

Negative Rectoanal Inhibitory Reflex in Anorectal Manometry...It is Not Always Hirschsprung's Disease

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Received: January 03, 2024; **Published:** January 29, 2024

DOI: 10.31080/ECCMC.2024.07.00874

Abstract

This study, conducted between June 2005 and June 2021, aimed to determine the presence of the rectoanal inhibitory reflex (RAIR) in patients with Anorectal Malformations (ARM) and its relationship with the height of the ARM. The study involved 63 patients aged 6 to 17 years who had undergone surgery for ARM using the Peña technique. The results showed that the RAIR was not found in 27% of the patients. Height of the ARM was significant in determining RAIR presence, being more frequent in patients with low lesions. The study also found higher frequency of rectosphincteric dyssynergia in high malformations than in low ones, which was statistically significant. The study concluded that the absence of RAIR is not always due to the presence of Hirschsprung's Disease (HD) and that taking a biopsy is not mandatory in all cases.

Keywords: Anorectal Malformations; Child; Anorectal Manometry; Fecal Incontinence

Introduction

In the study of anorectal function, anorectal manometry is commonly used in pediatric referral centers as diagnostic tool and to guide the treatment of children with intractable constipation and fecal incontinence [1].

Hirschsprung disease (HD) is characterized by the absence of enteric ganglion cells in the distal digestive tract, and is usually diagnosed in neonates, based on clinical symptoms, radiology, anorectal manometry, suction or surgical biopsies, and pathological anatomy. The manometric characteristic of HD is a negative rectoanal inhibitory reflex (RAIR).

Anorectal malformations (ARM) by definition: absence or narrowing of the anorectal canal or lack of communication between the rectum and the anus, with or without fistula to other organs [2]. They are rare anatomical anomalies, with a prevalence of 1:4000 to 1:5000 live newborns. ARMs originate from the interruption of the normal caudal development of the embryo in its first weeks of life. The associated malformations affect quality of life of these patients. Constipation and/or fecal incontinence occur in the majority of these children. In the therapeutic and prognostic approach of ARM, each patient must be considered chronic; it is necessary to inform the family of the requirement for controls and follow-up to minimize the impact of sequelae or possible complications.

There are very few papers on anorectal manometry in patients with ARM and all of them with few patients.

Objectives of the Study

- The main objective was to determine the presence of RAIR in a population of patients with ARM.
- The secondary objective was to study the relationship of the RAIR with the height of the ARM.

Materials and Methods

Descriptive, analytical, retrospective study. It was performed between June 2005 and June 2021. Patients with anorectal malformation operated on with Peña technique (posterior sagittal anorectoplasty), treated in the Pediatric Gastroenterology Service of the Prof. A. Posadas National Hospital in Argentina.

N: 63 patients. Age: 6 to 17 years (average 8.44 years), 58.7% (37) male.

30 patients presented associated malformations (the most common comorbidities were 8 cardiac and 8 renal). Previously colostomized: 43; 6 without colostomy and 14 without data about it.

According to the type of malformation they were classified: High (without fistula to the perineum): 26 (41%), Low (with perineum fistula): 29 (46%), Cloacae: 8 (13%).

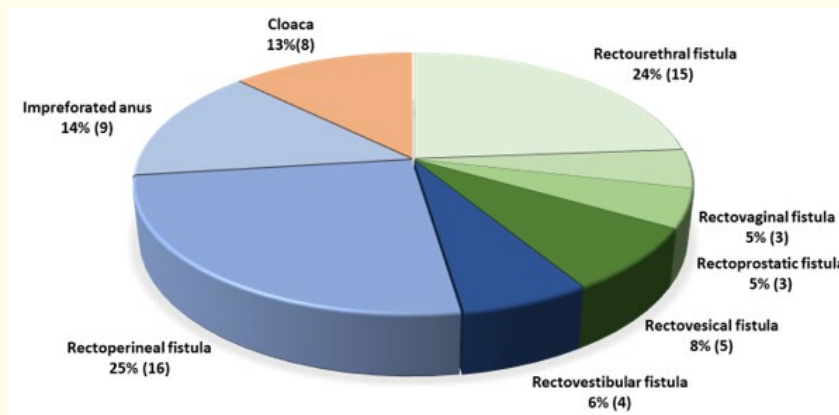


Figure 1: Type of malformation.

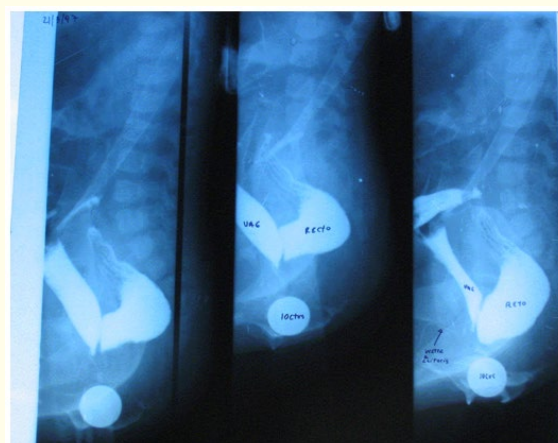


Figure 2: Cloacae.



Figure 3: Imperforate anus.

All patients underwent anorectal perfusion manometry. For the manometries, a STA 01 Manometer with water perfusion system and Nitrogen pump, with 6 perfusion channels, was used. A latex-free balloon catheter with 4 radiated channels was used. The patients were placed in left lateral decubitus position with knees flexed at 90°.

The probe was located 10 cm from the anus and was removed using the pull throw technique, cm by cm until anal sphincter was located [3,4].

RAIR was defined as the drop in the resting pressure at the anal channel, upon inflation of the balloon, with the appearance in the recording of a declining plateau, with a fall greater or equal to 5 mmHg that then recovers the initial pressure values. RAIR was obtained by inflating and deflating the rectal balloon in 2 seconds with increasing volumes up to 300 cc of air or when the maximum tolerance was reached [5-7].

The protocol was approved by the Bioethics and Research Committee of Professor Alejandro Posadas National Hospital. Registration code of this protocol for the CEPIC, ref.: 224 LUPOSO/18. It was exempt from requesting informed consent because it was a retrospective data collection work, with protection of the information of the research subjects (as stated in the documentation presented to the Bioethics and Research Committee).

Inclusion criteria: People over 6 years of age with anorectal malformation operated with the Peña technique, with constipation and/or fecal incontinence.

Exclusion criteria: Patients with neurological disorders who did not respond to the study instructions.

Statistical analysis: The Chi square or Fisher test was used for qualitative variables and the Student t test or Mann-Whitney test was used for quantitative variables according to distribution. A p value < 0.05 is considered significant.

Variables analyzed: Age, sex, resting pressure, squeezing pressure, Pushing, RAIR, type of ARM.

The data obtained does not always correspond to 100% of the patients, since it is a retrospective study and data may be missing from the report or chart.

Results

Manometric data:

- Resting pressure of the anal canal: 7 to 61 mmHg (mean 30).
- Squeezing pressure: 24 to 120 mmHg (average 69.5).
- Push: 20 relaxation, 22 paradoxical, 4 no response.
- In 40 (63.5%) positive RAIR; 17(27%) RAIR (-) and 6(9.5%) without data.
- RAIR duration: 8 to 22 seconds (average 12.67 seconds).
- Sensation: present in 40 (71.43%); absent in 16 (28.57%).

	High	Low	Cloacae
Resting pressure (mmHg)	27,11	33,99	27,80
Squeezing pressure (mmHg)	67,00	72,32	67,40
Push	6 relaxed (31,6%) 13 paradoxal (68,4%)	13 relaxed (61,9%) 8 paradoxal (38,1%)	1 relaxed 1 paradoxal
RAIR	15 positive 10 negative 1 no data	22 positive 4 negative 3 no data	3 positive 3 negative 2 no data

Table 1: Manometric findings according to the type of malformation.

The presence of RAIR was significantly more common in low ARM than in high ARM (p: 0.049).

We found higher frequency of paradoxical pushing in high malformations than in low ones and this was statistically significant (p: 0.022).

Discussion

Anorectal manometry is widely used and accepted for the evaluation of anorectal function in different types of anorectal malformations, as revealed by the study carried out by Kumar with conventional manometry [6].

More recent works such as that of M. Banasiuk with three-dimensional high-resolution anorectal manometry (3DHRAM), found that it is the most accurate tool to evaluate the function of anal canal in children after surgery to detect anorectal disorders. They prospectively included 43 children after surgery for Hirschsprung’s disease, anal atresia, or proctocolectomy. The lowest values of resting pressure, squeezing pressure and puborectalis muscle pressure were observed in the anal atresia group [8].

However, the benefit of 3DHRAM over conventional manometry in clinical practice is questioned in contrast to the esophageal field. There are no data showing additional benefits over conventional manometry in the treatment of fecal incontinence, and the role of 3DHRAM in discriminating healthy individuals from patients with dyssynergic defecation is unclear [9].

Anorectal manometry provides an assessment of pressure activity in the anorectum and provides complete information on rectal sensation, rectoanal reflexes, and anal sphincter function, at rest and during contraction and defecatory maneuvers. It is an established and widely used research tool for defecatory disorders.

The rectoanal inhibitory reflex is absent in Hirschsprung's disease. The qualitative evaluation of the RAIR can be valuable in the diagnosis of constipation, since this reflex is altered in the majority of patients with constipation and spinal cord injuries [10].

Regarding the modulation of the RAIR, our group published the differences in the duration of the inhibitory rectoanal reflex in patients with refractory functional constipation and myelomeningocele, in which we demonstrated that it is prolonged in patients with spinal cord alteration [7]. Other pathologies may have altered RAIR, such as organic pathologies like diabetes and autoimmune diseases with neurological involvement [11].

We found absence of RAIR in 27% of the patients operated on for ARM, but we did not find alterations in terms of its characteristics.

Complications after ARM correction are constipation and fecal incontinence; For these patients, a diet rich in fiber, habits and laxative medication are indicated, and if they do not respond, biofeedback [12,13], transabdominal or sacral electrostimulation [14-16] can be performed.

In Yang's paper, anorectal manometry was performed on 23 patients with ARM, and they found RAIR present in 80% of the patients [17].

Kumar, at Kuwait, performed anorectal manometry on 32 children operated on for ARM, finding lower resting pressures than controls (21 mm versus 42 mm) and RAIR (+) in 38% of the high ones and 58% of the low ones [18].

Caldara, in Rome, performed conventional manometry on 17 children with ARM, finding higher resting pressures in low malformations. The RAIR (+) in 83% of low, 50% of intermediates and 16% of high [19].

In a study with an elevated number of patients with high or intermediate ARM (85 children), Vittal Junior, *et al.* only found the presence of RAIR in 17% of the patients [20].

Our study had similar results with published papers, regarding the greater probability of negative reflex in patients with high malformations than in low ones, which for us was statistically significant.

We consider that the absence of the RAIR in patients with ARM may be due to the malformation itself or the surgical scar.

Conclusion

- Rectoanal inhibitory reflex was not found in 27% of patients.
- The height of the ARM was significant in determining the presence of the RAIR, being more frequent in patients with low lesions.
- We found a higher frequency of rectosphincteric dyssynergia in high malformations than in low ones and this was statistically significant.
- Based on the results of our study we can assert that the absence of RAIR is not always due to the presence of HD and that taking a biopsy, therefore, is not mandatory in all cases.

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Volume 7 Issue 2 February 2024

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