Solitary Rectal Ulcer Syndrome (SRUS): Spectrum of findings on MR Defecography

ABSTRACT

Objective: Radiological findings in solitary rectal ulcer syndrome (SRUS) are well described for evacuation proctography (EP) but sparse for magnetic resonance defecography (MRD). In order to rectify this, we describe the spectrum of MRD findings in patients with histologically proven SRUS.

Materials and methods: MRD from twenty-eight patients (18 female; 10 males) with histologically confirmed SRUS were identified. MRD employed a 1.5T magnet and a standardized technique with the rectal lumen filled with gel and imaged sagittally in the supine position, before, during and after attempted rectal evacuation. A single radiologist observer with 5 years' experience in pelvic floor imaging made anatomical and functional measurements.

Results: Sixteen patients (10 female) demonstrated internal rectal intussusception and 3 patients (11%) demonstrated complete external rectal prolapse. Anterior rectoceles were noted in 12 female patients (43%). Associated anterior and middle compartment weakness (evidenced by excessive descent) was observed in 16 patients (57%). Enterocoeles were detected in 5 patients (18%) and peritoneocoele in 5 patients (18%). None had sigmoidocoele. Sixteen patients (57%) demonstrated delayed voiding and 13 patients (46%) incomplete voiding, suggesting daefecatory dyssynergia.

Conclusion: MRD can identify and grade both rectal intussusception and dyssynergia in SRUS, and also depict associated anterior and/or middle compartment descent. Distinction between structural and functional findings has important therapeutic implications.

Key words

Rectal diseases

Ulcer

Magnetic Resonance Imaging

Defecography

Pelvic floor

Key points

- MRD can identify and grade both rectal intussusception and dyssynergia in patients with SRUS.
- MRD is an acceptable substitute to evacuation proctography in assessing anorectal dysfunctions when attempting to avoid ionising radiation.
- SRUS influences the pelvic floor globally. MRD depicts concomitant anterior and/or middle compartment prolapse.

Abbreviations

SRUS Solitary rectal ulcer syndrome

MRD Magnetic resonance Defecography

EP Evacuation proctography

PACS Picture archiving and communication system

PCL Pubococcygeal line

ARJ Anorectal junction

INTRODUCTION

In 1829 Cruveilhier first described solitary rectal ulcer syndrome (SRUS) as a chronic and benign rectal lesion **(1).** SRUS has an approximate incidence of 1 in 100,000 per year, with a small female predominance **(2).** Typically, patients present with functional symptoms of obstructed defecation, often needing to digitate the perineum or vagina when evacuating, and spending excessive amounts of time at stool. These functional symptoms are often accompanied by rectal bleeding, mucus discharge, external prolapse, and, rarely, fecal incontinence **(3)**. Biopsy helps exclude endoscopic mimics such as inflammatory bowel disease, ischaemic colitis, and malignancy **(4).** The prime role for imaging is to detect structural abnormalities such as rectal intussusception and rectal prolapse, and functional abnormalities, namely defaecatory dyssynergia (incomplete emptying) **(5)**. While this has been achieved using evacuating proctography (EP), magnetic resonance defecography (MRD) is now employed widely to evaluate pelvic floor dysfunction **(6)**. However, data regarding MRD features of SRUS are sparse **(7).** It cannot simply be assumed that EP findings will translate directly to MRD, since the two techniques have different temporal resolution, patient positioning, and the voided enema differs. Emphasizing this, a recent healthy volunteer study of MRD found differences when compared to historical EP data **(8)**. To rectify this, we reviewed the spectrum of

MRD findings in patients with histologically proven SRUS to assess the prevalence and severity of both structural and functional abnormalities.

MATERIALS AND METHODS

Ethical committee approval was waived due to the retrospective nature of the study and the fact that data presented would be anonymized.

Patients:

Patients were accrued from two institutions; BLINDED Hospital (institution 1) and BLINDED Hospital (institution 2). For institution 1, we searched the hospital pathology database for patients with histopathological diagnosis of SRUS between 2014 and 2019, providing 180 discrete cases. These were then cross-referenced against the hospital PACS to identify individuals who had also undergone MRD; any who underwent pelvic floor surgery prior to imaging were excluded. Nineteen patients were identified, with average temporal separation between histopathological diagnosis and MRD of 10 months (range 1 to 42 months). A single researcher (BLINDED) used the hospital Electronic Patient Record (EPR) system (Epic Hyperspace, Epic Systems Corporation) to extract age, sex, and symptoms. For institution 2, patients presenting from 2016 to 2019 with pathologically-proven SRUS were referred prospectively to the department of radiology within one month of their diagnosis for dynamic MRD; 9 patients were identified. Again, using the local EPR (PaxeraUltima, PaxeraHealth Corporation), age, sex, and symptoms were retrieved by the same researcher (BLINDED).

Imaging technique:

At institution 1, patients were scanned on one of four 1.5T units (Avanto and Symphony, Siemens). An optional phosphate enema (Fleet Enema, Fleet Laboratories, Prestige Consumer Healthcare) was administered immediately prior to the scan and patients asked to empty their rectum. After voiding the enema (if used), 150-180 ml of ultrasonographic gel was instilled into the rectum using bladder syringes. The patient was then positioned supine on the scanner and a surface phased-array coil placed over the pelvis. After initial planning sequences, an evacuation phase was acquired in the sagittal plane using a T2 Tru-FISP sequence (repetition time ms/echo time ms TR/TE 4.25/2.13 ms, FOV 300 mm, slice thickness 5.0 mm).

At institution 2, patients were scanned on one of two 1.5T units (Achieva and Intera, Philips Medical System) in the supine position using a pelvic phased-array coil. The evacuation phase was acquired in the sagittal position using a balanced-fast-field-echo (BFFE) sequence (TR/TE 5.0/1.6 ms, FOV 300 mm, slice thickness 3.0 mm) after rectal filling with 200-250 ml of ultrasonographic gel using bladder syringes. No oral or intravenous contrast agent was administered at either institution.

Image analysis:

MR image analysis was performed by a single radiologist (BLINDED) with 5-years' experience interpreting pelvic floor MR images. All measurements from institution 1 were recorded using a Picture Archiving and Communication Systems (PACS) workstation using Vue PACS software version 12.1.5.7014 (Carestream Health). All measurements from institution 2 were made using Philips MR extended workspace (software version 2013).

The pubococcygeal line (PCL) was used as the reference line from which to measure pelvic organ descent during evacuation. The PCL was drawn on the mid sagittal plane from the inferior aspect of the pubic symphysis to the last coccygeal joint. Posterior compartment descent was measured from the anorectal junction (ARJ) to the PCL; the ARJ was defined as the superior margin of the puborectalis sling. For the middle compartment, measurements were taken from the PCL to the anterior cervical lip, or the vaginal vault in case of previous hysterectomy. Finally, for the anterior compartment, the distance between the most inferior aspect of the bladder base and the PCL was measured. The distance from each reference point was measured perpendicularly to the PCL both at rest and during evacuation, or alternatively at maximum strain in those patients who did not void (6). Descent of anterior and middle compartments to diagnose cystocele and uterine prolapse were graded according to the "Rule of 3"; grade I (1-3 cm), grade II (3-6 cm) and grade III (> 6 cm) below the PCL) **(6)**.

Rectoceles were measured relative to a straight line drawn along the anterior wall of the anal canal and extended cranially; a rectal bulge of greater than 2 cm anterior to this line was defined as a rectocele, graded as follows "small" (≤2 cm), "medium" (>2-4 cm) and "large" (>4 cm) **(6)**. Rectal intussusception and prolapse were classified according to the Oxford grading system as follows: (1) "high" recto-rectal intussusception; when rectal in-folding descends no lower than the proximal

limit of the rectocele; (2) “low” recto-rectal intussusception, when rectal in-folding descends into the rectocele; (3) “recto-anal” intussusception, when rectal in-folding reaches the anal canal; (4) “external” rectal prolapse, when the rectum protrudes beyond the anal canal **(9)**. Enterocoeles were defined when small bowel loops prolapsed into the recto-genital space beyond the PCL during evacuation; sigmoidocoeles were analogous but required sigmoid colon prolapse; and peritoneoceles were diagnosed when the peritoneum herniated into the rectovaginal space and beyond the PCL but with no associated bowel herniation **(10)**.

For functional measurements, daefecatory dyssynergia was diagnosed if rectal emptying was prolonged and/or voiding incomplete **(11).** Evacuation was semi-quantitatively analysed by measuring the cross-sectional area of rectal gel visible on the mid-sagittal plane cine image stack, (a) at rest, (b) 60 seconds after initiation of evacuation and, (c) at the end of the evacuation effort. The percentage area of gel voided at 60 seconds and at the end of evacuation were both calculated, as a surrogate for the rate and completeness of voiding, respectively. After 60 seconds, the voiding rate was considered “normal” if more than two-thirds of the gel enema was voided, “mildly impaired” if one- to two-thirds of the enema was voided and “markedly impaired” if less than a third was voided. At the end of the evacuation attempt, completeness of voiding was considered normal if more than two-thirds of the enema was voided, mildly impaired if the amount voided was between one- and two-thirds, and markedly impaired if less than a third had been voided **(11).**

All measurements were transferred to an Excel spreadsheet (Microsoft Corporation) and descriptive statistics calculated.

RESULTS

Twenty eight patients were included; 18 female and 10 male. The mean age at MRD was 47 years (range 17 to 89 years). Symptoms at the time of MRD included constipation (17 patients, 61%); faecal incontinence (6 patients, 21%); diarrhoea (3 patients; 11%); rectal bleeding (3 patients; 11%). One patient (4%) presented with rectal prolapse and one (4%) with uterine prolapse.

Structural abnormalities

The positions of the urinary bladder base, anterior cervical lip and anorectal junction relative to the pubococcygeal line (PCL) both at rest and during evacuation in patients who showed any descent are summarized in figure 1.

Posterior compartment findings (table 1):

Nineteen of 28 patients (68%) had posterior compartment abnormality during evacuation; 16 of 28 (57%) showed rectal intussusception, while 3 patients (11%) showed complete external rectal prolapse (Oxford grade 4). Of the 16 patients with intussusception, this was most commonly low recto-rectal (Oxford Grade 2, 12 patients). Two patients had recto-anal intussusception (Oxford grade 3), and two had only mild, high recto-rectal intussusception (Oxford grade 1). In six patients (21%) with rectal intussusception, the thickness of the intussuscepting fold ranged between 1.5 and 2 cm **[figure 2]**.

Mean ARJ descent below the PCL was 5.7 cm (range 1 to 10 cm) **[figure 1]**. An anterior rectocele was found in 12 patients (43%), all female. Small sized rectocele was noted in 3 patients (11%) and medium sized in 9 patients (32%), the mean size of rectoceles was 3.1 cm (range 2 to 4 cm).

Anterior and/or middle compartment findings (table 1):

Sixteen of 28 patients (57%) showed associated excessive anterior and/or middle compartment descent **[figures 2, 3 and 4]**. Cystocele was found in 14 patients (50%) with mean descent of 2.3 cm below PCL (range 1 to 4 cm) and uterine prolapse was noted in 7 patients (25%) with average descent of 2.6 cm below PCL (range 1.5 to 4 cm). Uterine prolapse, when observed, was always associated with cystocele. Enterocoele was seen in 5 patients (18 %) with average descent of 4.7 cm below the PCL (range 4 to 6 cm) and peritoneocele was noted in 5 patients with average descent of 3.2 cm below the PCL (range 2 to 4 cm). No patient had a sigmoidocoele.

Functional abnormalities

16 patients (57%) showed impaired voiding; 10 patients (36%) showed mildly delayed voiding (between 1/3 and 2/3 of the rectal gel evacuated by 1 minute) and 6 patients (21%) showed markedly delayed voiding. The completeness of rectal of voiding was impaired in 13 patients

(46%): mildly incomplete in 9 patients (32%), and markedly incomplete in 4 patients (14%) **[figure 5]**. 13 patients (46%) showed prolonged and incomplete voiding, while 3 patients (11%) showed prolonged yet rather complete voiding by the end of the examination **(table 2)**.

DISCUSSION

The aetiology of SRUS remains largely unknown. Most patients present before fifty years, suffering from disordered evacuation **(12)**. The name "solitary" is now known to be a misnomer since a single ulcer is present in only 20% of cases. Indeed, multiple ulcers are present in 40% and remaining cases display other morphologies, including polypoid masses and no ulcer at all **(13).** Macroscopically, there is mucosal erythema and the presence of ulceration may lead to an erroneous diagnosis of rectal carcinoma if the clinician is unaware of SRUS **(14)**. Biopsy shows smooth muscle proliferation, replacement of the lamina propria by fibroblasts (typically arranged at right angles to the muscularis mucosa), mucosal ulceration, crypt distortion, and thickening of the muscularis mucosa **(15)**. The word "syndrome" was added to reflect an association with additional anorectal and pelvic floor disorder, such as prolapse and dyssynergia.

In agreement with previous EP studies **(13) (16)**, that showed increased prevalence of internal or external rectal prolapse in patients with SRUS, 57% of our cohort showed associated internal rectal intussusception and a further 11% demonstrated complete extra-anal rectal prolapse, confirming the strong association between SRUS and rectal intussusception/prolapse. Moreover, it supports the hypothesis that the process of rectal mucosal invagination leads to increasing sensation of obstruction, thereby causing patients to strain more, ultimately causing ischemia of the rectal mucosa that is responsible for ulceration **(16)**.

Our findings also support the hypothesis that intussusception visible on EP is also identifiable by MRD, despite the latter's supine position, suggesting that MRD is an acceptable substitute when attempting to avoid ionising radiation.

Another advantage of MRD may be that it affords the opportunity to image concomitant anterior and/or middle compartment prolapse, which is only possible when EP is combined with bladder, vaginal, and bowel opacification **(17)**. MRD presents a more global picture of pelvic floor dysfunction than EP alone, most notably extensive pelvic floor weakness contingent on chronic

straining. Our observation that 57% of patients had associated anterior and/or middle compartment dysfunction implies that SRUS influences the pelvic floor globally, probably due to chronic excessive straining. Additionally, we found an associated enterocoele or peritneocoele in 36% of patients (with the two being equally split in frequency). Accordingly, many pelvic floor practitioners believe that a global pelvic floor assessment should be performed prior to surgical intervention, even for patients who appears to have single compartment symptoms, so that any potential therapy, including surgery, is not overly restrictive **(18).**

SRUS has been reported sometimes to present as a mass or polyposis on endoscopy and/or MRI **(19, 20)** where malignancy was ruled out by biopsy. Notably, one of the cases included in our study showed circumferential mural thickening of the rectum on the static T2 WIs with evidence of recto-rectal intussusception during the attempted evacuation phase **[figure 6].** This patient presented clinically with rectal bleeding and colonoscopy revealed hyperemic ulcerated rectal mucosa. Histopathological assessment showed ulcerations, vascular congestion, thickened muscularis propria and tendency of villous formation with no evidence of neoplasia in keeping with SRUS diagnosis. Due to the versatile appearances of SRUS on colonoscopy and MRI, it is important to note that SRUS is a spectrum that includes mucosal prolapse syndrome as well as colitis cystica profunda **(21)**. Hence, MR imaging in light of endoscopic findings and high clinical suspicion can exclude malignancies.

Quantitative evaluation of rectal evacuation identified abnormalities in the majority of patients with SRUS. Our observations are in keeping with prior EP findings that suggest that evacuation rate is more important than completeness when attempting to identity dyssynergia because evacuation can eventually be complete if straining is prolonged and forceful **(11**). Notably, seven patients with significant evacuation impairment showed no rectal prolapse, either internal or external, suggesting a predominant functional abnormality where two of them only showed associated anterior rectocele, likely due to the elevated intra-rectal pressure. It should be borne in mind that intussusception cannot usually be identified unless the rectal ampulla empties, since prolapse tends to occur at the end of evacuation; patients with severe dyssynergia who cannot void during MRD, will not demonstrate prolapse. It is likely that defecatory dyssynergia (previously termed “anismus”) is a prime etiological factor in SRUS, whereby chronic, forceful straining secondary to a functional disorder leads to elevated intra-rectal pressure that predisposes to

prolapse and mucosal ulceration as a result of chronic ischemia **(16)**. This also suggests that surgery for prolapse may not be beneficial unless the underlying functional disorder is treated via behavioral and/or biofeedback therapy. A prime role for imaging, therefore, is to determine the relative contribution of any structural and functional abnormalities to patients' symptoms.

Our study does have limitations. It is based on a mixed retrospective/prospective case review from two centres. There will have been a degree of spectrum bias since a proportionately small number patients from institution 1 with a histological diagnosis of SRUS were imaged, presumably referred because they had severe symptoms. At institution 2, all patients with a histological diagnosis of SRUS were referred routinely for MRD. Yet, there was no noteworthy prevalence/severity of abnormality differences between the two groups.

In summary, although patients with SRUS classically present with posterior compartment symptoms, MRD typically shows global pelvic floor dysfunction. Most patients have significant prolapse, either intussusception or external prolapse; or defecatory dyssynergia. MRD with its lack of ionising radiation and accurate depiction of both structural and functional abnormalities is an acceptable substitute for EP to assess patients with SRUS. MRD may help stratify which patients require bowel retraining and biofeedback, and which are ultimately likely to need surgical intervention.

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Figure legends

Figure 1: ladder plots showing the positions of the urinary bladder base (anterior compartment), anterior cervical lip (middle compartment) and anorectal junction (posterior compartment) relative to the pubococcygeal line (PCL) at rest and during evacuation in patients who showed any descent. Each dot represents a patient and overlapping dots (more than one patient) are color coded as per the legend.

Figure 2: MRD of 29 year old female patient complaining of faecal incontinence and diarrhoea with histopathological diagnosis of SRUS. BFFE during evacuation showing medium sized rectocele (*) with thick-fold low recto-rectal intussusception (white arrow). Associated grade I cystocele (C) and grade I uterine prolapse (U) are also noted.

Figure 3: MRD of 30 year old female patient with SRUS showing excessive descent of the ARJ below the PCL with medium sized anterior rectocele (*) associated with grade I cystocele (C) and grade I uterine prolapse (U).

Figure 4: MRD of 56 year old female patient with SRUS showing excessive recto-anal intussusception (*) associated with large enterocoele (E) evident by herniation of small bowel loops beyond the PCL into the Douglas pouch with grade II cystocele (C) and grade I uterine prolapse (U).

Figure 5: MRD of a 63 year old female patient with SRUS complaining of constipation, gel area measured (a) at rest, (b) after 60 seconds and (c) by the end of evacuation: revealing functional impairment of evacuation with markedly prolonged emptying with only 11% (< one third) of injected gel voided after 60 seconds and mildly incomplete emptying with only 40 % (between one third and two thirds) of rectal gel voided by the end of evacuation attempt. Note the medium sized anterior rectocele measuring 3.5 cm in size (*).

Figure 6: MRI of 38 year old female patient complaining of rectal bleeding where endoscopy showed ulcerated mucosa and histopathological evaluation excluded malignant features. (A) Static T2WI revealed circumferential mural thickening of the rectum (white arrow) suggestive of mucosal prolapse (B) BFFE during evacuation showing small bulging of the anterior rectal wall and further prolapse (grey arrow).