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Symptom distribution and anorectal physiology results in male patients with rectal intussusception and prolapse

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ABSTRACT

Background: Rectal intussusception and external rectal prolapse are uncommon proctographic findings in men reflecting the lack of studies investigating such patients. The aim of this study was to identify the demographic, clinical, and physiological characteristics of this population with a view to appreciate the mechanism of development of this condition.
Methods: All men, presenting with symptoms of constipation or fecal incontinence, who were diagnosed proctographically with recto-rectal intussusception (RRI)/recto-anal intussusception (RAI) or external rectal prolapse (ERP) between 1994 and 2007 at a tertiary academic colorectal unit were studied. Demographics, relevant comorbidities, distribution and symptom duration, and anorectal physiology results were analyzed retrospectively for each proctographic group and intergroup comparisons performed.
Results: Two hundred five men (median age 50 y; range, 13–86) including 155 (75.6%) without any relevant comorbidities were studied. A significant proportion of patients in all proctographic groups reported rectal evacuatory difficulty ([RRI, 46.4%], [RAI, 39.4%], [ERP, 44.8%]; $P = 0.38$,analysis of variance). Patients also reported a combination of fecal incontinence symptoms (e.g., urge, passive, postdefecatory leakage) that did not differ across the proctographic groups. Anorectal physiological parameters were within normal range and were not found to be statistically different between the proctographic groups with the exception of anal resting pressure, which was lowest in ERP patients (62 cm H_2O; range, 14–155) compared with patients with RRI (89 cm H_2O; range, 16–250; $P = 0.003$) and RAI (92 cm H_2O; range, 38–175; $P = 0.006$).

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Conclusions: Men with rectal intussusception and prolapse present with a combination of symptoms, predominantly defective rectal evacuation. Anorectal physiological assessment has failed to shed light into the mechanism of development of this condition and thus, the need for large observational studies incorporating integrated defecographic and manometric assessments of the evacuation process.

1. Introduction

Rectal intussusception (RI) is a partial or full-thickness invagination of the rectal wall that may be confined to the rectum (recto-rectal intussusception, RRI), descend into the anal canal (recto-anal intussusception, RAI) or extend beyond the anal verge (external rectal prolapse, ERP) [1]. RI is diagnosed during defecating proctography in patients presenting with evacuatory difficulties, but its clinical significance is occasionally uncertain in view of its presence in healthy asymptomatic volunteers [2–4]. The vast majority of affected patients are elderly females due to the damaging effect of pregnancy and menopause on the pelvic floor [5].

The condition is also seen, rather unexpectedly, in men but little is known about its etiology and distribution of symptoms in this subgroup of patients, with no studies in the literature discussing these factors in a male cohort [6]. Factors likely to be involved include loose sacral fixation of the rectum with an underlying connective tissue disorder, increased intra-abdominal pressure (secondary to obesity, chronic cough, and weight lifting), colonic dysmotility with chronic straining, and perineal hypermobility [5,7]. Traumatic impairment of the levator ani is a major risk factor in women but unlikely to be the predominant factor in men. More recently, it has been suggested that morphologic changes in the enteric ganglia may also contribute to the development of RI and ERP and complement existing etiological parameters [6]. Despite these theories, the natural history of the condition is not understood and it is currently debatable whether RI and ERP are part of the spectrum of a progressive disorder or two separate clinical entities.

The aim of this study was to explore the demographic, clinical, and anorectal physiological characteristics found in symptomatic men with RRI, RAI, and ERP to define this cohort and potentially understand the mechanism of development of the condition. A secondary objective was to establish whether a progressive relationship between the proctographic cohorts exists, that is, whether RRI leads to RAI which leads to ERP.

2. Methods

Patients referred to a gastrointestinal physiology unit within a period of 13 y (1994–2007) with fecal incontinence or rectal evacuatory disorder, underwent routine anorectal physiological evaluation including measurement of anal resting and squeeze pressures by using water-perfused anal canal pull-through manometry, assessment of sphincter morphology by endoanal ultrasound (7 or 10 MHz; B-K Medical, Berkshire, UK), measurement of pudendal nerve terminal motor latency (PNTML) by using the St Mark's electrode (Dantec Ltd, Bristol, UK), and estimation of rectal sensory thresholds (e.g., maximum tolerable volume, MTV) to air-filled balloon distension. All patients underwent evacuation proctography and colonic transit study [8,9].

The upper limit of normal PNTML was age stratified with pudendal neuropathy diagnosed if PNTML >2.3 ms for subjects aged <40 y and >2.5 ms for subjects aged >40 y. Rectal hyposensitivity was diagnosed when the MTV was >325 mL [10]. A prolonged colonic transit was diagnosed when the subject retained ≥20% of 50 markers 100 h after ingestion.

All men diagnosed proctographically with RRI (Shorvon grading 3–4; grade 3 is noncircumferential infolding ≥3 mm; grade 4 is circumferential infolding >3 mm that remains intrarectal), RAI (Shorvon grading 5–6; grade 5 is a circumferential infolding that impinges on the internal anal orifice; grade 6 is circumferential infolding that descents into the anal canal), or ERP (Shorvon grading 7) were identified from a prospectively recorded database. Patients with a proctographic diagnosis of RI graded 1–2 on the Shorvon classification were excluded from the analysis given the uncertainty surrounding the clinical significance of these findings commonly seen in asymptomatic volunteers [2,3,11]. Data were reviewed retrospectively and included patient demographics (i.e., age at presentation), type and duration of symptoms, past medical and surgical (e.g., anal, pelvic surgery) history, and anorectal physiological measurements including proctographic and colonic transit findings.

2.1. *Statistical analysis*

The anorectal physiological findings (e.g., sphincter integrity and anorectal pressures, percentage of patients with rectal hyposensitivity), duration and type of symptoms, percentage of patients with a rectocele, and percentage of patients with a positive transit study were analyzed for each proctographic group and intergroup comparisons performed.

Data analyses were performed using commercially available statistical analysis software (GraphPad Prism, Version 5; GraphPad Software, Inc, La Jolla, CA). Data normality was assessed using the De Agostino-Pearson omnibus normality test. Nonparametric data were compared using the Mann-Whitney *U*(MWU) test. Fisher exact test was used to compare levels of comorbidity between groups. One-way analysis of variance (ANOVA) in median age, presence of rectocele, positivity of transit study, duration and type of symptoms, and anorectal physiological parameters between the proctographic groups (RRI, RAI, and ERP) was calculated using the Kruskal-Wallis test with Dunn multiple comparison posttest. If the studied variable was statistically different between the proctographic groups, post-ANOVA comparison was performed for the relevant groups using unpaired *t*-test or

Table 1 – Characteristics of male patients with RRI, RAI, and ERP.

Males	RRI	RAI	ERP	P value
Number of patients	138	38	29	
Age at presentation; median (range), y	51 (16–86)	49 (13–74)	39 (16–78)	0.46
Rectocele (%)	4 (2.9%)	1 (2.6%)	1 (3.4%)	0.98
Positive transit study (%)	4 (2.9%)	1 (2.6%)	1 (3.4%)	0.98
Duration of symptoms (mo); median (range)	36 (4–552)	48 (3–480)	36 (1.5–480)	0.58
Sphincter morphology				
Intact IAS and EAS (%)	97 (70.3)	27 (71.0)	19 (65.5)	0.75
Defective IAS and EAS (%)	17 (12.3)	2 (5.3)	5 (17.2)	0.75
Defective IAS or EAS (%)	24 (17.4)	9 (23.7)	5 (17.2)	
Pressures (cm H_2O) median (range)				
Maximum resting	89 (16–250)	92 (38–175)	62 (14–155)	**0.001**
Maximum squeeze	121 (14–440)	119 (22–341)	107 (22–304)	0.71
MTV (mL)				
Median (range)	195 (50–420)	180 (90–340)	180 (85–340)	0.85
Hyposensate (%)	12 (8.7)	5 (13.2)	4 (13.7)	0.56
Pudendal neuropathy				
Unilateral (%)	23 (16.7)	11 (28.9)	6 (20.7)	0.39
Bilateral (%)	13 (9.4)	2 (5.3)	4 (13.8)	0.40

Bold value represent statistically significant.

MWU test as appropriate. A P value <0.05 was considered significant.

3. Results

3.1. Demographics

A total of 205 male patients with a median age of 50 y (range, 13–86) were included in the analysis. One hundred thirty-eight (67.3%) patients were diagnosed proctographically with RRI, 38 (18.6%) with RAI, and 29 (14.1%) with ERP. The cohort included 155 (75.6%) patients without any relevant comorbidities and 50 (24.4%) patients with a history of abdominal/pelvic surgery ($n = 7$ prostatectomy, $n = 2$ bowel resection), anal surgery ($n = 19$), anal trauma ($n = 10$), heavy lifting ($n = 2$), spinal surgery ($n = 3$), and trauma to spine/pelvis ($n = 7$). The demographic and physiological characteristics of patients according to their proctographic diagnosis are shown in Table 1. Table 2 illustrates the distribution of symptoms per proctographic group.

3.2. Age and intussusception

The median age of patients with ERP (39 y; range, 16–78) was lower compared with patients with RRI (51 y; range, 16–86) and RAI (49 y; range 13–74), but the difference was not statistically significant ($P = 0.46$, ANOVA).

3.3. Type and duration of symptoms

Symptom duration did not differ between the proctographic groups (Table 1, $P = 0.58$, ANOVA) ranging from 36 mo in patients with RRI/ERP to 48 mo in those with RAI. Difficulty with rectal evacuation was the predominant complaint in all proctographic groups and urge incontinence was the least common. The proportion of patients with incontinence symptoms or evacuatory disorder did not differ between the proctographic groups (Table 2).

3.4. Incidence of rectocele and slow transit constipation

There were six patients in total with a diagnosis of rectocele and all of them had previously undergone prostatectomy. One further patient who had undergone prostatectomy did not have any evidence of rectocele. The incidence of rectocele ranged from 2.6% (RAI) to 3.4% (ERP), but the difference was not statistically different across the proctographic groups (Table 1, $P = 0.98$, ANOVA). A similar pattern was seen in the incidence of colonic dysmotility with approximately 3% of patients with a positive transit study ($P = 0.98$, ANOVA).

3.5. Sphincter morphology and anorectal manometry

Approximately 70% of patients in all proctographic groups had intact internal and external anal sphincter (IAS and EAS). A significantly smaller proportion of patients had damaged IAS and EAS. In both cases, the differences between the proctographic groups were not significant (Table 1). Median squeeze

Table 2 – Quantitive assessment of symptoms in patients with RRI, RAI, and ERP.

Proportion of patients with	RRI	RAI	ERP	P value
Rectal evacuatory disorder	64 (46.4%)	15 (39.5%)	13 (44.8%)	0.38
Urge incontinence	16 (11.6%)	3 (7.9%)	4 (13.8%)	0.58
Passive incontinence	34 (24.6%)	10 (26.3%)	5 (17.2%)	0.60
Postdefecatory leakage	24 (17.4%)	10 (26.3%)	7 (24.2%)	0.46

pressure was also similar across the groups, but the difference in resting pressures was statistically significant ($P = 0.001$, ANOVA) between RRI/ERP ($P = 0.003$, MWU) and RAI/ERP ($P = 0.006$, MWU).

3.6. *Rectal sensation and pudendal neuropathy*

No statistical difference between the proctographic groups was found when comparing MTV, proportion of hyposensate patients, and proportion of patients with unilateral or bilateral pudendal neuropathy.

3.7. *Comorbidities*

Although the ERP group had a higher percentage of diabetes diagnosis than the RAI and RRI groups (15% *versus* 3% *versus* 4%), this difference was not significant ($P = 0.102$, Fisher exact test). Similar results were found for those patients with cardiovascular disease, including stroke (5% *versus* 6% *versus* 4% $P = 0.613$, Fisher exact test).

3.8. *Management*

Thirty-eight percent of the ERP group underwent operative surgery for their prolapse, including both perineal approach (Delormes, Express Procedure [12]) and abdominal approach (Rectopexy, Hartmanns) with 60% undergoing concurrent conservative management. Follow-up was for a median of 6 y with a 40% recurrence rate. Fifteen of the ERP group underwent purely conservative management. None of the RAI patients underwent operative surgery (all undertook conservative management); they had a median follow-up of 1 y. Sixteen percent of the RRI group underwent operative surgery (express procedure and rectopexy) with an 80% recurrence rate. Twenty-nine percent of RRI group underwent conservative management. Follow-up was for a median of 6 y.

4. Discussion

The literature is characterized by a distinct lack of studies investigating the etiopathogenesis and symptomatology of RI and prolapse in male patients. This is the first study, to our knowledge, that has attempted to define the characteristics of this patient group according to their proctographic diagnosis with a view to identify clinico-pathological parameters that may shed light into the development of this condition.

Patients appeared to be middle aged with the ERP cohort being the youngest and were symptomatic for a median period of 36–48 mo depending on proctographic diagnosis. There was no evidence of a progressive relationship between age and symptom duration with the proctographic diagnosis casting doubt on the hypothesis that ERP is the end point of a condition that starts with internal intussusception.

Previous studies in predominantly female patients with defecatory dysfunction reported diminished rectal motor activity and abnormal rectal sensitivity suggesting that altered sensorimotor behavior of the colon and rectum as a potential risk factor for the development of intussusception and prolapse [13,14]. In addition, histolopathological assessment of rectosigmoid specimens from young men with symptomatic rectal prolapse demonstrated submucosal hyperganglionosis with features similar to that seen in intestinal neuronal dysplasia [6]. The low incidence of a positive transit study and low proportion of patients with rectal hyposensation, however, in this cohort imply that a sensorimotor abnormality might not be the predominant etiological factor.

Impaired pelvic floor structure and function, commonly seen in postpartum women, is another potential factor that requires further discussion because there is little doubt about the importance of the levator ani muscle in providing support for the pelvic organs [15]. Extensive distensibility ("ballooning") of the levator ani with increased hiatal area and avulsion has been proposed as a significant etiological factor [5]. Most men in this study did not appear to have an underlying pelvic floor injury as demonstrated by the absence of relevant surgical history. However, the impact of other factors such as obesity, chronic cough, type of employment (e.g., manual labor), and smoking on the structure and function of the pelvic floor was not investigated in detail and this is a limitation of the study. Nevertheless, the presence of rectocele, a marker for pelvic floor injury, was low in our study and appeared to be a consequence of prostatectomy in accordance with the findings of a previous study [16]. Consequently, the evidence regarding the contribution of the pelvic floor structure and function in male patients is inconclusive and warrants further investigation using radiologic and histologic assessments.

Physiological assessment revealed that the anorectal pressures and MTV for all proctographic groups were within normal limits. Moreover, the vast majority of patients had an intact anal sphincter complex, rectal sensation and no unilateral/bilateral pudendal neuropathy suggesting that none of these parameters independently appears to play a crucial role in the pathogenesis of this condition. None of the physiological variables investigated were found to vary between the proctographic groups with the exception of the maximum resting pressure that was found to be the lowest in ERP patients compared with those with RRI/RAI. This finding may be explained by persistent activation of the anorectal reflex in those patients with ERP rather than as a result of injury to the anal sphincter complex because there was no statistical difference in the proportion of patients with defective IAS and EAS across the proctographic groups. Consequently, the development of RRI/RAI and ERP in men does not appear to be related to any of the studied variables.

Histologic and radiologic studies have implicated a high-pressure zone at the rectosigmoid junction as the starting point of the intussusception [17]. This suggests that a connective tissue disorder with loose sacral attachment of the rectum might be involved in the etiology of the condition. Furthermore, according to Stelzner [18], the condition results from the different size and morphology of the "fixed" lower rectum and "upper" mobile rectum. If this hypothesis is correct, then severely affected patients may develop ERP, whereas a milder form of the condition may result in RRI/RAI.

In conclusion, the mechanism of development of RI and prolapse in men remains uncertain and this has implications for the prevention and precise treatment of the condition. Limitations of our study are its retrospective nature and lack of a control arm. Randomized controlled trials and

observational studies of symptomatic and asymptomatic volunteers undergoing simultaneous defecographic and manometric assessments of the structure and function of the rectum and pelvic floor may be more informative and reveal not only the hidden mechanistic aspects leading to the development of RI and ERP but also the true contribution of disorders such as intussusception, rectocele, and anismus on defective rectal evacuation.

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