

ROLE OF ANORECTAL MANOMETRY TO IMPROVE THE RESULTS OF BIOPSY IN DIAGNOSIS OF CHRONIC CONSTIPATION

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ABSTRACT

Objective: Most pediatric constipation cases may be treated with an adequate diet and moderate use of laxatives and enemas. A significant proportion of patients, however, does not show improvement with these therapeutic measures. In these cases, it is necessary to establish the differential diagnosis between functional and structural constipation. The objective of this study was to compare the result of two methods of evaluation, manometry before biopsy and biopsy alone.

Methodology: This cross sectional study was carried out in the children with chronic constipation that were referred to Mofid Children Hospital and underwent rectal manometry from 2002-2006. Chi-square test was used for the analysis.

Results: From 347 patients who underwent anorectal manometry, 134 patients had evidence that indicated neurologic abnormality and hirschprung disease. From these patients, based on parental agreement, biopsy were obtained from 69 patients. Histopathologic study was performed on 69 samples, and 57(82.6%) specimens had evidence of neuroanatomic abnormality as a cause of the disease. From the 423 biopsy that were obtained from rectal wall without previous anorectal manometry, only 125(29.55%) patients had pathological evidence of structural abnormalities due to Hirschprung disease. From all the biopsy (480), only 57(11.87%) patients had anorectal manometry. Based on there results of anorectal manometry they underwent biopsy.

Conclusion: Performing anorectal manometry as a first step to approach constipation could increase specificity of biopsy in contrast to performing biopsy without anorectal manometry. Due to some limitation, we did not obtain biopsy from patients whose manometry was normal.

KEYWORDS: Anorectal manometry, Hirschprung's disease, Constipation.

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INTRODUCTION

Most pediatric constipation cases may be treated with an adequate diet and moderate use of laxatives and enemas. A significant proportion of patients, however, do not show improvement with these therapeutic measures.¹ In these cases, it is necessary to establish the differential diagnosis between functional and structural constipation. The most difficult differential diagnosis is that of ultra-short segment Hirschsprung's disease. This is due to similarity of clinical manifestations and due to the fact that in the ultra-short varieties, barium enema does not reveal a spastic

aganglionic segment and intestinal dilatation that is identifiable in the most frequent form of Hirschprung's disease (congenital aganglionic megacolon). Assessments of anorectal manometry for diagnosis of Hirschprung's disease with ultrashort segment performed in both neonates and older children, have identified sensitivity varying from 75 to 100% and a specificity varying from 95% to 99%.^{2,3} False-positive results (incorrect diagnoses of aganglionosis) have been more frequently found in neonates and attributed to the immaturity of the myenteric nervous system.⁴ There were three false-positive and no false-negative results for manometry, corresponding to a sensitivity of 100%, specificity of 94%, positive-predictive value of 77% and negative-predictive value of 100%.⁵ The accuracy for the diagnosis of Hirschprung disease by manometry varies with the age of the patients.^{6,7} Anorectal manometry seem to be more accurate in older children,^{8,9} for whom recent studies have suggested an accuracy of 90 to 100%. Most authors report that the accuracy in neonates is lower.^{2-4,6,8} A recent study of 59 patients (2-90 days) reported a sensitivity, specificity, positive predictive value and negative predictive value of anorectal manometry for the diagnosis of Hirschprung disease 0.91, 0.56, 0.84 and 0.92 respectively.⁷ In other studies, the overall accuracy, sensitivity, specificity, and positive and negative predictive value were 90%, 0.79, 0.97, 0.94, and 0.88, respectively, whereas in neonates, it was 90%, 0.86, 1.1, 0.75, and in infants, it was 94%, 0.9, 1.1, and 0.89, respectively.² Thus, a functional constipation diagnosis based solely on a single radiological examination where the "Transition zone" has not been visualized may delay the solution of a condition for which surgery is required. Anorectal manometry is useful for identifying ultra-short segment Hirschsprung's disease. One of the aim of the manometric examination in the investigation of constipated patients is to demonstrate the presence of retrosphincteric inhibitory reflex. The observation of internal sphincter relaxation in response to the distention of the rectal wall eliminates the possibility of aganglionosis and makes the

diagnosis of the variable forms of neuronal dysphasia of the myenteric plexuses improbable. The objective of this study was to evaluate the power of anorectal manometry to identify the structural etiology in the investigation of patients with chronic constipation of childhood. In this study we compared two methods of approach (anorectal manometry + biopsy, biopsy alone) to investigation of Hirschsprung disease.

PATIENTS AND METHODS

This cross sectional study was carried out in the children with chronic constipation that were referred to Mofid Children Hospital and who underwent rectal manometry and rectal biopsy from 2002-2006. Chi-square test was used for the analysis. Due to some limitation, we did not obtain biopsy from patients whose manometry was normal, as some surgeons believe that there is no indication for manometry for the patients before rectal biopsy. This could be one of the limitations of this study.

RESULTS

From 347 patients who underwent anorectal manometry, 134 patients had evidence that indicated neurologic abnormality and hirschsprung disease. From these patients, biopsies were obtained from 69 patients. Histopathologic studies were performed, and 57 (57/69=82.60%) specimens had evidence of neuroanatomic abnormality as a cause of the disease. From the 423 biopsies that were obtained from rectal wall without previous anorectal manometry, only 125 (29.55%) patients had pathological evidence of structural abnormalities due to Hirschsprung disease. From all the biopsies (423+57), only 57 (11.87%) patients had anorectal manometry performed and based on their results of anorectal manometry they underwent biopsy. In fact, from 423 patients who underwent biopsy due to history and physical examination only 125 (29.55%) showed hirschsprung disease. As mentioned above, there is a significant differences between the results of biopsy with or without manometry ($p < 0.0005$).

DISCUSSION

The primary indication of anorectal manometry is to rule out Hirschsprung's disease in infants and children, although anorectal manometry is also used in the evaluation of fecal incontinence.¹⁰ The finding of sphincteric relaxation (RAIR) excludes Hirschsprung disease, particularly in older children⁶ and avoids the performance of more invasive testing such as a biopsy.¹¹ On the other hand, the lack of sphincteric relaxation strongly indicates the presence of Hirschsprung disease, but a confirmatory biopsy is necessary.^{8,12} Gil-Vernet JM reported that it is necessary to perform all three diagnostic procedures (Radiology/transition zone, anorectal manometry/absence of anal inhibitory reflex, anal suction biopsy/AChE study) in all patients with symptomatology given that not one test has the capacity to provide the accurate diagnosis alone. In one report of 26 children with Hirschsprung disease, three patients initially had normal manometries and when repeated later because of the persistence of symptoms, did not show a RAIR, and on histology subsequently these patients were diagnosed as of Hirschsprung disease.^{13,14} Da-Costa-Pinto EAL indicated that anorectal manometry is a diagnostic technique with very small possibility of error in differential diagnosis between constipation of a chronic functional nature and that which is secondary to ganglion cell abnormalities.⁵ Da-Costa-Pinto EAL has recommended that manometry should be included in the investigation of patients who do not respond satisfactorily to the initial clinical treatment. Given its high sensitivity, we believe that rectal biopsy is unnecessary in cases in which manometry demonstrates the presence of rectosphincteric inhibitory reflex.⁵

As mentioned above we recommend anorectal manometry in addition to rectal biopsy in the evaluation of non functional constipation.

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