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Analysis of three-dimensional image of anal sphincter muscle complex obtained from high-resolution 3D anorectal manometry in children who underwent surgery for anorectal defects. Image asymmetry analysis

Analiza trójwymiarowego obrazu kompleksu zwieraczy odbytu uzyskanego w manometrii anorektalnej 3D wysokiej rozdzielczości u dzieci poddanych zabiegom z powodu malformacji anorektalnych. Analiza asymetrii obrazu

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Abstract

Introduction and objective: The aim of this study was to evaluate the three-dimensional image of anal sphincter muscle complex which obtained from 3D anorectal manometry in children who underwent surgery for congenital anorectal defects. **Materials and methods:** The study group included 48 children operated on at the Polish Mother's Memorial Hospital – Research Institute from 2000 to 2020 due to anorectal malformations. All operations were performed using Peña's posterior sagittal anorectoplasty method. The reference group consisted of 28 children with no history of motility disorders of the distal gastrointestinal tract. Three-dimensional high-resolution anorectal manometry was performed in all children. The three-dimensional image of the anal sphincter complex was analysed in terms of its symmetry. The percentage of differences in the pressures in the images of both sides of the anal sphincter muscle, during squeeze and at rest, was calculated. Based on the pressure map, the puborectal muscle was visualised. **Results:** Asymmetry in the 3D image of the anal sphincter complex at rest and during squeeze in the study and reference groups was shown. In the study group, the mean percentage of asymmetry was 18% at rest, while in the reference group, it was 8%. It was 21% and 5%, respectively, during squeeze. In every patient in the reference group, the obtained image the identification of the puborectalis muscle. An image corresponding to this muscle was registered only in 22 patients (45%) in the study group. **Conclusions:** The techniques used in 3D high-resolution anorectal manometry provide the ability to reconstruct a computer image of the respective muscles of the anal sphincter complex and assess their contribution to the continence process.

Keywords: manometry, anorectal malformation, anorectal atresia

Streszczenie

Wprowadzenie i cel: Celem pracy była ocena trójwymiarowego obrazu kompleksu zwieraczy odbytu uzyskanego w manometrii anorektalnej 3D u dzieci po operacjach z powodu wrodzonych wad odbytu. **Materiał i metody:** Grupę badaną stanowiło 48 dzieci operowanych w Instytucie Centrum Zdrowia Matki Polki w latach 2000–2020 z powodu malformacji anorektalnych. U wszystkich pacjentów operację wykonano z dostępu strzałkowego tylnego metodą Peñy. Grupę referencyjną stanowiło 28 dzieci bez zaburzeń dystalnego odcinka przewodu pokarmowego w wywiadzie. U wszystkich dzieci wykonano trójwymiarową manometrię anorektalną wysokiej rozdzielczości 3D. Trójwymiarowy obraz kompleksu zwieracza odbytu analizowano pod kątem jego symetrii. Obliczono procentowe różnice ciśnień w obrazach po obu jego stronach, podczas skurczu oraz w spoczynku. Na podstawie mapy ciśnień uwidoczniono mięsień łonowo-odbytniczy. **Wyniki:** Wykazano asymetrię w obrazie 3D kompleksu zwieraczy odbytu w spoczynku i skurczu w grupie

badanej oraz grupie referencyjnej. W grupie badanej średni odsetek asymetrii wynosił 18% w spoczynku, zaś w grupie referencyjnej – 8%. W trakcie skurczu odnotowano odpowiednio 21% i 5%. U każdego pacjenta z grupy referencyjnej uzyskany obraz umożliwił identyfikację mięśnia łonowo-odbytniczego. Obraz odpowiadający temu mięśniowi zarejestrowano tylko u 22 pacjentów (45%) w grupie badanej. **Wnioski:** Techniki stosowane w manometrii anorektalnej wysokiej rozdzielczości 3D dają możliwość rekonstrukcji obrazu komputerowego poszczególnych mięśni kompleksu zwieraczy odbytu i oceny ich udziału w procesie trzymania stolca.

Słowa kluczowe: manometria, malformacje anorektalne, atrezja odbytu

INTRODUCTION

Anorectal malformations comprise a wide spectrum of developmental defects with varied severity and complexity. The incidence varies from one in 2,000 to 5,000 newborns and is slightly more common in males[1]. These infants require surgery in the first days or weeks of their life. The technique proposed by Alberto Peña – posterior sagittal anorectoplasty (PSARP) – was quickly accepted all over the world[2]. Unfortunately, despite even most skilfully performed procedure, the prognosis can be grave. The function of the newly created anus depends especially on the malformation type, but also on the development and function of the anal sphincter muscle complex and properly functioning nerve centres, including those in the sacrum. Anorectal manometry is a test allowing for the assessment of the neoanus. The aim of this study was to evaluate the asymmetry of the three-dimensional image of the anal sphincter muscle complex obtained from high-resolution 3D anorectal manometry (3D-ARM) in children who underwent surgery for anorectal defects.

MATERIALS AND METHODS

From 2000 to 2020, in the Department of Paediatric Surgery and Urology at the Polish Mother's Memorial Hospital – Research Institute, 212 patients with congenital anorectal malformation were operated on. In every patient, malformation repair was performed using Peña's PSARP method. The study group included 48 children aged from 3 to 17, consisting of 28 males and 20 females. The reference group comprised 28 children (14 males and 14 females) with no motility disorders of the distal gastrointestinal tract, such as constipation or incontinence, and children who had never undergone invasive treatment of the perineal region. Every patient and their legal guardians were informed about the study, its aim, and technique, and they gave written consent for participation.

	Age [years]	Treatment group	Reference group
Group 1	3–7	23	10
Group 2	8–13	14	10
Group 3	14–17	11	8

Tab. 1. Age groups of the patients

	Type of anorectal malformation according to Peña	Number of patients [*n*]
Group 1 (low risk of complications)	Rectoperineal fistula	14
	Rectourethral bulbar fistula	10
	Rectovestibular fistula	8
	Rectal atresia without fistula	6
Group 2 (high risk of complications)	Rectourethral prostatic fistula or rectobladderneck fistula	2
	Rectovaginal fistula	5
	Cloaca	5

Tab. 2. Low-risk and high-risk groups of long-term complications depending on the type of anorectal malformation

For the statistical analysis, patients were divided into three age groups, as presented in Tab. 1.

Additionally, two groups were differentiated according to the risk for complications, depending on the type of anorectal malformation (Tab. 2)[2].

Anorectal manometry was performed using the ManoScan™ AR which enables measurements of peripheral pressures in the region of the anal and rectal sphincters. The obtained three-dimensional computer image of the pressures generated by the anal sphincter muscle complex provided a functional visualization of the anatomy of these sphincters. In the subsequent part of the study, the three-dimensional image of the anal sphincter complex, obtained from both the study and reference group patients, was analysed in terms of symmetry. This analysis involved comparing both parts of the obtained computer image of pressures generated by the anal sphincter muscle complex and calculating the percentage of asymmetry between the left and right sides of the three-dimensional pressure map. In the literature, studies of healthy adults show that the spatial image of pressures in the anal canal on the left side is a mirror image of those on the right side. A deficit or weakening of muscles on one side of the complex causes lower pressure recorded on the respective side. The percentage of differences in the recorded pressures on the left and right sides of the anal sphincter muscle, both during squeeze and at rest, was calculated. A greater percentage difference between these values indicates greater asymmetry and motor dysfunction of one side of the muscle complex.

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	Reference group (*n* = 28)			Study group (*n* = 48)		
	n	***M***	***SD***	***n***	***M***	***SD***
Regardless of age group		0.08	0.05		0.18	0.14
Age group						
3–7 years old (*n* = 33)	10	0.07	0.04	23	0.21	0.16
8–13 years old (*n* = 24)	10	0.09	0.07	14	0.17	0.15
14–17 years old (*n* = 19)	8	0.07	0.05	11	0.16	0.11
n – number of patients; ***M*** – mean percentage of image asymmetry of pressures generated by the anal sphincter muscle complex; ***SD*** – standard deviation.						

Tab. 3. Percentage of asymmetry in 3D image computer reconstruction of the anal sphincter muscle at rest in the study and reference groups by age

	Reference group (*n* = 28)			Study group (*n* = 48)		
	n	***M***	***SD***	***n***	***M***	***SD***
Regardless of age group		0.05	0.03		0.21	0.12
Age group						
3–7 years old (*n* = 33)	10	0.05	0.03	23	0.21	0.13
8–13 years old (*n* = 24)	10	0.06	0.02	14	0.21	0.11
14–17 years old (*n* = 19)	8	0.04	0.03	11	0.19	0.13
n – number of patients; ***M*** – mean percentage of image asymmetry of pressures generated by the anal sphincter muscle complex; ***SD*** – standard deviation.						

Tab. 4. Percentage of symmetry in 3D image computer reconstruction of the anal sphincter muscle in squeeze in the study and reference groups by age

RESULTS

The asymmetry in the 3D image computer reconstruction of the anal sphincter complex at rest in the study group as well as in the reference group was shown (Tab. 3). However, in the reference group there was a lower percentage of asymmetry across all age groups in comparison to the study group. The first age group was characterised by the greatest asymmetry.

Asymmetry in the 3D image computer reconstruction of the anal sphincter complex during squeeze was observed in both the study group and in the reference group (Tab. 4). There was a lower percentage of asymmetry in the reference group in comparison to the study group across all age groups.

A correlation was shown between the type of anorectal malformation, categorised by low or high risk of complications, and the recorded asymmetry in the computer image of the anal sphincter complex at rest (Tab. 5). Children at low risk for these complications (group 1) had a lower percentage of asymmetry at rest compared to children at high risk for complications (group 2).

The study revealed a statistical correlation between the type of anorectal malformation (categorised as high or low risk for complications) and the asymmetry in the 3D image computer reconstruction of the anal sphincter complex during squeeze (Tab. 5). In children at low risk for complications, there was a lower percentage of asymmetry during squeeze in comparison to patients at high risk for these complications.

DISCUSSION

High-resolution 3D manometry generates a 3D image of the anal sphincter complex, reconstructed based on the pressures created by these muscles. The technique allows for the visualisation and assessment of anal sphincter function and asymmetry. It is of utmost importance for children who have undergone surgery for anorectal malformation. In the literature, multipoint and simultaneous measurement of pressures within the anorectum during high-resolution 3D manometry, followed by the analysis and processing of obtained data into an image of the anal sphincter complex, correlates with the images of these muscles obtained by magnetic resonance imaging (MRI) or transrectal ultrasound (TRUS)[3–6]. is the correlation has been confirmed in research conducted especially in adult patients[3,4,6]. Schuster's et al. conducted a study

	Type of malformation	*n*	*M*	*SD*	Median	Test value	Significance level (*p*)
Percentage of asymmetry in 3D image at rest	Group 1 – low risk of complications	38	0.13	0.10	0.11	-4.27^{U}	**<0.001**
	Group 2 – high risk of complications	10	0.38	0.12	0.40		
Percentage of asymmetry in 3D image in squeeze	Group 1 – low risk of complications	38	0.17	0.09	0.15	-4.07^{U}	**<0.001**
	Group 2 – high risk of complications	10	0.36	0.10	0.40		
n – number of patients; ***M*** – mean values; ***SD*** – standard deviation; ^{U} – nonparametric Mann–Whitney *U* test; ***p*** – significance level.							

Tab. 5. Dependent variables level: percentage of asymmetry in 3D image computer reconstruction of the anal sphincter muscle and type of anorectal malformation regarding defects with high risk (group 2) and low risk (group 1) of complications. Correlation analysis of these dependent variables

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involving 17 children after PSARP who underwent both anorectal manometry and pelvic MRI. The aim of this research was to assess the anal sphincter complex and the compatibility of the images in both exams. Results showed a significant correlation between the 3D images recorded in anorectal manometry and those obtained from MRI[7]. Emblem et al. conducted research with 40 patients (mean age: 16 years) who underwent surgery for anorectal malformations[6]. They had manometry and TRUS done. The results of the study showed a statistically significant correlation between the images obtained from TRUS and the reconstruction of pressures in 3D manometry. Similar results were presented by Caldaro et al., showing a correlation between the computer reconstruction of pressures in anorectal manometry and the images of the anorectum in 3D TRUS[8]. In several children, the TRUS exam showed minor disruptions in the internal and external anal sphincters that were not recorded in manometry. The study utilised one of the first models of an anorectal manometry device, which may have influenced the results. In the literature, there are no studies analysing the image of the anal sphincter complex obtained by high-resolution 3D anorectal manometry in children who underwent surgery for anorectal defects. Most assessments of the anal sphincter complex are conducted using MRI or TRUS. Single reports address the spatial assessment of pressures created by the anal sphincter complex based on 3D manometry in adults, which we reference in our paper.

The research based on manometry indicates that there is anterior-posterior asymmetry in the anal sphincter complex in both children and in adults[9–11]. The asymmetry is a result of the structure of the complex and corresponds to the functions of the external and internal anal sphincters and the puborectalis muscle. The puborectalis muscle presents as increased pressure in the proximal part of the image, at the back of the anal canal[12]. Data show that the spatial image of pressures in the anal canal on the left side is a mirror image of that on the right[9,10,13]. In the high-pressure zone (HPZ) on both sides of the image, there is the internal and external anal sphincter, and in the upper part of the image, centrally, there is the puborectalis muscle. Based on the measurements of these pressures, the manometry device visualizes areas of low pressure (blue) and high pressure (red), corresponding to the function and inferred structure of the anal sphincter complex.

We assumed, as Banasiuk et al.[11] did in their paper describing the results of 3D anorectal manometry in children who underwent surgery for Hirschsprung disease, that every deficit or underdevelopment in the anal sphincter complex would be visualised as a lack of or decreased pressure in the anal canal and reconstructed as an image of these deficits. During the examination, the percentage difference in registered pressures is calculated between the left and right side of the anal sphincter complex during squeeze and at rest. This allows the assessment of asymmetry in the 3D image created based on pressures in the anal canal. The greater the percentage of difference between pressures on both sides of the complex, the greater the asymmetry in the reconstructed image of the anal sphincters. Therefore, the greater the asymmetry in the image obtained from high-resolution 3D manometry, the greater the dysfunction of the anal sphincter complex on the respective side.

The asymmetry in the anal sphincter complex in patients who underwent surgery for various types of anorectal malformations is confirmed in many studies with the use of MRI and TRUS[5,6,14,15]. In the literature regarding patients after surgery for anorectal malformations, abnormalities in the anal complex are described as disruptions or thinning of the muscles[4–6,14,15]. Wong et al. presented a paper analysing asymmetry in MRI images of anal sphincters in children who underwent surgery for anorectal malformation using one of two methods: PSARP or laparoscopic-assisted anorectal pull-through (LAARP)[5]. Assessment of asymmetry was based on the thickness of the internal and external anal sphincters in MRI. The study included a group of just 10 patients, and a semiquantitive analysis of muscle structure abnormalities showed asymmetry in the anal sphincter complex in 40% patients who underwent surgery using LAARP and in every child operated on with PSARP[5].

In our paper, there was an asymmetry (right-left) of the anal sphincter complex 3D image during squeeze and at rest in both the study and reference groups. In the study group, the mean percentage of asymmetry was 18% at rest and 21% during squeeze, and in the reference group it was 8% at rest and 5% during squeeze. Thus, the difference in the percentage of asymmetry in the registered image in the study group was higher during squeeze than at rest.

Tang et al. conducted a study on group of 14 patients, of whom only six underwent surgery for anorectal malformation and eight were treated for Hirschsprung disease. They assessed anal sphincter asymmetry in 3D images obtained via anorectal manometry[16]. In their study, the reference group included three patients with constipation. In our study, children with any defecation disorder were excluded from the reference group. According to this study, asymmetry in the registered images of the anal sphincter complex in the study group was as high as 59–95% at rest and 45–81% during squeeze, which is considerably higher than in our study. In the group of children with Hirschsprung disease, asymmetry was also confirmed, with percentages of 23% at rest and 49% during squeeze[16]. In this study, asymmetry assessment was based on the calculation of asymmetry percentage in a 3D manometry image between the areas of the highest and lowest pressures, which is a totally different method from ours, where the percentage of asymmetry was calculated based on the entire image of the anal sphincter complex. According to our knowledge, the study by Tang et al.[16] is the only pilot study in the literature which assesses the anal sphincter muscle complex in children who underwent PSARP using 3D anorectal manometry. In the future, it will be essential to standardise

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the asymmetry assessment method. Although the results reported in both papers are divergent because of different asymmetry assessment techniques, it is remarkable that both Tang et al.'s study and ours found higher percentages of asymmetry (at rest and during squeeze) in the study groups in comparison to the reference groups. This raises the question of whether such slight asymmetry in the structure/function of the sphincter complex might be physiological, and, if so, what percentage of asymmetry would not affect the function of the anus and cause potential problems with defecation. Perhaps this slight asymmetry is due to the asymmetry in the anatomy of the human body, or possibly due to motor dominance of the right or left half of the body. Further research with a larger group of healthy children, including various age groups, seems crucial to clarify whether asymmetry (right/left) is possible in the recorded image of the anal sphincter complex in high-resolution 3D anorectal manometry.

Williams et al. carried out a study with 22 healthy adults who underwent TRUS[17]. Symmetry in the anal sphincter complex was assessed based on the maximal thickness of the sphincters on the left and right sides. In the internal anal sphincter, full symmetry was shown in 19 patients, and in the external anal sphincter – in 20 patients. Also, this study showed physiological asymmetry in the anal sphincter complex.

Our paper showed a statistically significant correlation between the percentage of asymmetry in an image of the anal sphincter complex in high-resolution 3D anorectal manometry and the type of anorectal malformation. In malformation types associated with a high risk for complications, the asymmetry in the anal sphincter image is greater than in those with a lower risk for long-term complications. Impaired development process of the anal sphincters is often observed during reconstructive surgery for anorectal defects (PSARP), especially in "high" malformation types. Even though this is quite a subjective assessment, an area of significant thinning/poor development of the anal muscle complex can often be identified with the "naked eye", as muscle stimulation shows very weak squeeze. Underdevelopment and asymmetry in the structure of these muscles are confirmed in MRI[7,15,18,19]. Madhusmita et al., in their study based on MRI images obtained in children after PSARP for anorectal malformations, classified as high and intermediate type of malformation (according the Krickenbeck classification), showed poorer development of the anal sphincter complex in comparison to the children with low-type defects, i.e. associated with a better prognosis. In this study, the MRI images of the anal sphincter complex was described as well-developed, moderately developed, or poorly developed. The authors did not comment on the symmetry in the obtained images[20]. In the study by Alamo et al., MRI showed significant underdevelopment, atrophy, and asymmetry in the structure of the anal sphincter muscle complex in patients with anorectal malformations classified as high[21].

Numerous previous studies indicate a correlation between the anatomical presentation of the anal sphincter complex obtained using imaging techniques and images of these muscles obtained in high-resolution 3D anorectal manometry based on pressures generated by the anal sphincter complex. Given this, along with the knowledge of the involvement of respective muscles in defecation and faecal continence, it can be presumed that asymmetry in the anal sphincter complex image obtained in 3D anorectal manometry corresponds to dysfunction of these muscles and can be partially responsible for complications, such as faecal incontinence and constipation.

The image of the anal sphincter complex obtained in high-resolution 3D anorectal manometry provides valuable information about the structure of the anal sphincters and allows for an assessment of the involvement of respective muscles in faecal continence. This is especially important for children who underwent surgery for anorectal malformations. In our study, based on the pressures recorded in the anorectum during manometry, the map of these pressures was reconstructed and the percentage of asymmetry was calculated in the image of the anal sphincter complex. In both the study and in reference groups, the image of the anal sphincter complex obtained at rest, as well as during squeeze, was asymmetrical – it was not a mirror image of the right and left sides, as previously described in the literature. This shows that there is physiological asymmetry in the anal sphincter complex. Further research with a larger group of healthy children of various age groups seem to be necessary to determine whether the asymmetry the in anal complex observed in high-resolution 3D anorectal manometry in our study is simply a result of a functional defect or whether is it caused by a structural abnormality of the anal sphincter complex, and at what point detected asymmetry might be linked to anorectal dysfunction. In the study group, the percentage of asymmetry is significantly higher in children who underwent surgery for anorectal malformation.

Summing up, our findings show that high-resolution 3D anorectal manometry is one of the most valuable diagnostic tests in children after surgery for anorectal defects. It does not require general anaesthesia, which is a significant benefit for children. It enables the functional assessment of the anal sphincter complex and sensation in the rectum, while also providing an opportunity to visualise the anal sphincter complex. Despite the fact that the anatomy of individual muscles of the anal sphincter complex is hard to capture, it seems that high-resolution 3D anorectal manometry offers meaningful insights into the function of the respective muscle of the complex. It is particularly significant in children who underwent surgery for anorectal malformations.

CONCLUSIONS

High-resolution 3D anorectal manometry is a valuable, non-invasive tool that allows detailed assessment of the

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structure of the anal sphincter complex in children who underwent surgery for anorectal malformations.
New techniques used in high-resolution 3D anorectal manometry enable the reconstruction of computer-generated images of respective muscles of the anal sphincter complex and assessment of their contribution to the continence process. It is extremely important for children following corrective surgery for anorectal defects.

Conflict of interest

The authors do not report any financial or personal connections with other persons or organisations which might negatively affect the content of this publication and/or claim authorship rights to this publication.

Author contributions

Original concept of study: BS. Collection, recording and/or compilation of data: BS, ML, KC, JŻ. Analysis and interpretation of data: BS. Writing of manuscript: BS, ML, KC, JŻ. Critical review of manuscript: APP. Final approval of manuscript: APP.

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