



Three-dimensional endoanal ultrasound and anorectal manometry in children with anorectal malformations: new discoveries[☆]

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Abstract

Background/Purpose: Fecal incontinence and constipation are common problems in follow-up of anorectal malformations (ARMs). We evaluated the anal sphincters using the 3-dimensional endoanal ultrasonography (3D-EAUS) and the anorectal manometry after ARMs repair.

Methods: Seventeen patients, divided into 3 groups according to Wingspread classification, underwent anorectal manometry and 3D-EAUS. Clinical, manometric, and endosonographic scoring systems were used.

Results: The average anal resting pressure (aARP) was significantly higher in low ARMs than in intermediate and high ARMs. The anal squeeze pressure was not statistically different between the 3 groups. Three-dimensional EAUS visualized internal anal sphincter (IAS) disruptions in 7 of 17 patients and absence of IAS in 6 of 17 children with high ARMs. Scars of the external anal sphincter were localized in low ARMs and generalized in the other groups. In the case of IAS disruption with aARP greater than 20 mm Hg, fecal incontinence and constipation improved with biofeedback and/or laxatives, whereas daily enemas were necessary in absence of IAS with aARP less than 20 mm Hg. Statistical correlation was observed between endosonographic and manometric findings and clinical outcomes.

Conclusions: Lesions of the anal sphincter are common in ARMs. Three-dimensional EAUS and anorectal manometry ensure a complete assessment of the anal sphincter and could provide useful information to define the most appropriate treatments to improve the quality of life.

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Defecation is a complex process resulting from rectal compliance, rectoanal sensation, colonic motility, and the coordinated actions of both voluntary and involuntary

muscular structures [1]. The integrity of all these mechanisms is essential to ensure fecal continence. In patients with anorectal malformations (ARMs), which occur in 1 in 5000 live births, a wide spectrum of fecal continence disorders is observed because of congenital alterations or surgical injury [2].

The level of ARM (high/intermediate/low) affects the degree of development of the internal and external anal

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sphincters (EASs). Associated sacral or spinal anomalies could worsen the prognosis [3]. Since 1980, the posterior sagittal anorectoplasty has radically changed the surgical approach to ARMs, improving the preservation of the anorectal sphincters thanks to their anatomical identification [4-6].

However, in long-term follow-up, fecal incontinence (FI) and severe constipation remain the most frequent and disabling postoperative clinical problems in patients with imperforate anus because of their impact on the quality of life [7]. The postsurgical management of these anorectal deficits is very complicated. Bischoff et al [8] developed a bowel management program based on clinical scenarios and radiologic examination of the rectum.

A careful morphological and functional evaluation of the anorectal sphincter could improve the outcome of children with ARMs, thereby defining different treatments according to the status of the internal anal sphincter (IAS) and EAS. Anorectal manometry and 3-dimensional (3D) endoanal ultrasonography (3D-EAUS) are diagnostic examinations used to assess FI and constipation etiology in adult patients. Although anorectal manometry is now routinely used in the pediatric age group to diagnose Hirschsprung disease and to evaluate patients after anorectal surgery, the introduction of 3D-EAUS is a more recent innovation. Both procedures are well tolerated, safe, cost-effective, and minimally invasive [9].

The aims of this study were to investigate the efficacy and usefulness of anorectal manometry and 3D-EAUS for the diagnosis sphincter deficits in patients who had undergone a surgical procedure for ARMs and to correlate the sphincter damage to the severity of defecation dysfunction.

1. Materials and methods

Starting in 2009, 17 neurologically healthy children older than 4 years, affected by constipation and FI, and who had surgical correction of an ARM in the Surgical Neonatal Unit of our hospital or in other hospitals were enrolled in this prospective study. All patients were referred to our surgery and endoscopy unit, a pediatric referral center for FI, for the assessment of their defecation disorder.

Patients were divided into 3 groups according to Wingspread classification (group 1, low ARMs; group 2, intermediate ARMs; and group 3, high ARMs) and underwent clinical evaluation, anorectal manometry, and 3D-EAUS to assess the functional and structural abnormalities of the anal sphincter muscles. Ethical approval for this study was obtained from our research ethics board.

A single investigator performed all procedures. An enema was administered the day before the examinations. For anorectal manometry, a computerized system equipped with a Polygram interface (Medtronic, Minneapolis, MN, USA) and a water-perfused 4 radial hole silicone rubber

catheters (outer diameter, 4.5 mm) was used. A balloon was located at the tip of the catheter to elicit the rectoanal inhibitory reflex (RAIR). The examination was performed without sedation to ensure patient cooperation.

The average anal resting pressure (aARP) and the average anal squeeze pressure (aASP) were established. Rectoanal inhibitory reflex and rectal sensation (threshold, urgency, and maximum tolerated volume) were elicited inflating the balloon slowly. A manometric scoring system was devised using the aARP, aASP, and RAIR values to determine the activity of the IAS and EAS (Table 1).

Three-dimensional EAUS scans were performed using a Pro Focus Ultra view 2202 ultrasound machine (BK Medical, Mileparken 34, DK 2730 Herlev, Denmark) with a 10 to 16 MHz high-resolution 360° rotating endoprobe covered by a hard cone (outer diameter, 1.7 cm) with a built-in 3D mover (Ultrasound Transducers 2052; BK Medical). High-resolution 2-dimensional (2D) images were recorded at the proximal, middle, and distal levels of the anal canal. The patient was placed in the supine or left lateral position. The probe was introduced into the anal canal until the typical endosonographic hyperechoic U-shaped image of the puborectalis muscle (PRM) was visible (proximal anal canal). The probe was then held in place while the ultrasound crystal was activated and moved caudally, allowing a 3D volume of the full length of the anal canal. The midanal canal was recognizable by the presence of the internal (hypoechoic ring) and external (hyperechoic ring) sphincter, whereas the lower anal canal was characterized by the hyperechoic ring of the external sphincter only (Fig. 1) [7,10].

An *endosonographic sphincter defect* is defined as a discontinuity in the images of the IAS and the EAS. The lesions were classified according to Starck criteria, which are used to define the degree of obstetric sphincter trauma (Table 2). Based on depth (defects involving $\geq 50\%$ but not the entire thickness of the sphincter muscle were classified as “partial”), length, and radial extension (Fig. 2A and B) of any IAS and EAS defect, the Starck classification assigns a score from 0 to 16. The damage is classified as small (score of 1-4), moderate (score of 5-7), or large (score of 8-16) [11]. On 3D-EAUS, scar tissue appears as endosonographic areas of mixed echogenicity, whereas atrophy is seen as a thin and poorly defined sphincter with heterogeneous increased echogenicity [12].

Table 1 Anorectal manometry scoring system

	Score
aARP >20 mm Hg	0
aARP <20 mm Hg	2
aASP >55 mm Hg	0
aASP <55 mm Hg	2
RAIR present	0
RAIR absent	1

Scores of 0 and 1 indicate good, and scores of 2 to 5, poor.

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