Author’s response to reviews

Title: Service use, unmet needs, and barriers to services among adolescents and young adults with autism spectrum disorder in Poland

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“Service use, unmet needs, and barriers to services among adolescents and young adults with autism spectrum disorder in Poland” (BHSR-D-19-00482)

Dear Editor Ehsan Zarei,

Thank you very much for the opportunity to revise our paper. We have now carefully taken all the Reviewers’ suggestions into account and introduced the requested revisions. Parts of the methods, results, and discussion sections were rewritten to address the points raised by Reviewers. In particular, following Reviewers’ suggestions, we decided to limit the analysis presented in the paper to one sample, namely, the parents of young people with ASD. We also referred to Andersen’s model of service use and we broadened the analyzed variables to include predisposing and enabling factors for service use. Moreover, we put more emphasis to situate our results in the context of the Polish healthcare system. Finally, language corrections were made, including more person-oriented language.

In the following, we respond to the comments one by one. Given the high number of changes, we used citations of the revised or newly introduced content. Answers to the in-text comments made by one of the Reviewers were added as comments to the manuscript.
We hope that the revised version of the manuscript improved its quality and will meet your expectations.

Kind regards,

Mateusz Płatos

Elizabeth Young, MD, FRCPC (Reviewer 1)

1. Comment: "Background was well written, gave good context to ASD population in Poland and how services are administered. Great to have a context that is not north American.

Methods - Polish Autism Survey - authors gave good background about the efforts to recruit participants from service providers, social media and press. Overall low number of adults with ASD and they were mostly high functioning. Wondered if there was any other way researchers could have recruited this cohort to make the numbers more similar to the adolescents. (64 vs 311). The sample for adults with ASD may not be representative of population and also weighted very heavily to high functioning and had already a psychiatric comorbidity. In contrast, the 311 adolescents who seem more representative (almost 50-50 high to lower functioning, and 60-40 split with comorbid psychiatric diagnosis). The characteristics of the adult group (esp high comorbid psychiatric comorbidity) directly lead to their need for more psychologic and psychiatric services. Would this be better as two separate papers describing both groups separately given the difference in the cohorts from the onset in terms of mental health and IQ? "
Thank you for pointing out an important issue of the imbalance between two presented samples in terms of cognitive functioning and psychiatric comorbidity. Additionally, a clear and concise presentation of two samples with different respondents (parents and young adults with ASD) that were not related to each other was challenging. Therefore, we decided to follow your suggestions fully and to separate this data into two papers. In this revised manuscript, we only present data regarding the sample of parents of young people with ASD. As you have pointed out, this sample was larger and more balanced in terms of cognitive abilities and psychiatric comorbidity, so we were able to use these variables as factors in our analyses. Age was also taken into account in all the analyses because ascertaining age-related changes in service use was an important aim of the paper.

A decrease in the number of participants with age is typical even in large surveys including people with ASD (for example Hewitt, A. S., Stancliffe, R. J., Sirek, A. J., Hall-Lande, J., Taub, S., Engler, J., ... & Moseley, C. R. (2012). Characteristics of adults with autism spectrum disorder who use adult developmental disability services: Results from 25 US states. Research in Autism Spectrum Disorders, 6(2), 741-751). This stems from a recent spurt in diagnostics of autism spectrum disorders that affects mostly children, making every younger cohort of diagnosed individuals more numerous. However, what we did was controlling age in all the analyses and using regression analyses, instead of non-parametric comparisons to take into account age-related variance in the sample.

2. Comment: "Results - interesting, well laid out, good use of table 2 and table 3. Clear in terms of services used, requested and perceived barriers

Discussion - Confirmed rates of service use to be 2/3, with 1/3 excluded. Discussed which were the top services needed by each age group and the top barriers including availability and affordability. While this is a good summary, I would encourage authors to dive a bit deeper and provide more of an interpretation of their results. While it is clear that mental health services including psychology and psychiatry would be needed in the adult ASD group with high prevalence of comorbid psychiatric diagnosis, can the authors explore this more? Why the high comorbidity for mental health? What would these services look like? How would they be different for ASD than general population? Is this an ASD specific need in Poland or is it that nationally there is a paucity of mental health services? In Poland, which sector do these services belong to - how much can health care providers do, what other parts of the system need to step up to provide these or are these up to parents and private agencies? How expensive are these services in Poland? Which then would put the barriers of availability and affordability more into context? The authors touch on the paucity of services in Poland in medium and small cities but again situating this in the context of all services in Poland would be helpful similar to the mental
health services. Is this an ASD specific issue or a general health care issue. Also that this is not
surprising as access for rural areas is a problem globally. For discussion I would have liked the
authors to discuss which service needs were surprising, and which correspond with the literature
in other parts of the world? What can Poland do to address these unmet needs? What have other
countries done? Limitations - were clear but again I would comment on how specific the adult
population became in terms of higher functioning and high comorbidity with mental health. And
how does that compare with other population studies specifically for Aspergers? Maybe they all
have high prevalence for mental health and as such it is representative after all? "

Thank you for all the inspiring questions that encouraged us to rewrite the discussion and include
more information on the comorbidity of mental health problems among people with ASD, on the
Polish institutional context and how the services for people with ASD could be improved.

High comorbidity of mental health problems is widely internationally reported among people
with ASD, although it may be surprising for researchers from other fields. First, we mentioned
the fact of high comorbidity and potential reasons for that in the introduction:

“In turn, social isolation and unemployment likely contribute to alarmingly high rates of
depression, anxiety, and other mental health problems (Hofvander et al., 2009; Lugnegård,
Hallerbäck, & Gillberg, 2011) and higher rates of suicidal ideation in this population (Cassidy et
al., 2014).”

Second, we referred to other international reports in a sample description in the ‘Methods’
section:

“Over half of the young people (58.2%) had a diagnosis of a coexisting mental health problem,
similarly to international reports on that population (Buck et al., 2014; Matilla et al., 2010).”

Third, in the discussion we refer to this issue in a more elaborated way:

“High level of needs for the latter type of services is not surprising, given very high rates of
coeexisting mental health problems in the sample. Young people with ASD and coexisting mental
health problems used 30% more mental health services than those without these problems.
Elevated incidence of anxiety and mood disorders, as well as conduct disorders and ADHD, is
reported internationally in both adolescent and adult populations of people with ASD (Buck et
al., 2014; Matilla et al., 2010). Findings from the current study indicate that individual and group
psychotherapy and, to the lesser degree, psychological and psychiatric consultations are largely
underserved in young people with ASD.”
In the next paragraph, we provide a broader context for the paucity of mental health services in Poland and how that affects the affordability of these services:

“Another result put this finding in the perspective – only 30.3% of young people with ASD used services provided by public mental health clinics that are main institutions offering state-funded mental health support. In contrast, 55.0% of young people with ASD used private or NGO-based services (the latter can operate commercially or using state funds). In Poland, mental healthcare is largely underfunded, with only 8.5 psychiatrists per 100 000 people, compared to the average 17.2 per 100 000 in the European Union (Sowada et al., 2019). Long waiting time for psychiatric and psychological services funded by the National Health Fund results in the proliferation of private mental health services. In the present sample, 20.6% of young people with ASD used only out-of-pocket services, while 37.7% combined them with state-funded services.”

We expanded also on the regional inequities in access to services by bringing in the broader institutional context of Poland:

“Moreover, the area of residence was an important enabling factor for young people with ASD. More than a quarter of parents indicated services were too far away from their home. Participants from medium cities (i.e. having 50-200 thousand inhabitants) were at 3 times higher risk of finding services unavailable, compared to those living in large cities. This resembles the broader context of specialized, ambulatory healthcare in Poland, which is mostly located in large cities and, as a result, less used by inhabitants of smaller cities and rural regions (Central Statistical Office, 2017). There are 70 cities of medium size in Poland and most of them do not have any services for individuals with ASD funded by the National Health Fund (Szymańska et al., 2016).”

Following your next suggestion, we have discussed results that were surprising for us and explored potential reasons for that:

“Both overall use of services and use of each type of services decreased with age. However, age was not an enabling factor. In contrast to one American study (Turcotte et al., 2016), our study did not show an increase in unmet service needs with age. This suggests that an age-related decrease in service use among people with ASD may reflect fewer needs for services in older age groups. Alternatively, it can be a cohort effect, with an older cohort being less aware of available services and thus indicating fewer unmet needs.”
Finally, we expanded both the introduction and the discussion and provided more information about Polish healthcare and support system of people with ASD, as well as our insight on how these services might be improved. As previously cited, we pointed out that NGOs and private agencies dominate the support market while public mental health clinics, including “Clinics for People with Autism, which are specialized facilities that often provide SSTs and ABA treatment, are scarcely and unevenly distributed in Poland (Szymańska, 2016), again directing many people with ASD to out-of-pocket services” (Discussion: paragraph 4). Moreover, we observed that sensory/motor services “are provided mostly in education settings (i.e. schools and educational counseling centers) and by private therapeutic centers. This results in limited access to sensory/motor services and SLT for a few groups of people with ASD. First, in the present study, people with lower income had a greater number of unmet needs specifically for sensory/motor services. Second, parents whose children attended integration classes, compared to special classes, were at higher risk of facing too high costs of services. Third, many adults are no longer eligible for services in education settings, so they must rely on out-of-pocket services. In contrast to school and education clinics that were service providers for, respectively, 45.9% and 22.1% of young people with ASD, institutions that provide support to adults with ASD, such as Occupational Therapy Workshops or Community Self-Help Homes almost did not appear in the results of the survey. This result corroborates a previous Polish report that pointed out that these institutions are mostly unavailable for people with ASD (Jankowska et al., 2013).” (Discussion: paragraph 5)

We expanded also an introduction, noticing the current unavailability of the institutions, such as Occupational Therapy Workshops or Community Self-Help Homes:

“Previous research points out to common barriers to the use of services by people with ASD, such as low staff-to-service-users ratio, no autism-related training of the staff or overcrowded rooms in residential centers (Jankowska et al., 2013).” (Background: paragraph 5)

Taken together, we discussed the role of different public and private service providers in the current support system of people with ASD and how this system can be improved to bridge the gaps in the delivery of services. We hope that we addressed the questions you have raised.

3. Comment: "Conclusion - well informed by the data."
Angela Jury, PhD (Reviewer 2):

We appreciate all the useful comments and corrections made in the manuscript. We have applied those changes in a ‘track changes’ mode and answered to the comments in the manuscript.

1. Comment: "The work is a useful examining an important issue for people with ASD, and considers other key factors. To aid the reader and clearer messaging I would recommend editing to improve clarity and succinctness, and reducing the inclusion of unnecessary detail. The methods section in particular needs to be more succinct and include relevant information. A clear description of the Polish Autism Survey - what type of survey it is, who was included and how it was distributed is required as it could be misinterpreted as a population survey. Limitations of this need to be included."

We have reviewed the manuscript in light of these recommendations and improved succinctness of descriptions. In particular, we deleted unnecessary details in the ‘Methods’ section. Convenience sampling and recruitment process were clearly stated both in the abstract, as well as in the ‘Methods’ and ‘Limitations’ sections.

2. Comment: "I would suggest including some of the results in tables to improve succinctness, and considering how the samples are described to aid the reader."

We followed this suggestion by introducing a new table (Table 2 in a revised version), as well as simplifying existing tables. The data has been reduced to one sample, which aids a clear understanding of the results.

3. Comment: "Some additional contextual information would be useful to aid interpretation of the results and their relevance to Poland."

Some additional information was added in the introduction and, mostly, in the discussion to help the reader who is not familiar with the Polish healthcare system.

4. Comment: "Recommendations have been made to consider the use of more people centred language, and avoid the use of diagnostic labelling of people where possible."

People-centered language has been applied throughout the article.
Mohammadreza Zakeri, MD (Reviewer 3):

1- It is better to use Anderson health care utilization model; including 3 main stages of feel need, refer and receive the services.

Thank you for that recommendation! We have followed it and included the Andersen Behavioral Model of Health Services Use in the ‘Background’ section (paragraph 2, from the beginning). We used a useful division of determinants of service use into predisposing factors, need factors, and enabling factors for subsequent analyses and in the discussion.

2- For the count data it is better to modelling the data with Poisson-like distribution.

Thank you for this suggestion. We used Poisson regression for the count data. Whenever there was an issue of overdispersion, we used Negative binomial regression with log link (see Results: paragraph 3 and further).

3- High rate of other barriers may suggest that some individual barriers may be neglected! It is better to categorize the barriers in two main groups as user and provider (demand side/supply side).

Thank you for this recommendation. As the participants were provided with a list of pre-defined barriers, we were unable to change those categories after the studies. However, we analyzed closely all the additional answers filled out in an open field of the survey. The rate of those who indicated other barriers than pre-defined was 14% in the remaining sample. All the open-ended answers were coded (bottom-top approach), so that we made sure that no barriers were neglected (see: Results: Section ‘What barriers to services do individuals with ASD face and ho is at the greatest risk of facing those barriers?’; paragraph 3).
4- I did not find any reasons for using 12 months recall period for outpatient services. Most international surveys recommend 2 weeks to 1 month recall period for outpatients and 12 months for inpatient services.

In the mental health services research (outpatient) a year recall period or even longer is common, as the services may be infrequent (e.g. psychiatric consultations twice a year), seasonal or time-limited (e.g. 4-month social skills training in a year). Examples:


Australian Rural Mental Health Study (1 year recall period; Perkins, D., Fuller, J., Kelly, B. J., Lewin, T. J., Fitzgerald, M., Coleman, C., ... & Buss, R. (2013). Factors associated with reported service use for mental health problems by residents of rural and remote communities: cross-sectional findings from a baseline survey. BMC Health Services Research, 13(1), 157);
Evelina Pappa (Reviewer 4):

1. Comment: "The aim of the study was to investigate the service use, the unmet needs and the barriers the adolescents and young adults with ASD face in Poland. It is an interesting study focusing on people with ASD, a category of users that little is known.

Six points should be taken into consideration

- Concerning the sampling procedure, authors report that they contact all the non-governmental organization and in sum 204 service providers correspond to their request. The organizations vary significantly from mental clinics and psychiatric wards to occupational therapy workshops. This consequently means the large diversity of the sample which includes inpatients and those who attend an occupational therapy workshop. Furthermore there are two different samples, of unequal size, thus the question is how will authors draw a reliable conclusions? The authors should clarify why they use adolescents and adults with ASD and parents. Was the difficulty of selecting adolescents and adults with ASD? It must also be mentioned the organizations the two samples derived. "

Thank you for pointing out two sampling issues that increased the heterogeneity of the studied samples. First, the recruitment process covered almost all service providers known to authors, as well as traditional and social media. There were a few reasons for that. In Poland, adolescents and young adults with ASD are very hard to recruit. Many of them do not use any services or use services only from one provider. Limiting sources of participants would make it very difficult to obtain a sample large enough to analyze it statistically and draw reliable conclusions. Moreover, most of the services are cross-sectional in terms of providers. For example, Social Skills Trainings are commonly provided in public, NGO-based, and private therapeutic center, as well as in schools and day hospitals. However, we clarified in the manuscript (Methods: paragraph 2) that we did not include inpatients in the sample. Only day hospitals were contacted. Finally, we took into account the heterogeneity of the sample (e.g. in terms of cognitive abilities or mental health problems) in the statistical analyses that were also improved to account for several demographic factors simultaneously (see Results: paragraph 3 and further)."
The second point concerned using two different samples in the study. Given the limited knowledge on the services for people with ASD in Poland, we wanted to rely on multiple informants in conclusions we drew. However, we fully subscribe to your concern that the samples differed in many ways, which made the interpretation of results difficult. Therefore, we decided to withdraw one of the samples from the manuscript and analyze only the parent sample, which was larger and better balanced. The second sample may be described in another prospective publication.

2. Comment: "-The services under study include a wide range of social, educational, health services and training. In my opinion the study should be restricted to one sector, health services. Investigating the service use, the authors gave only the percentage of use while no statistical analysis was addressed in order to explore the predictors of each service use. It is expected that each service will be investigated separately."

This is the first study on the services for people with ASD in Poland to be published internationally and the study itself was exploratory in nature, so it indeed covered a broad range of services. Due to the study design, it was impossible to limit most of the analyses to one type of service. However, in the revised version of the manuscript, we grouped services into categories (including ‘mental health services’; see ‘Statistical analysis’ and Table 3) and explored predictors of service use and unmet needs for each category (see Results: paragraph 3 and further). We applied regression analyses for binary and count data to model multivariable predictors of each category of services.

3. Comment: "-Following the previous point, how unmet needs were defined in each service? For example how unmet needs in animal-assisted intervention was defined?"

Unmet needs were defined as the services that participants would like to receive, but they did not receive them at the time of the study. Participants were provided with a list of 11 services they could choose from. In some cases, short examples were given. In the case of animal-assisted intervention, the item was: ‘Animal-assisted intervention (e.g. hippotherapy, therapy dogs)’.
4. Comment: "Unmet needs and the barriers related to accessibility, acceptability and availability should be studied separately controlled for socio-demographic, health and non health factors. It is expected that barriers would vary across different types of services."

Thank you for that suggestion. Each unmet need and each barrier were modeled separately in regression analyses, based on socio-demographic and health factors (see Results: sub-sections 2 and 3). However, due to the design of the study, we were unable to explore barriers across different types of services. The question regarding service barriers concerned all the services received. However, we might take into account this variability of barriers in the next, more in-depth studies.

5. Comment: "Data in tables 2 and 3 must also be given in numbers. How many young adults report each barrier?"

We have now reported the numbers in all the tables that contained descriptive data.

6. Comment: "In table 4 how the authors handled the participants who reporting more than one barrier?"

As noted in the footer of Table 4: ‘Analyses were performed separately for each barrier.’ Separate stepwise logistic regression was performed for each barrier, so reporting more than one barrier by participants was not a problem.
Steven Merahn, MD (Reviewer 5):

Comment: "These reports are very important to understand the inter-national and cross-cultural nature of ASD's effects on family and community beyond early childhood; and an excellent idea to subset out the age cohorts from the Polish Autism Survey and explore the current state of the 'transition to adulthood'. On a minor note, the manuscript could use one more round of editing: for example: "As pointed (out) by Shattuck and colleagues (Shattuck et al., 2012) most of the studies on..." could be simplified to: "Many studies (Shattuck et al., 2012) on..." -- this would make your findings more accessible to the reader."

Thank you for stressing out the value of our study and for language recommendations. We have reviewed the manuscript in light of conciseness and made corrections accordingly. For example, the sentence you commented was simplified to: ‘Other factors … were rarely taken into consideration in the previous research (Shattuck et al., 2012)’. 