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Constipation With Acquired Megarectum in Children With Autism

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ABSTRACT. *Objective.* Recent evidence suggests that autistic children may have significant gastrointestinal symptoms. Although constipation occurs in 2% to 5% of healthy children, its clinical diagnosis is often difficult in children with behavioral disorders. We thus aimed to assess the prevalence of fecal loading in autistic children with gastrointestinal symptoms and to identify possible predictors of constipation.

Methods. We studied abdominal radiographs of 103 autistic children (87 boys) who were referred for gastroenterological assessment, in comparison with 29 control radiographs from children who were referred to the emergency department, most with abdominal pain. Radiographs were scored independently, in blinded manner, by 4 pediatric gastroenterologists and a radiologist. The severity of constipation was determined using a validated index. Details of stool habit, abdominal pain, dietary history, and laxative use were obtained from case notes.

Results. The incidence of constipation in the control subjects with abdominal pain was higher than reported for normal children. Despite this, moderate or severe constipation was more frequent in the autistic group than in the control subjects (36% vs 10%). Analysis of rectosigmoid loading showed more striking differences (54.4% of autistic children had moderate/severe loading or acquired megarectum compared with 24.1% of control subjects). Multivariate regression analysis showed consumption of milk to be the strongest predictor of constipation in the autistic group, whereas stool frequency, gluten consumption, soiling, and abdominal pain were not predictive of constipation.

Conclusions. Constipation is a frequent finding in children with gastrointestinal symptoms and autism, particularly in the rectosigmoid colon, often with acquired megarectum. The absence of any correlation between the clinical history and the degree of fecal impaction in autistic children confirms the importance of an abdominal radiograph in the assessment of their degree of constipation. *Pediatrics* 2003;112:939–942; *autism, neurodisability, constipation, child, abdominal radiographs.*

Pervasive developmental disorders are a group of disorders of which childhood autism remains the prototype.¹ Typical features are qualitative impairments in social interaction and communication and a restricted repertoire of interests, activities, and behaviors, with onset in early

childhood. In classical autism, abnormal or impaired development is obvious from before the age of 3 years. Autistic spectrum disorders range from classical autism to the high-functioning Asperger's syndrome, in which language milestones are not delayed and cognitive skills may be above average.^{1,2} Hyperkinetic and attention-deficit disorders, obsessive-compulsive disorders, developmental disorders of motor function, and learning problems may coexist in children with autism.³

Gastrointestinal diseases are more common in children with a neurologic disability.⁴ In children with regressive autism, there have been recent reports of unexpected intestinal inflammation, with both low-grade colitis^{5,6} and duodenitis with reduced disaccharides.⁷ Immunohistochemical studies have suggested an immune response targeted at the gut epithelium, with a possible autoimmune cause.^{8,9} However, these findings have been largely restricted to those with a clear history of developmental regression, and the proportion of children within the overall autistic spectrum who have gastrointestinal problems remains unknown. An early report from Melmed et al¹⁰ of a large unselected population of autistic children, however, identified approximately half with gastroenterologic problems when formal assessment was made.

The combination of neurologic and developmental problems in children with autism makes toileting problems very common. In a questionnaire survey of 100 parents of autistic patients, among the most commonly reported problems were urinary incontinence and constipation.¹¹ Our own assessment of autistic children who were referred to us with gastrointestinal symptoms suggested that constipation was a particularly common finding and that effective management of this frequently provided global benefit.¹² Similar behavioral improvement was also noted by Horvath et al.⁷ This study thus aimed to define the prevalence of constipation in children with gastrointestinal symptoms and autism and to determine whether there are useful clinical predictors of the constipated autistic child.

METHODS

This was a retrospective study of children who had autism and were referred to a tertiary pediatric gastroenterology service for assessment of their gastrointestinal problems. All children who were referred to us during the course of this study had previously received a formal diagnosis by developmental pediatricians within the autistic spectrum and remained under appropriate neurodevelopmental follow-up. These diagnoses were made by pediatricians using criteria from the *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition* classification.¹ We reviewed

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all children who were younger than 18 years and referred with a diagnosis of an autistic spectrum disorder and gastrointestinal symptoms. Detailed histories were taken from all families, with particular emphasis on early development, dietary practices, recent gastrointestinal symptoms, and laxative use. All new referrals had a routine abdominal radiograph performed at presentation. All had received a diagnosis within the autistic spectrum by local developmental pediatricians or child psychiatrists. The median age of presentation was 5.9 years (range: 2.2–18.7).

To assess the discriminating power of the radiologic methodology, we used 29 consecutive, nonautistic, children who had required abdominal radiograph in the pediatric emergency department during the same time period as control subjects. The median age of presentation in this group was 8.5 years (M:F = 58.8%:41.2%). Of the total, 70.6% presented with abdominal pain, 17.6% had an radiograph to exclude suspected spinal injuries, 11.8% presented with foreign body ingestion, and 5.9% presented with vomiting.

Abdominal radiographs were scored independently, in a blinded manner, by 2 pediatric gastroenterologists, 1 senior radiologist, and 2 senior trainees in pediatric gastroenterology. The severity of constipation was recorded using a previously reported system, in which a score of ≥ 9 of 15 was defined as significant constipation.^{13,14} Each abdominal radiograph was scored in 3 segments, with a maximum of 5 points for severe fecal loading with bowel dilation. In view of the likely variability, we decided to use the mean scores of all 5 observers to determine whether a child was defined as “constipated” (≥ 9 of 15) or “not constipated” (< 9 of 15).

Clinical details that had been systematically recorded on all autistic children were extracted from the records. These included age at presentation, stool frequency, soiling, rectal bleeding, alternating diarrhea and constipation, abdominal pain, dietary history, and laxative use.

All statistical analyses were done using statistical package SPSS version 10.1. Analyses of significance were done using Wilcoxon rank sum test and χ^2 tests where appropriate.

RESULTS

Clinical records and abdominal radiographs were available for 103 children with autistic spectrum referred between 1996 and 2001. Of these, 87 (84.5%) were boys and 16 were girls. Ninety-four cases were of core autism, and 9 had Asperger’s syndrome. Only 13.6% of all of the autistic children referred were on treatment for constipation, with only 1 in 3 of the “constipated” children being on any laxatives. On referral, 1 child, subsequently defined as “constipated,” was being treated with Loperamide for recurrent soiling. Representative radiographs are shown in Fig 1.

Analysis of radiograph scoring confirmed systematic variation in the scoring system, as previously reported,¹³ with 2 assessors scoring consistently lower for all patients. Using the defined criteria, a diagnosis of constipation on the basis of mean overall score ≥ 9 of 15 was made in 37 of 103 autistic children. In the control group, only 3 of 29 were received a diagnosis of constipation. Thus, a significant difference in the prevalence of constipation was identified between the control subjects and the autistic group (10% vs 36% respectively; $P = .011$). When loading was analyzed in the sigmoid and rectal segment (identified as a score of ≥ 3), the differences were even more striking. Here, 54.4% of autistic children showed significant fecal impaction compared with 24.1% of control subjects ($P < .01$).

The stool frequency of the autistic children was not suggestive of a high incidence of impaction (overall mean 17.8 bowel motions per week; range: 1–50, with radiologically constipated children having mean 14.9

motions per week compared with nonconstipated 19.5 per week; not significant). On direct questioning, however, many of the parents reported that their children demonstrated avoidance posturing, such as hunching forward on the toilet or defecating while standing. Soiling was present in 25 (43.1%) of 58 of the overall autistic children, with a higher incidence (14 of 24 [56%]) of the radiologically constipated population. However, in 43% of cases, the parents had not reported soiling on direct questioning. There were no significant differences in clinical details between the “constipated” and “nonconstipated” children with autism, although the former were receiving more laxatives at referral ($P = .018$). The frequency of symptoms are given in Table 1.

Many different therapies have been used in the treatment of autism and its associated complications, with dietary exclusions being 1 of the more common practices.^{15–18} Of all of the autistic children in this study, 63 (61.2%) of 103 were on an exclusion diet (Table 1): 37 children were on combined dairy- and gluten-free diets, 16 were on dairy-free alone, and 10 were on a gluten-free diet alone. There were a very small number of children who were also excluding individual food items such as yeast, sugar, chocolate, egg, and soya. However, there was no significant difference in the use of exclusion diets between “constipated” and “nonconstipated” children (45.5% vs 46.2%, respectively). Univariate logistic regression found the use of laxatives at the time of referral to be the only significant predictor of the diagnosis of constipation (Table 2). However, a trend toward significance was found for both soiling and consumption of dairy products ($P < .09$). Multivariate regression analysis showed that the intake of dairy protein but not of gluten was a significant predictor of constipation in the total population ($P < .01$).

DISCUSSION

Constipation is a relatively common problem in children,¹⁹ with estimates of prevalence in otherwise normal children between 0.3% and 8%.²⁰ Whereas anecdotal reports and parental questionnaires have suggested that constipation may be common in autistic children, this is the first study reporting a definitive assessment of fecal loading in children with autism using abdominal radiograph as a diagnostic tool.²¹ This study underlines the difficulty in making a clinical diagnosis of chronic constipation in any child. History alone may only allow a diagnosis of constipation to be made in approximately 50% of children.²⁰ Making a diagnosis is even harder in children with autism. In our study, only one third of the referred children who finally received a diagnosis of being “constipated” were receiving any laxatives.

Clinical tools may be inadequate in making a diagnosis of constipation, and abdominal radiograph may be the only way to confirm the diagnosis in cases of clinical doubt.²² In view of the increased difficulties in clinical ascertainment of constipation in children with autism, we recommend the use of plain abdominal radiographs in initial assessment of possible gastrointestinal abnormality. In our study,



Fig 1. Representative abdominal radiographs from 3 children with autism. None had previously received a diagnosed of constipation and thus were not receiving treatment. A, Large fecal mass seen within distended rectum (acquired megarectum), with loading through sigmoid and transverse colon. This male child had a history of apparent abdominal pain, without overt constipation but showing avoidance posturing. B, Impaction with large acquired megarectum, without substantial colonic loading, in girl with intermittent diarrhea and soiling. C, Impacted rectal mass, filling most of the pelvis, in male child with history of episodic diarrhea.

soiling was the only symptom likely to be predictive (close to significance) in making a diagnosis of constipation.

Reporting and interpreting abdominal radiographs remains an art. We have confirmed the findings of Leech et al,¹³ that even when mutually agreed radiographic diagnostic criteria for constipation are used, there may be consistent differences in the observers' reports. Despite interobserver variability,

TABLE 1. Clinical Details of 103 Children With Gastrointestinal Symptoms and Autism

| Variable | Present | Absent |
|--|------------|------------|
| Dietary exclusion (<i>n</i> = 103) | 63 (61.2%) | 40 (38.8%) |
| Alternating diarrhea/constipation (<i>n</i> = 81) | 44 (54.3%) | 37 (45.7%) |
| Abdominal pain (<i>n</i> = 88) | 47 (53.4%) | 41 (46.6%) |
| Soiling (<i>n</i> = 58) | 25 (43.1%) | 33 (56.9%) |
| Blood in stool (<i>n</i> = 88) | 14 (15.9%) | 74 (84.1%) |

TABLE 2. Predictors of a Diagnosis of Constipation

| | Univariate Logistic Regression (P) |
|---------------------------------------|------------------------------------|
| Laxatives | .023 |
| Soiling | .087 |
| Milk exclusion | .0831 |
| Wheat exclusion | .154 |
| Dietary exclusion | .27 |
| Abdominal pain | .98 |
| Blood in stool | .453 |
| Alternating diarrhea and constipation | .2974 |

n = minimum 50 patients. On multivariate logistic regression, use of milk was found to be a significant predictor of the diagnosis of constipation, *P* < .01.

there was positive overlap in making a diagnosis in >70% of cases. Intraobserver variability seems to remain consistent,¹³ making abdominal radiographs the most effective means of diagnosing constipation in autistic children.

Constipation is present more often in children with neurodevelopmental disabilities. This may be because of the inherent problems encountered in toilet training. The same holds true for autistic children. However, the findings of colonic and duodenal immunopathology in children with regressive autism^{8,9} may suggest a more specific disturbance of gastrointestinal motility. Recent murine evidence suggests a direct link between mucosal immunopathology and antigen-induced dysmotility,^{23–25} with products of T_H2 cells and eosinophils directly affecting neural cell function within the gut. In humans, cow-milk protein has been particularly implicated in “allergic dysmotility,” with a pattern of constipation characterized by distal impaction as a result of antigen-induced external sphincter spasm.^{26,27} It thus is notable that cow-milk consumption was the only significant predictor of constipation on multivariate analysis. Whether the dominant cause of this distal constipation is indeed immune mediated and antigen driven or potentially caused by poor handling of dietary morphines,⁵ it is notable that behavioral improvement may occur if it is detected and effectively treated.¹² Our own practice in this regard is to soften initially with liquid paraffin before disimpaction with stimulant laxatives,^{8,12} as suggested for nonautistic children with similar impaction.²⁸

CONCLUSIONS

Constipation is a frequent and significant problem in children with autism and continues to remain underrecognized. It is particularly marked within the rectosigmoid colon, often in association with acquired megarectum. The absence of any correlation between the clinical history and the degree of fecal impaction in autistic children confirms the importance of an abdominal radiograph in the assessment of the degree of constipation. Effective treatment may potentially alleviate behavioral problems and disturbed sleep pattern in autistic children.

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