

Predictive value of internal anal sphincter thickness for diagnosis of rectal intussusception in patients with solitary rectal ulcer syndrome

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Background: The aim of this study was to determine any association between a thickened internal anal sphincter (IAS) on anal endosonography and high-grade rectal intussusception on evacuation proctography in patients with solitary rectal ulcer syndrome.

Methods: Anal endosonography was performed in 20 patients with solitary rectal ulcer syndrome and IAS thickness defined as normal or abnormal depending on age. Sphincter thickness was compared with the presence or absence of high-grade intussusception on subsequent evacuation proctography to determine any relationship between the two.

Results: Thirteen patients had an abnormally thick IAS, two of whom were unable to evacuate. Of the remaining 11 patients, ten showed high-grade intussusception (positive predictive value 91 per cent). Only three of seven patients with a normal IAS had high-grade intussusception (negative predictive value 57 per cent). Patients with a thick IAS were significantly more likely to have proctographic evidence of high-grade intussusception ($P = 0.047$).

Conclusion: Sonographic findings of a thick IAS are highly predictive for high-grade rectal intussusception in patients with solitary rectal ulcer syndrome.

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Introduction

Solitary rectal ulcer syndrome (SRUS) is an uncommon condition affecting young adults and is characterized clinically by rectal bleeding, mucus, tenesmus and difficult evacuation¹. Proctoscopy reveals a spectrum of mucosal changes ranging from reddening, through polypoid thickening, to frank ulceration². Abnormalities are anterior in most cases and biopsy characteristically shows collagenous replacement of the lamina propria and fibromuscular replacement of the mucosa¹. Full-thickness rectal wall examination also reveals that the circular muscle is significantly thickened and condensed into bundles, features resembling high-pressure sphincter tissue³. Circular rectal wall muscle is continuous with the internal anal sphincter (IAS), which is also abnormally thickened in patients with SRUS when examined by anal endosonography⁴. Muscular rectal wall thickening may be a feature of recurrent rectal prolapse^{3,5}, which affects approximately 75 per cent of patients with SRUS; prolapse is confined to the rectum (intussusception) in approximately two-thirds, with

the rest having complete rectal prolapse⁶. However, an association between intussusception and an abnormally thick IAS has not been proven in patients with SRUS. Sonographic and proctographic examinations of patients with proven SRUS have been reviewed in order to establish the relationship, if any, between IAS thickening and rectal prolapse.

Patients and methods

All patients with proven SRUS in whom anal endosonography and evacuation proctography had been performed were identified using both retrospective review and prospective data collection. No patient had undergone surgery for intussusception. Twenty patients, nine males and 11 females, of median age 33 (range 15–73) years, were identified. SRUS was confirmed by characteristic histological findings following rectal biopsy in all patients¹.

Anal endosonography was always performed first, using a Brüel and Kjær Medical 3535 scanner with an 1850 endoscopic probe (B&K Medical, Gentofte, Denmark). The first

ten patients were scanned with a 7-MHz transducer and the remainder using an improved 10-MHz transducer (type 6004) with superior spatial resolution. The transducer was covered with a hard plastic sonolucent cone with an outer diameter of 1.7 cm. The 10-MHz transducer has a focal range of 5–45 mm, an axial resolution of less than 0.05 mm and a lateral resolution of 0.5–1 mm. The assembly was filled with degassed water for acoustic coupling, and covered with a lubricated condom for insertion. Patients were scanned in either the left lateral or prone position⁷, and the endoprobe was inserted gently into the anus to the level of the anorectal verge. During slow withdrawal of the probe, images were taken at the following levels: high, mid and low anal canal. The high anal canal was defined as that level midway between the inferior border of the puborectalis and complete formation of the external sphincter ring anteriorly. Mid-canal level was defined by completion of the external sphincter ring anteriorly in combination with maximum IAS thickness. The low canal was defined as that level immediately caudal to the termination of the IAS, and comprised the subcutaneous external sphincter. Images were taken at two different magnifications and printed on laser film for subsequent analysis.

Evacuation proctography was subsequently performed using a simple standard technique⁶. Two glycerine suppositories were administered rectally and retained for 20 min; patients were then invited to empty the rectum in the toilet. With the patient in the left lateral position, 120 ml barium paste (E-Z paste[®]; E-Z-Em, Westbury, New York, USA) was instilled into the rectum using a bladder syringe. The patient was then seated upright on a specially designed commode and asked to empty the rectum as rapidly and completely as possible during lateral digital fluoroscopy with images acquired at one frame per second. Filming was intermittent if evacuation was delayed, prolonged, or both, so that the maximum screening time was kept to less than 60 s.

The anal endosonographic images were scanned electronically and images at mid-anal canal level were selected and stored on a personal computer. Using the centimetre scale reproduced on the sonographic image, an image analysis programme was calibrated for measurement (Scion Image 3b; Scion Corporation, Frederick, Maryland, USA), and measurements of IAS thickness were taken at 3 and 9 o'clock using the electronic callipers. All measurements were repeated three times by a single observer, and the mean was determined. Measurement reproducibility using this technique has been shown to be good⁸.

Internal sphincter thickness was defined as normal or thicker than expected for age by a single observer who used data derived from a sonographic study of 150 asymptomatic female volunteers whose bowel habit had been character-

ized as normal using validated questionnaires⁹. Subjects from that study were divided into seven age groups, and a thick IAS for the purposes of the present study was defined as the mean IAS thickness (at mid-anal canal level) plus two standard deviations. The following seven age groupings and upper limit of normal IAS thickness were defined: age 20 years or less, 2.6 mm; age 21–30 years, 3.2 mm; age 31–40 years, 3.6 mm; age 41–50 years, 3.6 mm; age 51–60 years, 3.4 mm; age 61–70 years, 3.7 mm; age 71 or over, 3.5 mm. Nine, 62, 43, 11, seven, 12 and six individuals, respectively, were used to define the seven age groups.

A second observer, unaware of the sonographic findings, reviewed the proctographic examinations. The presence or absence of rectal intussusception was noted and, if present, classified as high grade if the intussuscepting mucosa entered the anal canal or had a thickness greater than 3 mm. Low-grade intrarectal intussusception was considered a normal finding¹⁰.

The relationship between the presence of high-grade intrarectal intussusception and IAS thickness was determined and categorical data were examined using Fisher's exact test.

Results

Median IAS thickness was 3.9 (range 2.9–8.6) mm. Taking age into account, 13 of the 20 patients studied had an abnormally thick IAS on endosonography (*Fig. 1*). Two of the 20 subjects were unable to evacuate any contrast, possibly masking intussusception, and were excluded from

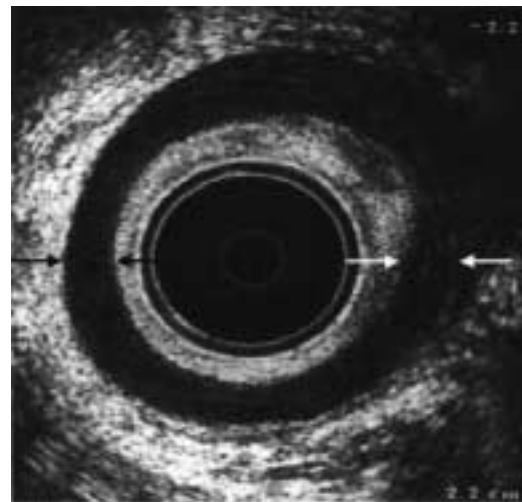
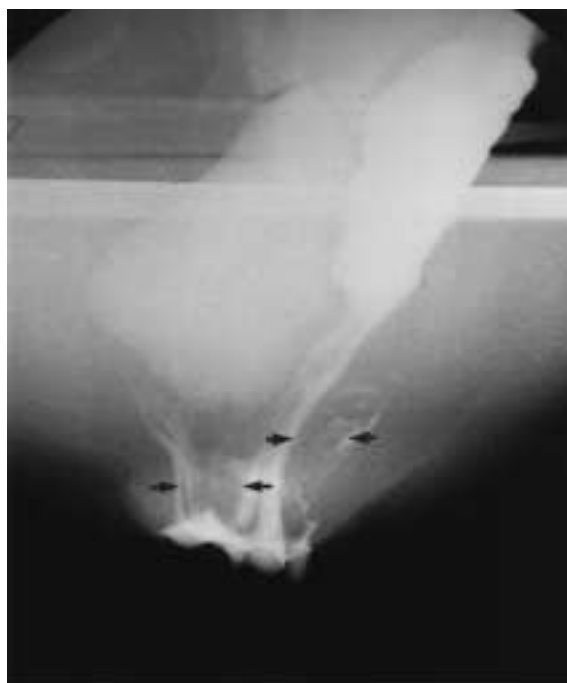


Fig. 1 Endoanal ultrasonogram at mid-canal level in a 54-year-old woman with solitary rectal ulcer syndrome. There is gross thickening of the internal anal sphincter (between arrows), which measured 7.3 mm



a During evacuation



b End of evacuation

Fig. 2 Evacuating proctography: lateral views during evacuation. **a** The intussuscepting mucosa descends from mid-rectal level, seen initially as a filling defect arising from the posterior rectal wall (arrow). **b** The intussuscepting mucosa (arrows) enters the anal canal at the end of evacuation, diagnosing high-grade intussusception

the subsequent analysis. Both of these had an abnormally thick IAS. Of the remaining 11 patients with a thick IAS, ten showed features of high-grade intussusception on evacuation proctography (Fig. 2). Overall, there were proctographic findings of high-grade rectal intussusception in 13 of the 18 patients who were able to evacuate, only three of whom had a normal IAS. Patients with a thick IAS had significantly more rectal intussusception than those whose sphincter was judged normal (ten of 11 *versus* three of seven patients; $P = 0.047$, Fisher's exact test).

Excluding the two patients who were unable to evacuate, the finding of a thick IAS on anal endosonography had a positive predictive value of 91 per cent for proctographic features of high-grade intussusception. Of the seven patients with a sonographically normal IAS, three had proctographic evidence of high-grade rectal intussusception, giving a negative predictive value of 57 per cent.

Discussion

The purpose of this study was to identify the relationship, if any, between IAS thickness and rectal intussusception in a group of patients with histologically proven SRUS. Morphological internal sphincter abnormalities in SRUS were first documented in a study of 21 patients that showed the IAS, subepithelium and external anal sphincter were significantly thickened in these patients compared with age- and sex-matched controls⁴. The study used a 7-MHz transducer that was unable reliably to differentiate the longitudinal muscle and intersphincteric plane from the external sphincter, so that these two structures were incorporated into external sphincter measurements. Longitudinal muscle thickening in SRUS may help to explain the surprising finding of apparent external sphincter thickening in that study³.

The reason for IAS thickening is unclear, but histological studies of rectal circular muscle, from which the IAS is derived, clearly demonstrate abnormal thickening in patients with SRUS³. Histologically, this is thought to be due to collagen deposition³. It has been suggested that mechanical stress due to intussusception and repetitive straining are the cause of this thickening⁵. Alternatively, it is postulated that the constant tenesmus typical of SRUS induces secondary hypertrophy of the muscularis propria¹¹. Rectal wall thickening may be demonstrated *in vivo* using rectal ultrasonography; a study of 15 patients with SRUS found that the rectal muscularis propria was thicker than the normal upper limit of 2 mm in all cases⁵. Furthermore, this thickening was associated with rectal wall intussusception⁵. The present authors postulated that rectal intussusception would similarly be associated with IAS thickening.

This study has confirmed earlier observations that the IAS is abnormally thick in patients with SRUS. Thirteen of the 20 patients had a thicker IAS than would be expected for their age. Indeed, the thickest sphincter (8.6 mm) was found in the youngest patient, a boy aged 15 years. Furthermore, the study has also shown a significant association between this thickening and rectal intussusception, with a positive predictive value of 91 per cent. No patient in this group had complete rectal prolapse. Although rectal muscularis propria thickening and IAS thickening both seem to be associated with rectal intussusception, it is not known whether thickening of the latter is similarly due to excess collagen deposition; sphincter biopsy would be required to prove this. Furthermore, it remains unknown whether the IAS changes are secondary to mechanical strain, either directly on the sphincter or due to intussusception occurring in the rectal wall above. Indeed, a primary sphincter abnormality cannot be excluded. Alternatively, both intussusception and IAS thickening may be independently associated with some other, as yet unexplained, feature of the syndrome.

Data from a prospective sonographic study of asymptomatic women were used to define the upper limit of normal IAS thickness⁹. All had normal bowel habit confirmed using validated questionnaires. Limits were stratified for age because it is known that the normal IAS thickens with age¹². A possible criticism is that only female control values were available. However, the authors considered that this was acceptable because previous sonographic studies have shown no difference between male and female IAS thickness^{13,14}. Another potential criticism relates to the use of two different transducers, 7 and 10 MHz, although this seems unlikely to affect measurements of IAS thickness adversely because this muscle is so well delineated on ultrasonography, even when using the older transducer. In contrast, the external sphincter and longitudinal muscle are much better defined using the newer probe.

The present findings suggest that anal endosonography could be used as an alternative to evacuation proctography for the diagnosis of rectal intussusception in SRUS, thus perhaps avoiding irradiation in these patients, many of whom are women in their childbearing years. To date, evacuation proctography has been the 'gold standard' for diagnosis of intussusception in SRUS. A proctographic study⁶ of 53 patients with proven SRUS found high-grade internal rectal intussusception in 36, and some studies have found an incidence of up to 94 per cent¹⁵. Approximately 50 per cent of patients with SRUS also have muscular incoordination of the pelvic floor, which can be diagnosed reliably by means of evacuation proctography when there is incomplete or prolonged emptying¹⁶; two of the patients in the present study failed to evacuate. When both prolapse

and disorders of evacuation are considered, evacuation proctography demonstrates abnormalities in 75 per cent of patients with SRUS⁶. Rectal wall changes in SRUS are also demonstrable on barium enema, but the lack of functional information with respect to intussusception or muscular incoordination limits the usefulness of this technique¹⁷.

Demonstration of intussusception and any coexisting functional disorder of evacuation is central to current management strategy for SRUS. Biofeedback may have a role in restoration of normal pelvic floor function, especially where behavioural disorder is dominant¹⁸. Conversely, surgery is likely to be helpful only when there is significant intussusception or complete rectal prolapse and, even then, a consensus is emerging that it should be reserved for selected patients with severe symptoms only. A proctographic study of 23 patients with SRUS found that rectopexy successfully treated prolapse in all but one patient, but 16 remained symptomatic¹⁹. Most authors would agree that the aetiology of SRUS is multifactorial and that management is consequently difficult.

In summary, this study has demonstrated that excessive IAS thickening in patients with SRUS is significantly associated with high-grade rectal intussusception. Endosonography may be used to identify patients suffering from intussusception, but the cause of these alterations in IAS morphology remains unknown.

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