

Anal ultrasonography in monitoring and postoperative evaluation of anorectal malformations

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Abstract

Objectives: We aimed to describe anal ultrasound for monitoring anorectal malformations. **Methods:** A descriptive and retrospective study of patients diagnosed and/or intervened with anorectal malformations rated by anal ultrasonography, between 2016-2017, was performed. We employed a transducer of 10 megahertz to identify the anatomy of the anal channel.

Results: Eight patients, 5 men and 3 women were included, with a mean of 8.37 years old (range, 3-11). Three patients had a diagnosis of an anterior anus (not operated). Surgery was performed in 5 patients: 2 had a rectourethral fistula, 1 had a rectovesical fistula, and 1 had a rectovestibular and a perineal fistula.

The most significant ultrasound findings were partial absence of the anterior part of the external sphincter, internal sphincter hypertrophy in the middle channel, and absence of internal sphincter in the median and high channel plus the absence of external sphincter in the anterior region. These results belonged to a patient with clinical incontinence and to 2 patients that were previously diagnosed with an anterior anus.

Conclusions: Anal ultrasonography is a non-invasive method, economic, and feasible to be performed without anesthesia. It offers useful anatomical information to establish the best therapeutic option. Its main limitation is the unfeasibility of carrying it out on patients under 3 years old.

Keywords: pediatric, ultrasonography, anorectal malformation, anorectoplasty

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La ultrasonografía anal en el seguimiento y evaluación postoperatoria de malformaciones anorrectales

Resumen

Objetivos: Describir la ecografía anal en el seguimiento de malformaciones anorrectales. **Material y Métodos:** estudio descriptivo y retrospectivo de pacientes diagnosticados y/o intervenidos de malformaciones anorrectales, evaluados por ecografía anal, entre 2016-2017. Empleamos un transductor de 10 megahercios, identificando la anatomía del canal anal alto, medio y bajo.

Resultados: Se incluyeron ocho pacientes, 5 hombres y 3 mujeres, con una media de 8,37 años (rango, 3-11). Tres pacientes tenían diagnóstico de ano anterior (no operados previamente). Se intervinieron 5 pacientes: 2 presentaban fístula recto-uretral, 1 fístula recto-vesical, 1 recto-vestibular y 1 fístula perineal.

Los hallazgos ecográficos más significativos fueron: ausencia parcial de la parte anterior del esfínter externo, hipertrofia del esfínter interno en el canal medio en un paciente; y ausencia de esfínter interno en el canal medio y alto más ausencia de esfínter externo en la región anterior, en un paciente con incontinencia clínica, ambos pacientes previamente diagnosticados de ano anterior que posteriormente fueron intervenidos.

Conclusiones: La ecografía anal es un método no invasivo, económico, factible de realizarse sin anestesia y ofrece una información anatómica útil para establecer la mejor opción terapéutica. Su principal limitación es la imposibilidad de realizarlo en pacientes menores de 3 años.

Palabras clave: pediátrico, ecografía, malformación anorrectal, anorrectoplastia.

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Introduction

The reported incidence of anorectal malformations (ARM) is 1 in every 2500-5000 live births but may be more frequent in certain developing countries¹. ARM include a spectrum of anomalies on the pelvic floor with different subtypes of ARM².

The reconstruction is mainly made by Posterior Sagittal Anorectoplasty (PSARP), implemented worldwide in the late 80s³. The long-term outcome after a PSARP is reported to largely depend on the ARM subtype⁴⁻⁶. Some patients evolve unfavorably despite surgery and present with fecal incontinence or constipation. We believe that the anal ultrasound can provide valuable information in the diagnosis.

Anal ultrasonography (AUS) is a well-established imaging tool used in adults for the assessment of conditions including perianal Crohn's disease, anal sphincter evaluation, and rectal cancer staging⁷⁻¹⁰. It is widely regarded as being safe, well tolerated, and provides clinically relevant information. The feasibility and importance of AUS in the assessment of pediatric anal and perianal disease

are clear, although, there are only a few existing publications in this group of patients.

We aimed to demonstrate our use and the effectiveness of anal ultrasound in the monitoring and in the postoperative evaluation of ARM.

Material and Methods

We performed a descriptive and retrospective study of all patients seen in our clinic for ARM between 2016-2017, and for which an AUS had been performed. Before beginning this review, approval from the institute's research board and verbal and written consent were obtained from the parents of all the participants in this study.

We employed a BK Medical - Merlin 1101 Ultrasound System with a 10megahertz transducer with a metal adapter of 17 mm and a plastic cap with a perforated vertex (purge bubbles). The tube was filled with distilled water employing a 20 ml Luer syringe (degassed water/ bidistilled water/ water + emulsified dimethicone - eliminates bubbles). The probe was covered with sonographic gel, then coated with a condom and lubricant jelly on it (Figure 1).



Figure. 1 Anal ultrasound

The night before the test, a saline enema was given to the child. To perform an anal ultrasound, the patient was placed in left lateral decubitus with flexed lower limbs and knees to the chest

or if applicable in the lithotomy position. The anal probe when correctly positioned should show the puborectalis branches going upward with the transducer in a 90° angle (probe-anus).

The objective of AUS was to identify the anatomy of the high, middle, and low anal channels in all patients. This imaginary division allowed a better radiological evaluation and was based on the structures that were expected in each area, as described in the following text. The high anal canal was defined as the level midway between the inferior border of the puborectalis muscle. The complete formation of the external sphincter ring anteriorly the puborectalis was imaged as a horseshoe-shaped, mixed-echogenic structure forming the lateral and posterior portion of the upper anal canal. In the high anal channel, we looked for the integrity of the puborectalis muscle and the external anal sphincter (EAS) through the evaluation of the circumference, thickness, and disposition.

The middle canal level was defined by the completion of the external sphincter ring anteriorly in combination with maximum internal sphincter thickness; the internal anal sphincter (IAS) is represented by a hypoechoic band surrounded by the hyperechoic EAS. In the middle segment, the integrity (total circumference) and measures of the external sphincter, IAS, and disappearance of the internal sphincter, when the patient contracted the anal canal, were evaluated.

The low canal level was defined where the IAS was no longer seen. Only the hyperechoic EAS and surrounding soft tissues were visualized. In the low channel, the integrity (total circumference) and measures of the subcutaneous EAS were evaluated.

In all patients, we performed the anorectal manometry, for the functional study of patients, using a 4-channel probe- 3 for measuring pressure and 1 channel to inflate the balloon. To assess the mo-

tor function of the EAS, we identified the mean baseline pressure and range, the presence of anal inhibitory reflex, and voluntary contraction. All patients were evaluated at 3, 6, and 12 months after diagnosis or surgery when applicable. Continence was assessed by the clinical history of soiling and the number of stools per day.

Results

Eight patients were included, 5 men and 3 women with a mean age of 8.37 years (range, 3-11). No patients under 3 years old were included, as the ultrasound is not performed in this population. Three patients (37.5%) had a diagnosis of an anterior anus with clinical constipation and conservative management. Five patients underwent surgery: 2 had a rectourethral fistula (one of them incontinent), 1 had a rectovesical fistula, 1 had a perineal fistula, and 1 had a rectovestibular fistula (incontinent).

Four patients did not have any associated malformations. Comorbidities in our patient population included 1 patient with Noonan syndrome, 1 with esophagus atresia, 1 with hypospadias, and 1 patient with colonic dysplasia type B. Clinically, 4 patients had symptoms of constipation, 2 of encopresis, and 2 of incontinence. The surgical history included one primary repair and 4 repairs with a colostomy.

For the preparation for an endoanal ultrasound, all patients received a normal saline enema the night before the procedure. The most significant ultrasound findings in 2 patients previously diagnosed with an anterior anus, were: partial absence of the anterior part of the external sphincter and internal sphincter hypertrophy (mean of 4.5 mm) in the middle channel (Figure 2).

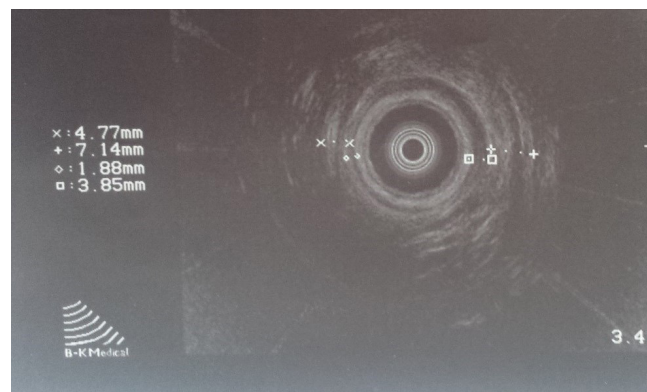


Figure 2. Anal ultrasound findings in patients with anorectal malformation. Partial absence of the external anal sphincter and internal sphincter hypertrophy in the middle channel.

In a patient with a rectovestibular fistula and with clinic incontinence, an echography absence of the internal sphincter in the middle and high channel plus the absence of the external sphincter in the anterior region was observed. (Table I) (Figure 3)

Table I. Findings of anal ultrasonography in patients with anorectal malformations

	Anorectal Malformation	Initial Symptoms	Findings of anal ultrasonography
1	Perianal fistula*	Constipation	Partial absence of the anterior EAS**, internal sphincter hypertrophy in the middle anal channel.
2	Anterior anus*	Constipation	Without alterations except for the anterior anus. Adequate stimulation of the anal sphincter.
3	Perianal fistula	Encopresis	Patchy defect of IAS*** in the high anal channel. EAS with normal measures and into its entire circumference.
4	Rectourethral fistula	Encopresis	Patchy defect of EAS and IAS in the high, medium, and low anal channel
5	Rectovesical fistula	Constipation	Low anal channel with thinning patched EAS.
6	Rectovestibular fistula	Incontinence	High anal channel without IAS. In de middle anal channel absence is almost complete of the IAS in all its circumferences, and the anterior quadrant of the EAS.
7	Rectourethral fistula	Incontinence	In the high anal channel patched EAS, least patched in the middle and low anal channel.
8	Perianal fistula*	Constipation	Middle anal channel with partial absence of the anterior EAS, internal sphincter hypertrophy

*Patients without previous surgery

**External anal sphincter (EAS)

***Internal anal sphincter (IAS)

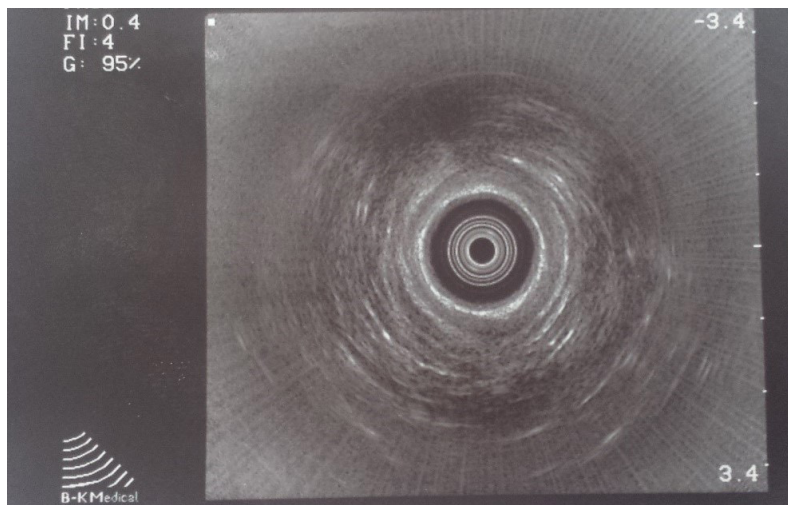


Figure 3. Anal ultrasound findings in patients with a rectovestibular fistula. In the middle channel, the absence of almost the complete internal anal sphincter in all its circumferences and the absence of the anterior quadrant of the external anal sphincter

Rectal manometry was performed in all patients but valid results were obtained only in six patients and in the remaining two lack of cooperation rendered invalid results. Among the most important findings, the mean resting pressure was 21.02 millimeters of mercury (mmHg) (range 8.54-54.7). Poor voluntary contraction in 2 patients with perianal fistulas and 1 patient with a rectourethral fistula already intervened. Additionally, a poor expulsive maneuver was identified in 2 patients with the new diagnosis of perianal fistulas and in a patient with vestibular fistula presenting incontinence.

After analyzing both anatomical and functional findings in these patients, surgical correction through PSARP was performed in the 2 patients previously diagnosed with anterior anus and currently diagnosed with a perineal fistula. In the same way, in a patient with a history of a vestibular fistula and now malpositioned, a redo anorectoplasty helped the patient to achieve continence. All patients were socially continent, with only one patient that had a rectourethral fistula being incontinent but clean on an enema regimen. The global average of stools per day was 1.18 (range 0.5-2).

Discussion

The behavior of ARM varies markedly and its management and in many cases it's a real challenge for the pediatric surgeon. The postoperative course depends on the type of ARM, the surgical technique used for correction, and significant comorbidities especially relating to the spinal cord.

Despite attempts at the management of these types of malformations at a global standardized level, some patients develop some degree of rectal incontinence, encopresis, or constipation that requires further diagnosis and treatment.

Anal ultrasounds are widely used in adult coloproctology because of their capacity to provide anatomic information about the anal channel. Due to its importance for continence, analysis of the muscular complex of the anal channel should be a fundamental part of the study of patients with the ARM, that present a torpid evolution. Previous studies, on rectal sonography in children with ARM, as published by Caldarò et al., have shown a positive correlation between the number of scars in IAS and incontinence¹².

In the same way, Wang Z et al., in 2016, included 47 children who had undergone PSARP or transperineal anorectoplasty for the repair of ARM. Endoanal ultrasonography showed significant differences in the thickness of the interior sphincter between the ARM patients and the healthy controls ($P < 0.05$)¹³. The authors considered that AUS must be complemented by other techniques that provide functional information from the anal canal such as anorectal manometry, thus realizing the anatomical and functional analysis of these patients.

In the present series, we found that AUS has multiple advantages. All of the procedures were performed on outpatient basis and without sedation. Our results, although limited, suggest that AUS findings have a great influence over clinical decisions in the treatment of patients with ARM.

In our series, a surgical correction through a PSARP was performed in 2 patients previously diagnosed with the anterior anus and then diagnosed with perineal fistula. In the same way, in a patient with a history of rectovestibular fistula and a malposition anus, who had already gone through an anorectoplasty, a new anorectoplasty was performed achieving continence. This latter finding differs from that published by Stenström et al., who studied 40 women with a history of ARM who were operated, and found that they have considerable defects on the pelvic floor but without any significant correlation to bowel symptoms¹⁴.

A potential limiting factor to the use of AUS in children is that it is not available in most pediatric centers, however, it is available in numerous adult centers and most pediatric centers are affiliated with adult counterparts. Another limitation is the age because the size of the ultrasound probe is not adequate for patients younger than 3 years old.

In conclusion, pediatric AUS for the study of the ARM has shown to be a non-invasive method, safe, feasible to be performed without anesthesia. It offers anatomical information, and thus guides the physician to the best therapeutic option to perform. Its main limitation is the ultrasound size, leading to the unfeasibility of carrying it out on patients under 3 years old.

However, it is a promising technology in the pediatric coloproctological area. However, due to

the small sample size, more prospective studies with more patients are necessary to generalize these results.

Abbreviations: anorectal malformations (ARM); Posterior Sagittal Anorectoplasty (PSARP); anal ultrasonography (AUS); external anal sphincter (EAS); internal anal sphincter (IAS); computed tomography (CT) and magnetic resonance imaging (MRI); millimeters of mercury (mmHg)

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Conflicts of Interest

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