RESEARCH ARTICLE

Hypermobility among children with autism spectrum disorders and its correlation with anthropometric characteristics

Jaya Shanker Tedla, Faisal Asiri, Mastour Saeed Alshahrani, Kumar Gular

Abstract

Objective: To determine the extent of hypermobility in children with Autism Spectrum Disorder and to determine the correlation between age, gender, height, weight, BMI and hypermobility.

Methods: This cross-sectional study included 117 children with Autism Spectrum Disorder aged 2 to 17 years, of whom 91 were boys and 26 were girls. After obtaining the written informed consent from the parents of these children with Autism Spectrum Disorder, we assessed their level of hypermobility using the Beighton score.

Results: Out of 117 children, 47 (40.17%) were normal and 70 (59.83%) showed an abnormal increase in mobility. The average Beighton score was 5.33 ± 2.42. There is a moderate negative correlation between height, weight, and age with hypermobility.

Conclusion: The extent of hypermobility among children with Autism Spectrum Disorder was 60%, that is, 70 out of 117 children had hypermobility. Our results also suggested that the age, height, weight, and BMI of the child had a moderate negative correlation with hypermobility.

Keywords: Autism Spectrum Disorder, Hypermobility, Beighton score, Children and Ligament laxity.
(JPMA 71: 1076; 2021) **DOI: https://doi.org/10.47391/JPMA.436**

Introduction

Autism Spectrum Disorder (ASD) is an umbrella term, which includes conditions with problems of socialization, communication, and stereotypic problems.[1] Even though psychological and behavioural problems are commonly observed in children with ASD, motor impairments are not uncommon, the motor problems in these children include difficulties in gait,[2] balance, coordination, fine and gross motor skills[3] and sensory motor integration.[4] Moreover, all these motor impairments are not stable, and they are different between age groups.[5,6] The reasons for these motor problems are hypothesized directly to involve minimal brain damage, deficiency in motor control, and motor learning, and indirectly to muscle weakness, hypotonicity, hypermobility, and ligament laxity. Joint hypermobility refers to the excess of range of joint extensibility beyond the normal limits.[7]

Some studies indicated hypermobility in children with ASDs, but the clear amount of clarification is not there concerning the quantification of ligament laxity.[8,9] Klein et al. compared hypermobility between typically developing children and children with ASD. This study used a general range of motion (ROM) measured using a universal goniometer. The study did not use any criteria or a valid measurement tool that is specific to quantify the overall extent of hypermobility.[9]

Clark et al. reported that adults with joint hypermobility had chronic widespread pain, autonomic symptoms, gastrointestinal symptoms, and chronic fatigue syndrome.[10] Children with ASD who have hypermobility might also have similar issues, which the child would not be able to express due to issues with communication and social interaction. Hypermobility in the joints might lead to arthritis, low back pain,[11] altered neuromuscular control during activity[12] and it might even affect the muscle activation pattern.[13]

A recent review was published by Velasco et al. in 2018 in Frontiers of Psychiatry titled "Autism, Joint Hypermobility-Related Disorders and Pain" concluded that "Considering the high probability that pain remains disregarded and untreated in people with ASD due to communication and methodological difficulties, increasing awareness about the interconnection between ASD and hypermobility-related disorders is relevant, since it might help identify those ASD patients susceptible to chronic pain".[14]

We, as Paediatric Physical Therapists, see this problem in children with autism on a day to day basis. The children with ASD were suffering from hypermobility and related problems like pain and proprioceptive deficits. Due to their communication difficulties children cannot express this problem. Their sensory behavioural pattern changes

Department of Medical Rehabilitation Sciences, College of Applied Medical Sciences, King Khalid University, Abha, Kingdom of Saudi Arabia.
Correspondence: Jaya Shanker Tedla. Email: jtedla@kku.edu.sa

due to these problems.

Even though hypermobility exists in this population, there is no published evidence about this issue. This scientific research would create awareness among the medical team and and identify possible solutions to overcome this problem. Hence the current study aimed to determine the extent of hypermobility using the Beighton score among children with ASD and to find factors affecting this hypermobility.

Methods

In this cross-sectional study, we used the center for clinical research and biostatistics website[15] for calculating sample size, which amounted to a total of 117 children. As per our previous observation of such cases in our clinic, we observed that at least eight children out of one hundred cases of ASD had ligament laxity and we used this as a pilot observation while calculating the sample size. The formula used for sample size calculation was Z1-α/2 2 X p(1-p)/d2 where Z1-α/2 is standard normal variate, at 5% type I error ($p<0.05$) it is 1.96. In the formula p is the expected proportion in population based on previous studies or pilot studies. In our study we kept the p as 8 percent based on our pilot observations. In the equation d is the absolute error or precision which must be decided by the researcher, in our study the absolute error was kept as standard 5 percent. By considering all the above-mentioned values we calculated the sample size and it was 1.96 X 0.08(1-0.08) / 0.05 = 117 approximately.

These children with ASD were included in the study by convenience sampling. The Research Ethics Committee approved this study (REC # 2017-01-13). A developmental paediatrician, paediatric neurologist, psychiatrist or clinical psychologist diagnosed the child. They used the Diagnostic and Statistical Manual of Mental Disorders 5 (DSM-5) criteria for diagnosing ASD. We recruited all the children by referral to our center by paediatricians, neurologists, special schools and psychologists. The study is conducted in King Khalid University Hospital of Saudi Arabia. The duration of the study was one year.

Children with a formal diagnosis of ASD of either gender between the ages of 2 and 17 years were included in the study. The mean age was 7.23 ± 3.62 years. Children with ASD who had active convulsive disorders or recent musculoskeletal injuries or were under medications that alter the muscle tone were excluded.

Written consent was obtained from the parents of children with ASD. Out of 146 children selected, data was collected from 117 children; the other 29 children were excluded due to various reasons, such as unable to relax their body for measurement, anxious, and hyperactivity.

The general demographic characteristics of each child was noted. Detecto Eye-Level Beam Scale with Height Rod was used to measure weight and height. By using weight in KG/ Height in meter square we calculated BMI on the excel sheet. The evaluation was done by a 10-years' experienced physical therapist as per the standard protocol of the Beighton scoring system.[16] The Beighton score is one of the common methods for testing hypermobility in children.[17,18] The test has good intra and interrater reliability[19] and validity.[16] A major part of the test is passive except for forward bending, so it is easy to administer. The score was from 0 to 9 based on bilateral measurements.

As per the Beighton score, the children were divided into three categories. If the Beighton score was 0-4, they were grouped under a Normal ROM, if it was 5-6, they were grouped under an increased ROM, if it was 7-9 then they were grouped under hypermobile.[16] The Beighton score has five components; they are one passive dorsiflexion of the fifth metacarpophalangeal joint (one point on each side), two passive hyperextension of the elbow (one point on each side), three passive opposition of thumb to the flexor aspect of the forearm (one point on each side), four passive hyperextension of the knee (one point on each side) and five forward flexion of the trunk (one point). Among these, the forward flexion of the score has one point; the other four components are tested bilaterally with a score one on each side. All the range of motions were measured by using a universal goniometer The total score of the test should be nine.[11] During the data collection of the Beighton score, each component was taken three times and the best measurement was considered for analysis. There were different investigators for data collection and analysis.

SPSS version 20 was used for the analysis. The univariate analysis of the parameters such as age, height, weight, body-mass index (BMI) and Beighton scores was performed using descriptive statistics. The correlations between age, gender, height, weight, BMI and Beighton scores were obtained using the Spearman's correlation at the level of significance 0.05.

Results

Anthropometric parameters such as height, weight, BMI were measured and Beighton scores for hypermobility were assessed in these children. The details of these measurements are presented in Table-1.

Table-1: Age, weight, height, BMI and Beighton scores of children with ASD.

Parameter	Gender	Mean ± SD
Age (years)	Boys	6.92 ± 3.31
	Girls	8.34 ± 4.45
	Together	7.23 ± 3.62
Weight (Kg)	Boys	27.12 ± 15.61
	Girls	33.73 ± 20.94
	Together	28.59 ± 17.06
Height (Cm)	Boys	119.96 ± 20.99
	Girls	125.34 ± 24.82
	Together	121.16 ± 21.90
BMI	Boys	17.44 ± 3.88
	Girls	19.35 ± 5.65
	Together	17.86 ± 4.38
Beighton Score	Boys	5.30 ± 2.46
	Girls	5.42 ± 2.30
	Together	5.33 ± 2.42

Table-2: Diagnosis of laxity based on Beighton score.

Gender	Total Number of Children	Diagnosis of laxity based on Beighton score		
		Normal Normal ROM	Abnormal Increased ROM	Hypermobile
Boys	91	36	25	30
Girls	26	11	7	8
Total	117	47	32	38

Table-3: Correlation between age, gender, weight, height, BMI and Beighton score.

Parameter	Rs-value
Age	-.390(**)
Gender	.020
Weight	-.353(**)
Height	-.411(**)
BMI	-.285(**)

Note: ** Correlation is significant at the 0.01 level (2-tailed). BMI: Body Mass Index

Out of 117 children, 47 (40.17%) had a normal range of motions and 70 (59.83%) showed an increased ROM. Out of 70 children, 32 (27.35%) had increased ROM and 38 (32.48%) were hypermobile. The details of this abnormality are provided in Table-2.

The correlations between age, gender, weight, height, BMI and Beighton score were obtained using Spearman's correlation coefficients. We found a negative correlation between Beighton score and age, weight, height and BMI. The details of the correlations are presented in Table-3.

Discussion

The main aim of the study was to find the extent of hypermobility among children with ASD. In this study, approximately 60 percent of the sample had ROM more than normal. Till date many authors have spoken about hypermobility issues in children with ASD but the concrete quantitative evidence is lacking and this study provided unique literature about hypermobility among children with ASD.

Klein et al. conducted a study to determine the abnormalities in joint mobility and gait in children with ASD.[9] In their study, they measured index finger, wrist, elbow and ankle joints' flexion, and extension and compared them with their typically developing peers.[9] They found that except for the elbow joint, all three other joints showed significant hypermobility. In our study, we also found 60 percent of children with ASD showed increased ROM and hypermobility.

In our study, we had more boys who are affected with autism than girls, approximately 3.5:1 ratio, this was on par with other existing evidence, which mentions a ratio of incidence 4:1.[20] In the current study, we had a negative correlation of BMI with hypermobility, which was also shown in another study of normal children in the Indian population by Sanjay et al.[21]

Velaso et al. also believed that the children with ASD will have joint hypermobility due to which the children may suffer with multi system fragility leading to weak proprioception and motor system which causes musculoskeletal trauma and pain. Moreover, the children with ASD may not be able to express these issues due to communication difficulties which leads to irritability and increased stress levels.[22]

A recent study conducted on adults with Autism and related neuro developmental disorders by Csecs et al. also supported our finding that people with autism have hypermobility. They compared 109 adults with autism related neurodevelopmental disorders with general population in UK and found that this population had elevated rates of hypermobility than the general population. They also found a strong relationship between hypermobility and autonomous dysfunction and pain.[23]

Even though our study intended to find hypermobility in children with ASD, in some of the older children we found more muscle tightness.[24,25] During the evaluation, we were not ready for the quantitative measuring of tightness, which is the reason we could not determine the details of it. This was also supported

in our result by correlation analysis, which shows a negative correlation between age, height, weight and Beighton score.

The negative correlation is an association between two variables whereby they move in conflicting directions. In our study, there is a moderate negative correlation between height, weight, and age with hypermobility. This indicated that as height, weight and age increase the hypermobility decreases to a certain extent. The study supported the clinical hypothesis that there is hypermobility among children with ASD. The study proved the extent of hypermobility objectively among children with ASD by using the Beighton score. The current study showed that out of 117 children with ASD 70 children had hypermobility.

Restricting the sample to one city and having a wide age group from 2 to 17 years are some of the limitations of this study. Even though we have seen some correlations, which does not mean association and does not identify the cause.

Conclusion

The current study showed that there was increased ROM and hypermobility among children with ASD. Out of 117 children, 70 (59.83%) had increased ROM and Hypermobility values in Beighton score testing. There was a moderate negative correlation between age, height, weight, BMI, and hypermobility.

Disclaimer: The abstract has not been presented or published in a conference or an abstract book.

Conflict of Interest: The authors do not have any conflicts of interests

Funding Disclosure: We sincerely thank the Deanship of Scientific Research, King Khalid University, Abha, Kingdom of Saudi Arabia for funding this research project numbered R.G.P.1/52/42.

References

1. Schaefer GB, Mendelsohn NJ. Professional Practice and Guidelines Committee. Clinical genetics evaluation in identifying the etiology of autism spectrum disorders: 2013 guideline revisions. Genet Med. 2013; 15:399-407.
2. Esposito G, Venuti P, Apicella F, Muratori F. Analysis of unsupported gait in toddlers with autism. Brain Dev. 2011; 33:367-73.
3. Green D, Charman T, Pickles A, Chandler S, Loucas T, Simonoff E, et al. Impairment in movement skills of children with autistic spectrum disorders. Dev Med Child Neurol. 2017; 51:311-6.
4. Piek JP, Dyck MJ. Sensory-motor deficits in children with developmental coordination disorder, attention deficit hyperactivity disorder and autistic disorder. Hum Mov Sci. 2004; 23:475-88.
5. Van Waelvelde H, Oostra A, Dewitte G, Van Den Broeck C, Jongmans Mj. Stability of motor problems in young children with or at risk of autism spectrum disorders, ADHD, and or developmental coordination disorder. Dev Med Child Neurol. 2010; 52:e174-8.
6. Ming X, Brimacombe M, Wagner GC, Nelson KB, Grether JK, Croen LA, et al. Prevalence of motor impairment in autism spectrum disorders. Brain Dev. 2007; 29:565-70.
7. Sinibaldi L, Ursini G, Castori M. Psychopathological manifestations of joint hypermobility and joint hypermobility syndrome/ Ehlers-Danlos syndrome, hypermobility type: The link between connective tissue and psychological distress revised. Am J Med Genet C Semin Med Genet. 2015; 169C:97-106.
8. Baeza Velasco C, Hamonet C, Baghdadli A, Brissot R. Autism Spectrum Disorders and Ehlers-Danlos Syndrome Hypermobility-Type : Similarities in clinical presentation. C Med Psicosom. 2016; 118:49-58.
9. Shetreat-Klein M, Shinnar S, Rapin I. Abnormalities of joint mobility and gait in children with autism spectrum disorders. Brain Dev. 2014; 36:91-6.
10. Clark C, Khattab A, Carr E. Chronic widespread pain and neurophysiological symptoms in joint hypermobility syndrome (JHS). Int J Ther Rehabil. 2014; 21:60-8.
11. Beighton P, Grahame R, Bird H. Assessment of Hypermobility. In: Hypermobility of Joints [Online] 2012 [Cited 2017 January 27]. Available from: URL: http://link.springer.com/10.1007/978-1-84882-085-2_2
12. Junge T, Wedderkopp N, Thorlund JB, Søgaard K, Juul-Kristensen B. Altered knee joint neuromuscular control during landing from a jump in 10–15year old children with Generalised Joint Hypermobility. A substudy of the CHAMPS-study Denmark. J Electromyogr Kinesiol. 2015; 25:501-7.
13. Luder G, Schmid S, Stettler M, Mueller Mebes C, Stutz U, Ziswiler H-R, et al. Stair climbing-An insight and comparison between women with and without joint hypermobility: A descriptive study. [Online] 2015 [Cited 2017 January 29]. Available from: URL: http://linkinghub.elsevier.com/retrieve/pii/S1050641114001461
14. Baeza-Velasco C, Cohen D, Hamonet C, Vlamynck E, Diaz L, Cravero C, et al. Autism, Joint Hypermobility-Related Disorders and Pain. Front Psychiatry. 2018;
15. Sample size calculation. Centre for Clinical Research and Biostatistics. The Chinese University of Hong Kong. [Online] [Cited 2019 June 13]. Available from: URL: https://www2.ccrb.cuhk.edu.hk/web/?page_id=1016
16. Smits-Engelsman B, Klerks M, Kirby A. Beighton Score: A Valid Measure for Generalized Hypermobility in Children. J Pediatr. 2011; 158:119-23.
17. Kulik O, Gębska M. Comparison of Beighton score and Brighton Criterion in order to diagnosis of joint hypermobility in children. J Educ Heal Sport. 2018; 8:133-48.
18. Smits-Engelsman B, Klerks M, Kirby A. Beighton Score: A Valid Measure for Generalized Hypermobility in Children. J Pediatr. 2011; 158:119-23.
19. Evans AM, Rome K, Peet L. The foot posture index, ankle lunge test, Beighton scale and the lower limb assessment score in healthy children: a reliability study. J Foot Ankle Res. 2012; 5:1.
20. Werling DM, Geschwind DH. Sex differences in autism spectrum disorders. Curr Opin Neurol. 2013; 26:146-53.
21. Sanjay P, Bagalkoti PS, Kubasadgoudar R. Study of Correlation between Hypermobility and Body Mass Index in Children aged 6–12 Years. Indian J Physiother Occup Ther. 2013; 7:247-9.
22. Baeza-Velasco C, Cohen D, Hamonet C, Vlamynck E, Diaz L, Cravero C, et al. Autism, Joint Hypermobility-Related Disorders and Pain. Front Psychiatry. 2018 Dec 7;9:656.
23. Csecs JLL, Iodice V, Rae CL, Brooke A, Simmons R, Dowell NG, et al. Increased rate of joint hypermobility in autism and related

neurodevelopmental conditions is linked to dysautonomia and pain. [Online] [Cited 2020 September 15]. Available from: URL: https://doi.org/10.1101/2020.09.14.20194118

24. Barrow WJ, Jaworski M, Accardo PJ. Persistent Toe Walking in Autism. J Child Neurol. 2011; 26:619-21