

Anorectal manometry in children with chronic functional intestinal constipation refractory to treatment

Manometria anorretal em crianças com constipação intestinal crônica funcional refratária a tratamento

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ABSTRACT

Objective: To evaluate anorectal manometry results in children with chronic functional intestinal constipation refractory to conventional treatment regarding mean anal sphincter resting pressure, presence of recto-anal inhibitory reflex (RAIR), expulsion capability of the intra-rectal balloon and behavior during the defecation maneuver.

Methods: Retrospective study by review of medical records from January 2003 to June 2007 with 31 patients who presented chronic functional constipation refractory to standard treatment. Anorectal manometry was performed by a four-channel catheter (MPX 816 processor and Proctomaster 5.0 software, Dynamed).

Results: Among the 31 studied children, 24 (77%) were male. The mean age was 8.9 ± 2.6 years. The mean maximal squeeze pressure of the anal sphincter was 82 ± 38 mmHg. Among the patients, 15 (48%) had normotonic anal sphincter and 16 (52%) hypertonic anal sphincter. RAIR was present in all patients. Intra-rectal balloon expulsion was positive in 12/31 patients, being 4/12 with normotonic sphincter and 8/12 with hypertonic sphincter ($p=0.22$). Anism was detected in 6/15 patients with normotonic sphincter (40%) and in 4/16 patients with hypertonic sphincter (25%) ($p=0.45$).

Conclusions: There was a predominance of normotonic anal sphincter in children with refractory chronic functional constipation. More than 1/3 of the patients were able to expulse the balloon during the defecation test, regardless of anal sphincter resting pressure, and about half of them presented anism not related to the anal sphincter tonus. The anorectal manometry is appropriate to study physiol-

ogy and evacuatory dynamic, and can be useful to guide the treatment.

Key-words: constipation; manometry; child.

RESUMO

Objetivo: Descrever os resultados da manometria anorretal dos pacientes com constipação intestinal crônica funcional (CICF), refratária aos tratamentos habituais, quanto ao tônus esfínteriano, presença do reflexo inibitório retoanal (RIRA), capacidade de expulsão do balão intrarretal e comportamento durante manobra evacuatória.

Métodos: Estudo retrospectivo por meio de análise de prontuários médicos entre janeiro de 2003 e junho de 2007, avaliando-se 31 pacientes ambulatoriais com CICF refratária a tratamentos convencionais por manometria anorretal com cateter de quatro canais (processador MPX 816 e *software* Proctomaster 5.0, Dynamed).

Resultados: Dos 31 pacientes, 24 (77%) eram do sexo masculino. A idade média foi $8,9 \pm 2,6$ anos. A média de pressão máxima fisiológica do canal anal foi 82 ± 38 mmHg. Dos 31 pacientes, 15 (48%) apresentaram esfínter anal normotônico e 16 (52%), hipertônico. O RIRA esteve presente em todos os casos. A prova de expulsão do balão intrarretal foi positiva em 12/31 pacientes, sendo 4/12 com esfínter normotônico e 8/12 com esfínter hipertônico ($p=0,22$). Anismo foi detectado em 6/15 pacientes com esfínter normotônico (40%) e em 4/16 com esfínter hipertônico (25%) ($p=0,45$).

Conclusões: Houve predomínio de esfínter anal normotônico nos casos de CICF refratária. Pouco mais de 1/3

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dos pacientes conseguiram a expulsão do balão durante a prova evacuatória, independentemente da tonicidade do esfíncter anal e cerca de metade dos pacientes apresentavam anismo sem relação com a tonicidade do esfíncter anal. A manometria anorretal é apropriada para o estudo da fisiologia e dinâmica evacuatória e pode ser útil para orientar o tratamento.

Palavras-chave: constipação intestinal; manometria; criança.

Introduction

Chronic functional constipation (CFC) is the most common presentation of childhood constipation. It is characterized by persistent difficulty in defecation and most commonly is caused by painful bowel movements, resulting in withholding of feces by the child, without objective evidence of an organic disorder. Stool-withholding behavior can lead to prolonged fecal stasis in the colon, with reabsorption of fluids and increase in the size and consistency of the stools⁽¹⁾, resulting in increased rectal compliance and decreased rectal sensitivity. The patients may also have abdominal distension, pain or discomfort. Approximately 25% of appointments at a pediatric gastroenterology clinic and 3% of general pediatric outpatient visits⁽²⁾ are related to chronic constipation. Thus, this condition is an important factor for psychosocial disorders among children and their families.

The management of chronic constipation depends on two critical factors to achieving good results: the child's adherence to conventional treatment (adequate dietary fiber intake, fecal disimpaction, behavioral and drug therapy) and parents' understanding of the importance of integrate actions. Nevertheless, long-term follow-up studies have shown that approximately 50% of children require treatment for long periods of time⁽³⁾, showing no significant improvement with

the use of conventional therapeutic measures. In these cases, it is necessary to establish a differential diagnosis between functional and organic constipation, such as Hirschsprung's disease and other systemic diseases, neurological disorders, or malformations⁽⁴⁾.

Over the past few decades, anorectal manometric examination of constipated children has provided information on anorectal physiology, showing abnormalities such as decreased rectal sensitivity and paradoxical contraction of the anal sphincter during a defecation attempt⁽⁵⁾. One goal of the manometric examination, within the investigation of constipated patients, is to demonstrate the presence of a rectosphincteric inhibitory reflex. The observation of internal sphincter relaxation in response to distension of the rectal wall rules out the possibility of aganglionosis and makes the diagnosis of some of the various forms of neuronal dysplasia of the myenteric plexus unlikely⁽⁶⁾. This knowledge has also enabled the development of techniques for adjuvant treatment, such as biofeedback training.

The objective of this study was to describe the anorectal manometric profile of patients with CFC refractory to conventional treatment, regarding anal sphincter tone, presence of rectoanal inhibitory reflex (RAIR), ability to expel the intrarectal balloon, and behavior during the defecation maneuver.

Method

This is a retrospective study based on the review of medical records. All patients met Rome II criteria for constipation (Chart 1). Patients with pathologic causes of constipation, such as Hirschsprung's disease, neuropathies, hypothyroidism, spina bifida, or who were using drugs that influenced gastrointestinal function were excluded.

All children were under regular ambulatory monitoring and initially underwent conventional treatment, receiving guidance on diet, including adequate dietary fiber and fluid intake, fecal disimpaction with glycerin or phosphate enemas every four days if no spontaneous evacuation occurred, toilet training, and prescription of laxatives such as magnesium hydroxide (1 mL/kg/day). The children were reassessed in the outpatient clinic every two months, with evidence of lack of response to treatment, i.e., the investigation on the characteristics and frequency of bowel movements remained unchanged, although treatment

Chart 1 – Rome II criteria for the definition of constipation

Rome II criteria (children and infants)

Continuous or recurrent symptoms for at least 12 weeks in the preceding 12 months. Two or more of the following symptoms:

- straining, hard stools, sensation of incomplete evacuation, sensation of anorectal obstruction or blockade, and manual maneuvers to facilitate defecation in >25% of defecations;
- <three defecations per week.

adherence was positive. After a six-month period, those who failed therapy in at least two cycles of an eight-week treatment were considered refractory to treatment. Following the usual routine of the clinic, anorectal manometry was indicated for those patients.

Anorectal manometry was performed after preparation of the colon with phosphate enema, followed by digital rectal examination to ensure an empty rectal ampulla. A catheter with four radial and four axial channels 1 cm apart was used for perfusion. Pressures were measured by transducers in the perfusion line, perfused with distilled water at a rate of 0.56 mL/min/channel, and connected to MPX 816 processor and Proctomaster 5.0 Dynamed software. Rectal distension was produced with a distending balloon tied to the tip of the catheter. One examiner performed all manometry sessions, and no sedation was used.

When inserted into the anus, the catheter was drawn and pressure was recorded every centimeter, thus determining the length of the anal canal and the highest-pressure zone that characterizes the anal canal.

Anal sphincter resting pressure was measured, allowing the patient to calm down for three to five minutes.

Maximal squeeze pressure of the anal sphincter was determined by asking the child to squeeze the sphincter muscles as tight as possible for five times, considering the highest measurement. RAIR was tested by inflating the rectal balloon with 10 to 50 mL of air. After each 10-mL inflation of the balloon, the sphincter pressure was allowed to stabilize to resting pressure values. The reflex was defined to be normal when rectal distension produced a reduction in the anal sphincter pressure of at least 5 mmHg.

Defecation dynamics were studied by observing the ability to expel the intrarectal balloon. The latex balloon was inserted into the rectum and inflated with air to simulate the presence of feces. The patients were instructed to contract the abdominal muscles and to relax the pelvic floor during an attempt to expel the balloon. A normal response occurred when the anal sphincter pressure decreased during straining. A paradoxical anal response was characterized by increased anal sphincter pressure during the defecation maneuver. The examination was considered inconclusive in the presence of atypical tracing.

The study project was approved by the Research Ethics Committee of the School of Medicine of Santa Casa de São Paulo (FCMSCSP), Brazil. Statistical analysis was performed using Fisher's exact test.

Results

Thirty-one patients with difficult-to-manage CFC were included in the study, 24 were male and mean age was 8.9 ± 2.6 years. Patients were followed at FCMSCSP Pediatric Gastroenterology Outpatient Clinic from January 2003 to June 2007. Clinically, all study patients showed infrequent defecation and scybalous or hard stools for at least 3 months, despite conventional treatment, most frequently accompanied by abdominal pain (cramps), abdominal distension, pain during defecation, and presence of fecal soiling.

In the manometric examination, the assessment of anal sphincter resting pressure revealed that 15 (48%) patients had normotonic and 16 (52%) patients had hypertonic anal sphincter. Mean maximal squeeze pressure of the anal sphincter was 81.7 ± 38 mmHg. RAIR was present in all cases (Figure 1).

As for the balloon expulsion test, 12 (39%) patients were able to expel the intrarectal balloon. It was possible to correlate this finding with sphincter tonicity: 4/12 (33%) patients had normotonic and 8/12 (67%) patients had hypertonic anal sphincter, with no statistical difference between them ($p=0.22$).

During the defecation maneuver, anismus was observed in ten cases (32.2%), six of these children were among the 15 patients with normotonic anal sphincter and four were among the 16 patients with hypertonic sphincter ($p=0.45$).

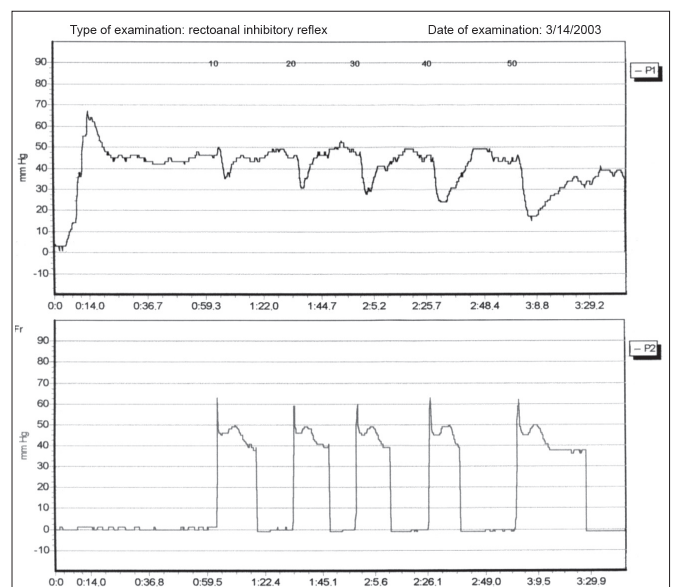


Figure 1 – Anorectal manometry tracing – rectoanal inhibitory reflex present.

Discussion

This study evaluated the anorectal manometric profile of constipated patients refractory to standard treatment. According to the recommendations of the North American Society for Pediatric Gastroenterology⁽¹⁾, whenever objective evidence of constipation refractory to treatment is observed, it is important to consider the possibility of Hirschsprung's disease. To rule this possibility out it is very important to perform anorectal manometry and rectal biopsy the only tests that can reliably exclude this diagnosis.

Several authors have reported changes in anorectal function. Some of these authors found normal anal resting tone, whereas others reported anal hypertonia or hypotonia in relation to the control group⁽⁶⁾. According to Benninga *et al*⁽⁷⁾, mean anal resting pressure in 22 healthy neonates was 40 mmHg (range: 7-65 mmHg). Loening-Baucke and Younoszai⁽⁸⁾ studied 116 constipated children, aged between two months and 18 years, demonstrating that anal resting pressure ranged from 25 to 72 mmHg in control children (mean 51±10 mmHg) and from 10 to 72 mmHg in constipated children (mean 39±12 mmHg). Anal resting pressure was, therefore, lower in constipated children. In the present study, normal anal sphincter tone was predominant. According to Loening-Baucke and Younoszai⁽⁸⁾, decreased motor activity in constipated children suggests that the internal anal sphincter is less active in constipated than in healthy children or in those with chronic rectal distension that can contribute to reduce anal resting pressure. Bigélli *et al*⁽⁹⁾ suggest that children with chronic constipation can have involuntary additional contraction of the anal sphincter, probably secondary to retention, to avoid the elimination of hard, large and consequently painful stools.

In the present study, maximal squeeze pressure of the anal canal, which is related to the external anal sphincter and puborectalis muscles, after conventional treatment, was 78±42 mmHg. These data are consistent with the study by Voordeckers *et al*⁽¹⁰⁾ of 91 children aged one month to 11 years with refractory CFC, in which mean maximal squeeze pressure was 73.7±34.4 mmHg. This measurement has limited clinical value; however, lower pressures and the inability to maintain contraction may be related to fecal incontinence.

RAIR was present in all our patients, thus excluding the diagnosis of Hirschsprung's disease and allowing a more reliable patient treatment and follow-up. The absence of RAIR

suggests Hirschsprung's disease, a situation that indicates a rectal biopsy for diagnostic confirmation. Steffen *et al*⁽¹¹⁾ stress the importance of repeating the anorectal manometry within one-two weeks in children less than one year of age with equivocal RAIR. Those authors studied a hundred anorectal manometry reports, and repeating the manometry was recommended in 18 cases. Of these, 17 patients had already undergone rectal biopsy for a diagnosis of Hirschsprung's disease and the second manometric screening showed the presence of RAIR in 16⁽¹¹⁾. These difficulties are described in the literature especially when dealing with infants, who feel insecure or even traumatized by frequent enemas, which results in poor compliance, thus hindering examination as well as its interpretation.

Children with long-standing functional constipation may show absence or a slight decrease in anal pressure (minimal relaxation of the rectosphincteric reflex), as observed in Hirschsprung's disease, whose probable etiology is chronic ischemia of the rectum⁽¹²⁾. In these cases, RAIR can be triggered after distension of the rectal ampulla due to volumes of air larger than the maximum tested⁽¹³⁾.

The literature reports paradoxical contraction in 36 to 78% of constipated children^(14,15). Corazziari *et al*⁽¹⁶⁾ observed paradoxical contraction in 30% of children examined, being significantly more common in children with fecal soiling.

Anismus occurs due to attempts to avoid painful bowel movements after a defecation stimulus, because of fears and pains experienced during defecation of hard stools early in life⁽¹⁷⁾. Anismus results from a lack of synergy between the functions of the levator ani muscle and external sphincter relaxation, leading to the contraction of both of these muscles during straining. In this study, during the defecation maneuver, anismus was observed in 32% of cases, unrelated to internal anal sphincter tonicity.

Thus, anal sphincter hypertonia was not prevalent among our cases of difficult-to-manage CFC. RAIR was present in all cases, ruling out Hirschsprung's disease. Just over one third of patients were able to expel the rectal balloon during the test, regardless of anal sphincter tonicity. There was no correlation between the presence of anismus and sphincter tone. The present data allow us to conclude that anorectal manometry is an adequate method to evaluate anorectal physiology and defecation dynamics in patients with chronic functional constipation. In addition, manometric examination can assist with the differential diagnosis of organic diseases and may also be useful to guide the therapeutic approach.

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