The Turkish Journal of Pediatrics 2018; 60: 547-553
DOI: 10.24953/turkjped.2018.05.012

Original

Frequency and findings of the acquired anorectal disease in the pediatric population with chronic constipation

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Received: 28th March 2017, Revised: 5th June 2017, 7th July 2017, 15th August 2017, Accepted: 30th October 2017

SUMMARY: Sanchez-Avila MT, Garcia-Valencia OA, Rivas-Calderon M, Morales-Garza LA, Jacobo-Velazquez P, Chavez-Caraza KL. Frequency and findings of the acquired anorectal disease in the pediatric population with chronic constipation. Turk J Pediatr 2018; 60: 547-553.

To determine the frequency of anorectal disease associated with chronic constipation in children we conducted an observational, descriptive, retrospective study. One hundred and sixty eight patients were evaluated by anorectal manometry over a 10-year period. The population was divided into organic constipation (OC) and functional constipation (FC) per ROME III criteria. Of those: 95 (56.54%) presented with anorectal disease. The age range was 1-17 (mean 5.3). Eighteen presented with OC and 77 with FC. Acute anal fissure was found in 45 patients (38 FC, 7 OC), chronic anal fissure in 38 (30 FC, 8 OC), perianal erythema in 26 (23 FC, 3 OC), scar tissue was found in 28 (24 FC, 4 OC), anal mucosa congestion in 27 (22 FC, 5 OC), hemorrhoidal disease in 26 (20 FC, 6 OC) and perianal venous dilatation in 21 (16 FC, 5 OC). Anal fissure, perianal erythema, and venous dilation occurred more frequently in patients with an anterior ectopic anus. An anal fissure was present in more than 80% of patients who had puborectalis muscle and external anal sphincter dysfunction.

We concluded that the frequency of acquired anorectal disease in children with chronic constipation is high; early diagnosis and treatment are priorities for the successful management of these patients.

Key words: constipation, children, anal fissure, functional constipation, hemorrhoids, rectal disease.

In the pediatric population, constipation has a prevalence up to 29%[1] and approximately 3% of the general pediatric consultations and 25%[2,3] of pediatric gastroenterology consultations are due to it. There is no statistical gender predominance.[1,4] This condition has an important negative impact both on the quality of life of the children and the economic aspect amounting to health care related costs of around $6.9 billion annually.[5]

In the pediatric population, chronic constipation can be divided into 2 categories: functional and secondary to an organic cause. The functional type is by far the most prevalent (90%) in the population outside the neonatal period and is usually multifactorial in origin.[2,6]

The lack of appropriate diagnosis and treatment of chronic constipation can result in the development of gastrointestinal complications such as fecal incontinence, fecal impaction, abdominal pain, anal itching, rectal bleeding, anal or perianal fissures, sentinel skin tag, hemorrhoids, rectal prolapse, anorexia or nausea.

There are other non-GI complications such as enuresis or urinary tract infections. Affected patients often present with complaints like difficulty defecating, large, hard stools, pain and/or bleeding when defecating and edema in the anal region.[7,8]

On the approach of the children has with constipation it is important to identify the

acquired anorectal diseases because if not treated early in the course can cause pain and discomfort to evacuate, perpetuating constipation and affecting significantly the quality of life of the pediatric patient.

The objective of this research is to determine the frequency of acquired anorectal disease associated with chronic constipation in children, to have evidence of the incidence of such diseases in the pediatric population.

Material and Methods

We reviewed the records of children referred to the Motility and Pelvic Floor Laboratory of the Hospital San Jose Tecnologico de Monterrey for chronic constipation over a period of 10 years (2004-2014).

Patients were sent for an anorectal manometry as they had been treated for functional constipation with conventional therapy based on osmotic laxatives and lifestyle changes without resolution or improvement of the problem for 1 month.

The population was divided into those with organic constipation, in which a cause or history of disease that accounted for the

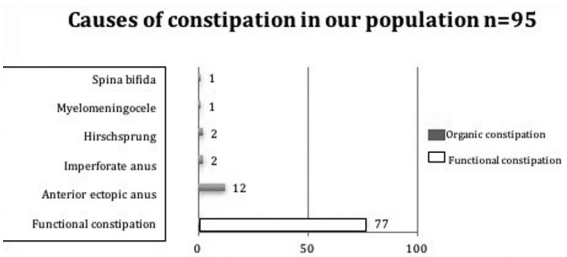

Fig. 1. Causes of constipation.

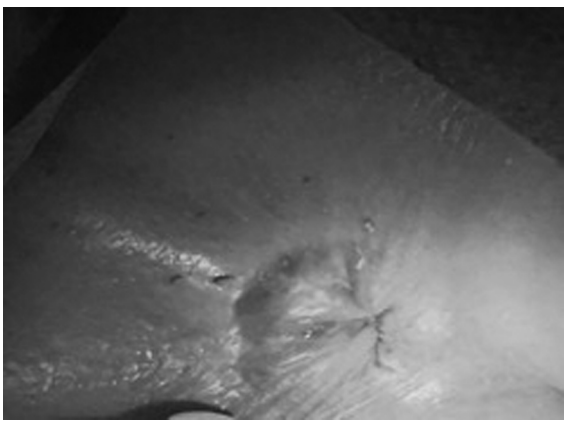

Fig. 2. Acute and chronic anal fissure: 10-year old-girl with constipation for 6 months associated with bleeding and rectal pain.

Fig 3. Acute anal fissure in a 4-year old boy with constipation for 5 months associated with pain.

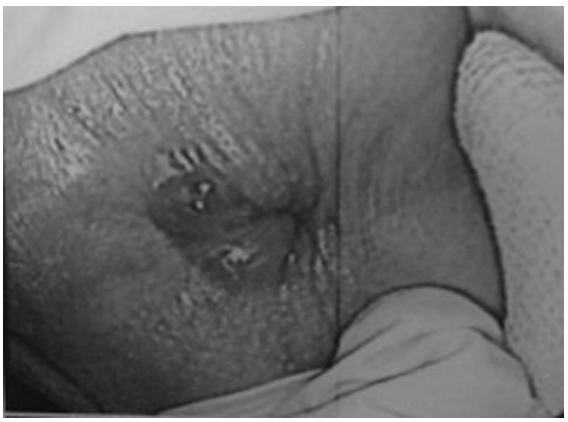

Fig. 4. Chronic anal fissure with external hemorrhoids.

Fig 5. External hemorrhoids in a 17-year-old male with constipation for 14 years associated with bleeding.

constipation was found, and those with functional constipation, in which had no constipation-associated cause. We used the ROME III criteria classification system as it is the most accepted and widely recognized as the only standardized symptom-based criteria for the diagnosis of functional chronic constipation (Table I).[9,10]

We searched retrospectively for sex, age, underlying disease, prior surgical procedures, history of biofeedback training, history of bleeding, difficulty or pain when defecating, large or hard stools and urinary disturbances.

As per protocol just prior to the manometric evaluation both groups underwent a physical examination directed towards finding manifestations of acquired anorectal pathology associated with chronic constipation. We searched for acute anal fissure, chronic anal fissure, scarring, hemorrhoidal disease, perianal erythema, anal mucosa congestion, perianal venous dilatation, perianal fistula or rectal prolapse.

The anorectal manometry was performed with a perfusion probe (Synectics®) with 5 sensors that have a distance between them of 1 cm in radial order and a balloon in the distal portion. The Camodi technique was used for patient evaluation, who was placed in left lateral decubitus position with flexed legs, the lubricated probe was then inserted into the anal region until it reached a length of 10 cm. The required values were obtained and the probe was removed with the "station pull-through" technique for the evaluation of the internal and external anal sphincter pressures. Those patients who did not cooperate received Chloral hydrate (70 mg/kg PO) as sedative. The parameters used were Internal Anal Sphincter (IAS) resting pressure, External Anal Sphincter (EAS) resting pressure, IAS squeeze pressure, EAS squeeze pressure, anal canal length, rectal ampulla pressure, Presence of Rectal Anal Inhibitory Reflex, Volume of conscious perception, volume of defecation desire, maximum tolerable volume and abdominopelvic coordination.

Table I. Diagnostic Criteria for Functional Constipation.[25]

Diagnostic Criteria For Functional Constipation In children >4 Years Of Age
Must include at least two of the following in a child with insufficient criteria for diagnosis of IBS
Two or fewer defecations in the toilet per week
At least one episode of fecal incontinence per week
History of retentive posturing or excessive volitional stool retention
History of painful or hard bowel movements
Presence of a large fecal mass in the rectum
History of large diameter stools which may obstruct the toilet
Criteria fulfilled at least once per week for at least two months prior to diagnosis.
Diagnostic Criteria For Functional Constipation In Children <4 Years Of Age
Must include at least two of the following for at least a month
Two or fewer defecations per week
At least one episode/week of incontinence after the acquisition of toileting skills
History of excessive stool retention
History of painful or hard bowel movements
Presence of a large fecal mass in the rectum
History of large diameter stools which may obstruct the toilet Accompanying symptoms may include irritability, decreased appetite and/or satiety. The accompanying symptoms disappear immediately following passage of large stool.

Table II. Associated Complications.

	Functional constipation (77)	Anterior ectopic anus (12)	Imperforate anus (2)	Myelomeningocele (1)	Spina bifida (1)	Hirschsprung (2)
Acute anal fissure	38	4		1	1	1
Chronic anal fissure	30	6	1	1		
Scar tissue	24	4				
Perianal erythema	23	1			1	1
Anal mucosa Congestion	22	5				
Hemorrhoidal Disease	20	3	1	1		1
Perianal venous dilation	16	5				
Rectal prolapse	5			1		
Perianal erosion	3		1			
Anal fistula	1					1
Fibrosis	1		2			

This study was approved by the Ethical Committee (13CEI19039139) and the National Bioethics Commission (CONBIOETICA19CEI00820130520).

Statistical analysis

Results are described as means and standard deviation for continuous variables and as frequencies and proportions for the discrete variables. MS Excel 2013 and SPSS20 were used for the statistical analysis. We calculated the odds ratio for the risk factors in the development of anorectal pathology.

Results

A total of 168 patient records were examined. Of those 95 presented with anorectal pathology, 43 being girls (45.26%) and 52 boys (54.72%). The age range of the studied population was 1-17 years old with a mean of 5.3 years.

Of those who had anorectal disease 18 presented with constipation secondary to an organic cause (18.94%) finding a history of ectopic anus in 12 (66.66%), Hirschsprung 's disease in 2 (11.11%), history of imperforate anus in 2 (11.11%), 1 patient with myelomeningocele (5.55%) and 1 with spina bifida (5.55%) (Fig. 1). On the other hand, 77 patients had functional constipation (81.05%). Regarding anorectal disease 62 patients (65.26%) presented with 2 or more coexisting lesions. Acute anal fissure was found in 45 patients (38 FC, 7 OC), chronic anal fissure in 38 (30 FC, 8 OC), perianal erythema in 26 (23 FC, 3 OC), scar tissue in 28 (24 FC, 4 OC), mucous anal congestion in 27 (22 FC, 5 OC), hemorrhoidal disease in 26 (20 FC, 6 OC) and perianal venous dilatation in 21 (16 FC, 5 OC). Other injuries found were rectal prolapse in 6 (5 FC 1 OC), perianal erosion in 4 (3 FC 1 OC) perianal fibrosis in 3 (1 FC, 2 OC) and anal fistula in 2 (1 FC 1 OC). (Table II).

Patients with anterior ectopic anus presented more frequently with anal fissure (83.33%), perianal erythema (41.66%) and perianal venous dilation (33.33%).

Relevant manometric findings associated with acquired anorectal pathology were found in 43 patients: 26 presented with puborectalis muscle dysfunction, 8 with external sphincter dysfunction and 9 with combined dysfunction. The most common anorectal finding in these patients was anal fissure presenting in 84.6% of the patients with puborectalis muscle dysfunction, 87.5% on those with external sphincter dysfunction and all of those with combined dysfunction.

Table III. Risk factors for the development of Anorectal Pathology

	Variable	Odds ratio	Confidence interval	P value
Constipation				
	Functional	Referent		
	Organic	1.01	(0.37-2.74)	0.98
Sex				
	Female	Referent		
	Male	0.76	(0.40-1.46)	0.42
Age (yr)				
	≥9	Referent		
	<9	2.14	(1.11-4.13)	0.02
Treatment with biofeedback				
	No	Referent		
	Yes	3.05	(0.87-10.64)	0.08

We tested the importance of the population characteristics such as type of constipation (functional vs organic) sex, age and history of biofeedback treatment as risk factors for the development of anorectal pathology. Only age accounting for significant statistical difference (p value 0.02). (Table III).

Discussion

The acquired anorectal disease has been extensively studied in adults, so its diagnosis and treatment are well established in this population. However, this is not true in the pediatric population as this disease is poorly described and thought to be uncommon in this age group. This lack of information often leads pediatricians to overlook its manifestations and therefore not detect them and treat them in properly fashion.

Regarding the findings in this study, there was no difference between the proportion of males and females evaluated, which is consistent with the results of Van den Berg's systematic review[1] and described by Medeiros et al.[4] and Rajindrajith et al.[11] who found no significant association between gender and constipation.

Anal fissures are defined as the laceration of the anal mucocutaneous junction. It is an acquired lesion of unknown etiology likely secondary to the forceful passage of a hard stool related with constipation.[12] They can be classified into acute (<6 weeks) or chronic (6 weeks or more). The former usually appear as a simple laceration (linear appearance) in the anus (Fig. 2 and 3) while the latter have a triangular appearance and can be accompanied by edema and fibrotic tissue (Fig. 4) tend to be more difficult to treat and can be associated with an external elevation and/or extra tissue within the anal canal referred as hypertrophic papillae. They mostly occur along the posterior midline and the presence outside this position should lead to thinking of other conditions or comorbidities. In some cases, as previously described, they can be the cause and not the consequence of constipation. The percentage of patients with a fissure in our study was much higher (88%) than reported in previous studies such as Dehghani et al.[9] (7.2%) and Aydogdu et al.[13] (26.9%). Likewise, perianal erythema was more prevalent (29.8%) when compared to Dehghani et al.[9] (13.1%).

History of surgically corrected imperforate anus (usually by posterior sagittal ano rectoplasty) has been associated with a slightly higher percentage of long-term constipation (30.7%) [14] being the most common disorder found in these patients.[15] In our study 50% of patients with a history of imperforate anus presented with chronic anal fissure, however being a small sample (n=2) we recommend further investigation.

Anterior ectopic anus refers to an anorectal malformation observed only in women and characterized by an essentially normal appearing anus in an abnormally anterior position of

the midpoint between the fourchette and the coccyx.[16] The anal canal is primarily located within the voluntary sphincter complex differentiating it from the perineal fistula. Kyrklund et al.[17] found a prevalence of constipation of 36% in patients with this disease as compared with the 13% found of their control cases. In our study, the high prevalence of anal fissure (83.33%), perianal erythema (41.66%) and perianal venous dilatation (33.33%) in these cases suggests a possible association between the two conditions.

In patients with pelvic floor dysfunction, the ability to relax the muscles during defecation is altered resulting in a paradoxical (inappropriate) contraction. This has been recognized as a major cause of chronic functional constipation presenting in up to 50% of these patients.[18] The etiology of this pattern is not entirely clear yet it has been associated with a brain-intestinal axis dysfunction and/or acquired behavioral failure secondary to pain, stress or trauma.[19] The latter being the basis for anorectal biofeedback and psychotherapy as treatment, both demonstrating positive changes in rates of motility and symptomatology.[20] The excessive tension exerted against an unrelaxed, closed anal canal can contribute to the development of other anorectal pathology with complications such as anal fissures, hemorrhoids, rectocele, solitary rectal ulcer or coccidynia.[19] This is consistent with our manometry findings suggestive of a relationship between the puborectalis muscle and external anal sphincter dysfunction with the presence of anal fissures being higher when combined dysfunction is present than either one alone.

Hemorrhoids are extremely rare in children with an increasing prevalence in adolescents and adults,[21] however there is insufficient data regarding the incidence in the pediatric population.[22] They are more common in children with portal hypertension with an estimate between 4 y 32% [23] yet these are rarely symptomatic.[24,25] When present symptoms may include hemorrhage, prolapse, itching and pain; (Fig. 5) the stools may be bloody or parents may observe a bulge or dilated venous plexus at the anal orifice. The latter being more typical in children with chronic constipation.[21] In our study the prevalence of children with hemorrhoids was common (27%), being higher in the patients with organic vs. functional constipation (33.33% vs. 25.97%). Even as this is not regarded as a problem of childhood, in our study we found a mean age of 8 years old for the patients with this presentation, this would suggest a strong correlation between chronic constipation and early onset of hemorrhoidal disease.

We acknowledge in the limitations of our study that the higher prevalence of anorectal disease found in our patients when compared with most studies could be explained because our study was conducted in patients with a long-standing pathology and who had already been treated by another specialist without improvement in the symptoms and therefore were referred for a more thorough evaluation, representing this as a selection bias.

In the present study, a high prevalence of acquired anorectal disease in the children with chronic functional constipation was found unlike previously reported in the literature. Thus, we recommend an adequate management and follow up in the assessment of the children with a history of constipation in order to avoid the development of acquired anorectal lesions as they could contribute to pain and discomfort to evacuate perpetuating the behavioral pain-retention cycle and its very important negative impact on the quality of life of the pediatric patients and the family economics.

When assessing children with constipation and a history of pain or bleeding a thorough examination of the perianal region is warranted to identify acquired anorectal lesions such as hemorrhoids and anal fissures that can be perpetuating the symptomatology. Among the causes of constipation anterior ectopic anus should be considered, as it is strongly associated with the anorectal pathology. The frequency of acquired anorectal disease in our studied population is high; early diagnosis and treatment are priorities for the successful management of these patients.

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

For this type of study formal consent is not required.

Acknowledgements

We would like to thank Dr. Gonzalez-Camid, Estrella, Dr. Uscanga-Vicarte, Victor, Dr. Urquidi-Rivera, Martha and Dr. Cura-Esquivel, Idalia for helping us review each of their cases.

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