mutyzmu wybiórczego. Badania

przeprowadzono w czterech lokalizacjach metodą kwestionariuszową, ankiety wypełnili nauczyciele wychowania przedszkolnego oraz szkół podstawowych; w przypadku wstępnych podejrzeń mutyzmu wybiórczego także rodzice. Częstotliwość występowania aktywnego („niepokonanego”) mutyzmu wybiórczego (na tysiąc dzieci) wyniosła 5.7 wśród 3-6 letnich przedszkolaków (95% przedział ufności: 3.7-8.8); 2.0 (1.1-3.9) w klasach 1-3; oraz 2.1 (0.9-5.1) w klasach 4-6. Precyzja tej estymacji jest ograniczona niskim odsetkiem odpowiedzi. 56% grupy dzieci z mutyzmem stanowiły dziewczynki. Mniej niż 20% dzieci z mutyzmem miało formalną diagnozę tego zaburzenia, co sugeruje brak świadomości tego problemu oraz adekwatnego wsparcia.

Keywords 1 [English]

selective mutism, anxiety disorders, prevalence, children

Keywords 2 [Polish]

mutyzm wybiórczy, rozpowszechnienie, zaburzenia lękowe, dzieci

Introduction

Selective mutism (henceforth SM) is disorder characterized by a persistent difficulty with speaking in specific social contexts. It is included in the DSM-5 and ICD-11 classifications, which recommend the following diagnostic criteria:

- Remaining persistently silent in certain social situations that require speaking (e.g., at school).
- Normal or near-normal language competence, manifested by talking freely in other social situations (e.g., at home).
- Duration of silence is of over 1 month (not limited to the 1st month at school).
- Silence is not due to the lack of language (e.g., a child who has just begun education in unfamiliar language) or comfort in using that language.
- Silence cannot be adequately accounted for by other disorders (e.g., autism spectrum disorder or schizophrenia).
- The problem causes a significant impairment in functioning: interferes with education, occupation or social communication.

The manifestations and severity of SM vary considerably. A ‘classic’ picture would be that of a child who is completely mute and anxious in the public sphere (at school, interacting with strangers) but chatty and confident in the private sphere (at home, interacting with very familiar people). However, some children with SM are mute yet not socially anxious, happy to interact through gesture, facial expressions or writing. Yet others (called ‘low profile’ SM: Johnson & Wintgens, 2016) do respond verbally to questions, but do not *initiate* verbal communication. SM can be *highly* selective (e.g., the child speaks to peers but not to teachers).

Selective mutism typically starts manifesting itself fully when the child first enters preschool or primary school. While fortunately it tends to resolve with development, this may take several years; in a minority of cases it may become a lifelong condition. Longer-term SM, even once resolved, carries a risk of subsequent adjustment problems and psychiatric conditions (Remschmidt et al., 2001; Steinhausen et al., 2006).

Contemporary researchers agree that mutism is a type of anxiety disorder (Muris & Ollendick, 2015). Some authors (e.g., Johnson & Wintgens, 2016) conceptualize it as *a type of specific phobia* – a phobia of the expectation to speak. Seen from that perspective, the condition is similar to, e.g., spider phobia or agoraphobia. Others (e.g., Black & Uhde, 1995) see SM as *a variant of the social anxiety disorder* (social phobia).

SM – just like any other disorder – may be conceptualized in terms of predisposing, triggering and maintaining factors (Johnson & Wintgens, 2016). Factors predisposing to SM are likely to reside in child’s highly sensitive temperament and cognitive style: factors such as behavioral inhibition, negative emotionality, or attention bias to threat. Communication difficulties (e.g., developmental speech and language delays, or receiving education in a language other than the mother tongue) may also predispose to SM. The triggering factor is usually the preschool or school entry, though there are other possibilities (e.g., hospitalization). Maintaining factors reside mostly in the environment: well-meaning but ill-guided efforts to help the child may create the situation when the child is always ‘spoken for’, and has few incentives – and, indeed, few opportunities – to practice speaking.

SM can be treated effectively using behavioural and cognitive-behavioural techniques (such as stimulus fading, shaping, systematic desensitization, contingency management, auto-modelling)

(Cohan et al., 2006). It is essential to identify and remove the environmental maintaining factors, such as being always ‘spoken for’ and thus having no opportunity to practice speaking – or, conversely, being put under pressure to speak, which increases anxiety (Johnson & Wintgens, 2017). Pharmacotherapy has also been shown to be effective in some cases (Manassis et al., 2016).

The context and the aim of the present study

The research reported here is the first phase of our ongoing research programme into selective mutism in Poland: its prevalence, symptoms, mechanisms and effective therapy. The second author (Monika Burzyńska) undertook this study as a part of her PhD project.

Our experience suggests that SM is a significant problem that gravely affects many Polish children and some young adults, as well as their families and teachers. There are several SM support groups (the membership of which runs into thousands), designated support centres and training courses (which are in high demand). However, virtually no systematic Polish research on SM has been published, apart from a handful of case studies. A few publications that did appear (most of them in the last 5 years) are mostly literature reviews or handbooks.

Given this research lacunae, we decided to begin with the most basic of questions: **how prevalent is SM among Polish children, and how does it manifest itself?** In this paper we report a population survey which we set up to answer these questions.

Our literature searches identified 8 studies from other countries which attempted to estimate the prevalence of SM in the general population of preschool or school-age children (Bradley & Sloman, 1975; Brown & Lloyd, 1975; Kopp & Gilberg, 1997; Kumpulainen et al., 1998; Bergman et al., 2002; Elizur & Perednik, 2003; Karakaya et al., 2007; Sharkey & Nicholas, 2012). Their results make several trends apparent:

- The existence of SM has been documented in several countries.
- The condition is relatively rare; the prevalence is typically less than 1 per 100 school age children – but more than 1 per 1000.
- The prevalence decreases with age.
- The prevalence rate depends on how strict the diagnostic criteria are.
- The prevalence is markedly higher in bilingual than monolingual children.

We expected our study to produce similar findings.

Method

Study design & instruments

Our study used two-stage survey methodology. At the first stage, teachers were asked to report the number of boys and girls in the class/group under their care, and then answer four questions about those children: Is there a child in your class that:

- Never, or hardly ever, speaks to his or her peers – spontaneously says nothing, or hardly anything, in their company?
- Never, or hardly ever, answers questions asked by his or her peers?
- Never, or hardly ever, speaks to his or her teachers – even when appropriate?
- Never, or hardly ever, answers questions asked by his or her teachers?

If the answer to all of the following questions was negative, the survey was terminated and teachers were thanked for their participation.

If the answer was positive to at least one of those questions, the teachers were asked to answer further four questions (about child's sex, home language, ability to talk freely at home, the duration of mutism at school) and also to seek parental consent for two follow-up surveys:

- For the parent. This covered: history and severity of child's problems with talking (including their functional impact on the child and family), other developmental problems and concerns (and their impact on the life of the child and the family) as well as the formal clinical diagnoses which the child received or which were under investigation. The survey also included two psychometric instruments: Selective Mutism Questionnaire (Bergman et al., 2008) and Strengths and Difficulties Questionnaire (Goodman, 1997; <http://www.sdqinfo.com>).
- For the teacher. This covered: history and severity of child's problems with talking at school/preschool (including their functional impact on the child and the teacher), other developmental problems and concerns spotted by the teacher (and their impact on the child's functioning at school, and on the teacher). The survey involved two psychometric instruments: School Speech Questionnaire (Bergman et al., 2002) and the Strengths and Difficulties questionnaire.

Respondents

Our survey targeted four populations of children, all living in the Lower Silesia Voivodeship (province) in the south-west Poland:

- Wrocław county (*powiat wrocławski*) which consists of three small towns and several villages south of the City of Wrocław,
- Kamienna Góra county (*powiat kamiennogórski*), which consists of two small towns and several villages,
- Bogatynia commune (*miasto i gmina Bogatynia*), consisting of one town and several villages,
- Several districts in the City of Wrocław, encompassing roughly 1/5th of the city population.

Within that population, we targeted 3-6 years-old children attending state-run preschools, as well as 6-11 years old children attending state-run mainstream primary schools (all six grades, plus reception (so called ‘zero grade’). We excluded children attending private pre-schools and schools, specialist schools for special needs children, or not attending any of those (e.g., pre-schoolers staying at home; home schooled school-age children).

Overall, 43 preschools and 63 schools were invited to take part in the study. Of these, 28 (65%) and 36 (57%) did respond, respectively. The questionnaires returned by teachers reported on 496 groups/classes, and 10,208 children in total.

Procedure

The surveys were coordinated by four Psychological and Pedagogical Counselling Centres (*Poradnia Psychologiczno-Pedagogiczna*) specific to each location. Those Centres are state-run enterprises offering psychological and pedagogical support and career guidance to children, their parents and teachers, especially in case of special needs. Most of them operate on the catchment area (zoning) principle, being responsible for supporting schools and children in one particular administrative division.

Paper copies of all questionnaires and associated materials were distributed to teachers by designated staff at each of the four Centres. The teachers, in turn, contacted parents of children whom they suspected of SM, and distributed questionnaires to those parents.

The principle of informed consent was observed throughout. Information letters (containing definition of SM) and consent forms were distributed to school and preschool principals, teachers and the parents of children suspected of SM. The parents were also advised

where to seek help should they have any concerns about the development of their children.

Results

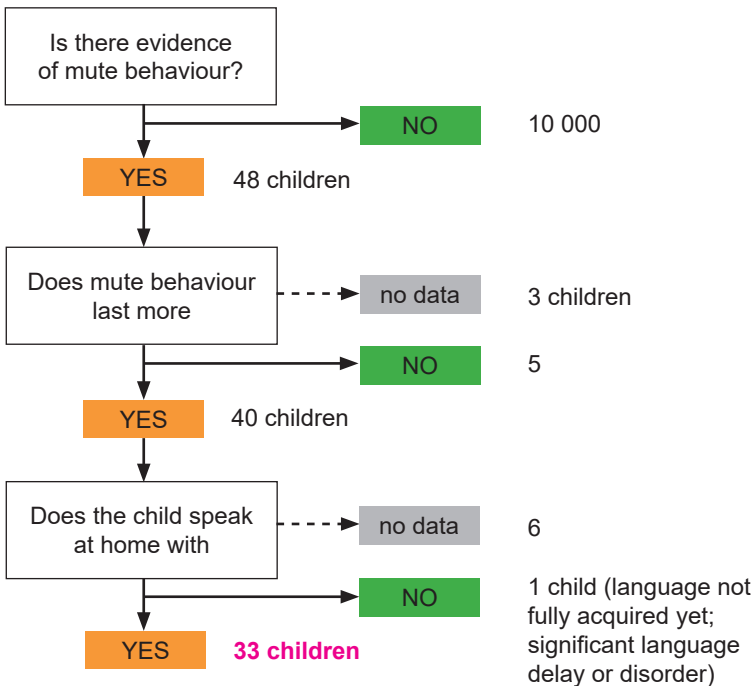
Prevalence of SM

We adopted three criteria of SM:

- 1) Evidence of mute behaviour (operationalized through teacher's affirmative answer to at least one of the four initial questions listed in the Method section above).
- 2) Evidence of mute behaviour being persistent (more than 1 month).
- 3) Evidence of mute behaviour being selective (child talking freely at home when in company of close relatives).

33 children met all three criteria (see figure 1) – though it is likely that the actual number is higher, as some criteria were impossible to verify for some children due to missing data.

Figure 1. Identification of children suspected of SM: a flowchart.



Additionally, one teacher identified a child with a history of SM, who, according to her, does not meet criterion 1 any more (i.e., he speaks freely). However, additional information (full parent and teacher questionnaires were available) indicated that that boy still shows residual signs of mutism and had been formally diagnosed with the condition. Consequently, we decided to include that child in our SM sample.

The prevalence estimates based on the final group of 34 children whom we identified as probable SM cases (15 boys and 19 girls) are presented in table 1.

Table 1. Prevalence rate analyses, grouped by gender and age.

	Boys		Girls		Total	
	n (sample)	Prevalence rate (95% CI)	n (sample)	Prevalence rate (95% CI)	n (sample)	Prevalence rate (95% CI)
Pre-school	8 (1783)	4.5 (2.3 – 8.8)	12 (1724)	7.0 (4.0 – 12.0)	20 (3507)	5.7 (3.7 – 8.8)
1 st – 3 rd grade	5 (2328)	2.1 (0.9 – 5.0)	4 (2093)	1.9 (0.7 – 4.9)	9 (4421)	2.0 (1.1 – 3.9)
4 th – 6 th grade	2 (1165)	1.7 (0.5 – 6.2)	3 (1145)	2.6 (0.9 – 7.7)	5 (2310)	2.1 (0.9 – 5.1)
TOTAL	15 (5276)	2.8 (1.7 – 4.7)	19 (4962)	3.8 (2.5 – 6.0)	34 (10238)	3.3 (2.4 – 4.6)

n (sample): number of children identified with SM, against the total sample size

prevalence rate: calculated PER THOUSAND children (i.e., 4.5 = 4.5 children with SM per 1000)

95% CI: 95% confidence interval for the estimate, calculated using Wilson score interval for the sample proportion.

We also considered including the 4th criterion of SM: that of a significant detrimental impact of mute behaviour on child's daily functioning. While such ecological criterion is fully warranted – indeed, mandated by DSM and ICD classifications – we dropped it for practical reason: the relevant data were missing for 6 out of 34 children. It is curious, however, that in 5 cases (out of 28) where the impact data were available, both parents and teacher declared that child's mutism has small or negligible impact on his/her functioning, which makes the diagnosis of SM questionable. Interestingly, of those five children, only one had significant difficulties speaking with their peers, for the other four mutism was apparent only speaking with adults.

The SM group: key characteristics

Detailed characteristics of the 34 children whom we identified as likely SM cases will be presented in a separate article. Here we present just a snapshot.

There were more girls (19) than boys (15) in the group, but the difference was not statistically significant – not even in the preschool sample where it was most pronounced (see table 1).

Virtually all of the children were monolingual. Only one child was identified as bilingual (the father and the father's family speak language other than Polish), though information on bilingualism was missing for some SM children.

Teachers confirmed that, in all 34 cases, they had been observing symptoms of SM for at least 3 months. Parents were asked to provide more specific information; out of 16 who did, 13 declared that the symptoms had been seen for 2 years or longer. Thus, a sizeable proportion of our SM sample appears to exhibit an entrenched problem.

Both parents and teachers were asked to state their concerns about child's functioning in four domains: emotional, concentration/hyperactivity, behaviour (being aggressive, rebellious or antisocial) and peer relations. Concerns about emotional difficulties and peer relationships were declared frequently by both parents and teachers – in contrast to other two domains that caused concern much less frequently.

Interestingly, according to parents' declarations, only six out of 34 children had a formal diagnosis of SM.

Discussion

Our study constituted the first systematic attempt to ascertain the prevalence of selective mutism in Poland. Indeed, to the best of our knowledge it is only the second group study of Polish SM children ever reported (after Bystrzanowska, 2017) and the first reported in the peer reviewed literature.

Our findings are broadly consistent with those reported elsewhere. Children with SM are rare, but they do exist. The impact of the condition is mostly on child's emotional wellbeing and peer relations. Disruptive, oppositional, hyperactive or inattentive behaviours are much less likely.

Only few children in our SM group were formally diagnosed with the condition. While this may sound surprising, it does not surprise us. It is only in the last few years the condition became more widely recognized in Poland, becoming the topic of public and professional

discourse. The awareness of SM, and knowledge about true nature of the condition and its seriousness remains low, even among professionals. Dismissing the condition as benign is common.

The main – and significant – limitation of the study was its survey-only methodology. Lack of resources made it impossible to follow up cases of suspected SM with individual interviews and in-depth clinical assessment, which would verify survey results (especially concerning the impact of SM on child's wellbeing) and fill in missing data points. We might have underestimated the true prevalence of SM, as we excluded 9 cases with incomplete data. Yet we might have overestimated it also, as we included 5 cases where the impact of mutistic behaviour on child's functioning appears to be small or negligible. The fact that some schools and preschools – and some individual teachers and parents – refused to participate, adds further uncertainty to our estimates.

The clinical experience of the 2nd author (Monika Burzyńska), who works with one of the populations covered by our survey, where she deals with ever increasing case load of SM referrals, suggests that *underestimation* is more likely.

According to the Polish Office of National Statistics (GUS, no date) there were some 1.51 million 3-6 years-old; 1.28 million 7-9 years-old, and 1.15 million 10-12 years-old living in Poland in 2017. Thus, assuming that our prevalence rate estimates of SM are broadly accurate, we can infer (using confidence intervals reported in table 1) that there are thousands of Polish children suffering from SM now: 5,600-13,300 3-6 year olds, 1,400-5,000 7-9 year olds, and 1,000-5,900 10-12 year olds. An overall figure of ten thousand is a conservative estimate.

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- Poradnia Psychologiczno-Pedagogiczna w Bogatyni.

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