

## Solitary Rectal Ulcer Syndrome about 26 Cases and Literature Review

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### Abstract

**Introduction:** The solitary rectal ulcer syndrome (SRUS) is a rare disorder of the rectal wall and its course is chronic but always benign. The therapeutic management of SRUS remains a challenge and relies on a multidisciplinary management. The objective of our study is to describe the epidemiological, clinical, therapeutic and evolutionary characteristics of SRUS in a Moroccan series. **Materials and Methods:** This is a monocentric descriptive retrospective study including all patients with SRUS in the Department of Hepato-Gastro-Enterology and Proctology "Médecine B" of the Ibn Sina University Hospital, over a period from January 2000 to August 2021. **Results:** A total of 26 patients were included with a mean age of 45 years [15-70 years] and a sex ratio (M/F) of 0.6. The most frequent clinical symptomatology was rectal bleeding found in all patients (100%), followed by anorectal pain (84.6%), bloody mucus evacuations (73.1%), chronic constipation (38.5%) and rectal prolapse in 34.6% of the patients at the proctologic examination. The low endoscopic exploration had detected mainly a single ulceration with clean bottom (50%), multiple ulcerations (20%) and a pseudo-polypoid aspect (15.4%). Histological examination of the biopsies taken from the ulceration and the periphery confirmed the diagnosis by showing the typical histological aspect of SRUS. The majority of our patients (77%) underwent anorectal manometry, which revealed anorectal dyssynergia and sphincter tone abnormalities. The therapeutic management was based on medical treatment in all patients, defecation rehabilitation "biofeedback" (53.8%) and surgical approach was considered in 19.2% of cases. **Conclusion:** The SRUS is an entity defined by its endoscopic and histological aspect, manifesting itself mainly by rectal bleeding. Recto-sigmoidoscopy and histology are the key to diagnosis. Treatment is primarily medical. Surgical treatment is indicated in case of failure of medical treatment or in case of occurrence of complications.

**Keywords:** Solitary rectal ulcer syndrome, rectal bleeding, rectal prolapse, rectal ulceration, anorectal dyssynergia, biofeedback, rectopexy.

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## I. INTRODUCTION

The solitary rectal ulcer syndrome (SRUS) is a rare disorder of the rectal wall and its course is chronic but always benign. It was described for the first time by Cruveilhier [1] in 1829 as an unusual rectal ulcer and it was in 1969 that a description of the clinical presentation and histopathological characteristics was made, but its physiopathology remains poorly understood [2].

Its incidence is classically estimated at 1/100,000 inhabitants/year [3, 4], it is likely underestimated and SRUS is often underdiagnosed. The peak prevalence occurs between the ages of 30 and 40, however the prevalence of SRUS in men and women is almost the same and can occur at any age [5].

The pathogenic mechanism that leads to the occurrence of rectal lesions is imperfectly determined but most often associates internal prolapse of the rectal wall and an anal obstacle to evacuation (anorectal dyssynergia).

The diagnosis is suspected on a combination of clinical and endoscopic arguments and is confirmed by histology [6-8]. The diagnosis can be difficult to make and requires histological evidence to exclude cancer, ischemic colitis and chronic inflammatory bowel disease [2, 9].

The therapeutic management of SRUS remains a challenge and relies on multidisciplinary

management. It should aim to improve the patient's symptoms and quality of life [9, 10].

The objective of our study is to describe the epidemiological, clinical, therapeutic and evolutionary characteristics of SRUS in a Moroccan series.

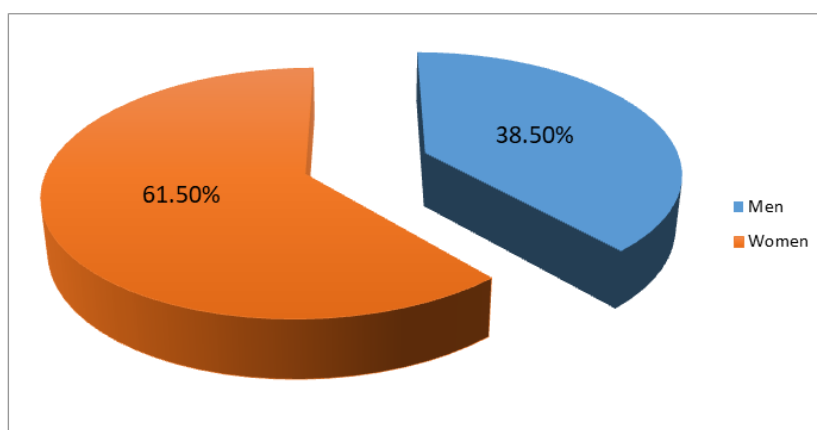
## II. MATERIALS AND METHODS

This is a monocentric descriptive retrospective study of SRUS collected over a period from January 2000 to August 2021 in the Department of Hepato-Gastro-Enterology and Proctology "Médicine B" of the Ibn Sina University Hospital. Were included all the

cases whose diagnosis of SRUS was confirmed on the clinical, endoscopic and anatomopathological criteria. We collected epidemiological data (age, sex), data on the clinical, endoscopic, anatomopathological and therapeutic presentation. We excluded patients with a diagnosis doubt or without histological confirmation.

## III. RESULTATS

We collected 26 patients with a mean age of 45 years and extremes ranging from 15 to 70 years. There were 10 men (38.5%) and 16 women (61.5%) with a sex ratio (M/F) of 0.6 with a female predominance (Chart 1).



**Chart 1: Distribution of patients by gender**

All patients were symptomatic, some patients could present several symptoms (Table 1). This symptomatology was represented by rectal bleeding in all patients, anorectal pain in 22 patients (84.6%), bloody mucus evacuations in 19 patients (73.1%) and chronic constipation in 10 patients (38.5%).

The proctologic examination showed a rectal prolapse in 34.6% of patients (n=9) and hemorrhoidal scars in 30.7% (n=8).

**Table 1: Frequency of symptoms**

Clinical symptomatology	Patients	
	(n)	(%)
Rectal bleeding	26	100
Anorectal pain	22	84.6
Bloody mucus evacuations	19	73.1
Chronic constipation	10	38.5
Rectal prolapse	9	34.6

Rectosigmoidoscopy is the key test for positive diagnosis. All our patients underwent a lower digestive endoscopy (Table 2) and it mainly detected a single clean-bottom ulceration in 50% of cases (n=13) (Figure 1), multiple ulcerations in 20% of cases (n= 6) (Figure 2) and a pseudo - polypoid appearance in 15.4% of cases (n=4) (Figure 3).

**Table 2: Endoscopic presentation**

The endoscopic aspect	Patients	
	(n)	(%)
Single clean-bottom ulceration	13	50
Multiples ulcerations	6	20
Pseudo- polypoid appearance	4	15.4
Proctitis without ulceration	2	7.7
Mid-rectal stenosis	1	3.8

Histological examination showed the typical histological appearance of SRUS in all patients with a pseudo-villous aspect of the rectal mucosa with deformed, shortened and branched glands.



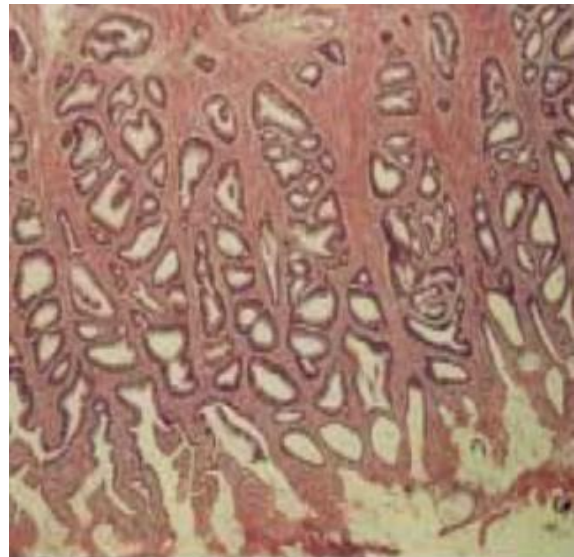
**Figure 1: Single, light-bottomed, shallow ulceration surrounded by an erythematous mucosa**



**Figure 2: Two small ulcers, resting on an erythematous mucosa**



**Figure 3: Pseudo-polypoid aspect**



**Figure 4: Histological aspect of the SRUS**

Anorectal manometry was performed in 77% of cases (n=20) and objectified dyssynergia anorectal bone and abnormal sphincter tone. Therapeutic management was based on medical treatment in all patients associated with defecation rehabilitation of "biofeedback" in 53.8% of patients (n=14). The surgical approach was considered in 5 patients (19.2%) of the cases including rectopexy in 15.4% (n=4) and resection of the stenosis with recto-anal anastomosis in 3.8% (n=1).

**Table 3: Therapeutic modalities**

Therapeutic modalities	Patients	
	(n)	(%)
Medical treatment	26	100
The defecation rehabilitation of the "biofeedback"	14	53.8
Rectopexy	4	15.4
Resection of the stenosis with recto-anal anastomosis	1	3.8

#### IV. DISCUSSION AND LITERATURE REVIEW

SRUS is a rare condition with a chronic course. Although SRUS is benign, it can affect patients' quality of life [11]. It is mainly due to prolapse and trauma to the rectal mucosa. Inappropriate contraction of the puborectalis muscle, abnormal perineal descent, and overt rectal prolapse have all been cited as possible mechanisms for the development of this affection [12].

In our series, we note that SRUS affects young adults with an average age of 45 years, but this pathology must be evoked at all ages when faced with suggestive lesions [5]. It is therefore likely to have severe social impact in patients in full professional activity [2, 13].

Several authors agree on the fact that this pathology affects young adults and occurs during the 3<sup>rd</sup>

decade of life in men and the 4<sup>th</sup> decade of life in women. However, it has also been described in children and in the geriatric population [5, 14].

The series of Tjandra, JJ *et al.*, [15], Ennaifer R *et al.*, [16], AlGhulayqah AI *et al.*, [17] and Abid, S. [18] found a mean age of 48.7 years, 42.6 years, 42.5 years and 37 years respectively. The series by C. Gouriou *et al.*, [10] noted an average age of 53 years in a series of 102 patients. Our study showed a female predominance (61.5%) with a sex ratio (M/F) of 0.6. Several recently published series corroborate our results, in particular the Tunisian series of Ennaifer R. [16] and that of AlGhulayqah AI [17] in the respective proportions of 53.3% and 55% of female cases. The results of the series by Gouriou C. *et al.*, [10] revealed a more marked female predominance of around 78% of women against 22% of men, which could be explained

by the prevalence of pelvic floor disorders which are more frequent in women.

Some authors have found results in favor of a male predominance such as Abusharifah O *et al.*, [19] who reported a male predominance of 81% in his study including 21 patients with a mean age of 11.4 years. The same results were reported by Anjum MN *et al.*, [20] and Dehghani SM *et al.*, [21] at 76.19% and 74.5% male cases respectively. However, this male predominance has only been observed in pediatric studies.

In our series, the clinical symptomatology was polymorphous and all our patients were symptomatic. Rectal bleeding was the most common symptom found in all patients (100%). They were associated with anorectal pain, bloody mucus evacuations and chronic constipation in 38.5%. In the literature, all authors agree on the fact that rectal bleeding is the most frequent and constant sign found. They are present by 56% for Tjandra JJ *et al.*, [15] to 100% of patients for Chiang JM *et al.*, [23]. Other authors have found similar results, where rectal bleeding can be isolated or associated with other symptoms to varying degrees such as dyschezia, rectal syndrome or abdominal pain [17, 14, 21, 24]. Nevertheless, it has been suggested by Tjandra JJ *et al.*, [15] that up to 26% of patients may be asymptomatic, discovered incidentally during investigations for other pathologies.

In our study, proctologic examination revealed rectal prolapse associated with SRUS in 34.6% (n=9). According to some authors, internal rectal prolapse constitutes, with anismus (absence of opening of the anorectal angle and marked imprint of the pubo-rectal muscle when pushing), factors favoring SRUS that must be systematically sought by anorectal manometry and defecography [4, 25].

Endoscopically, the examination is based on rectoscopy which objectifies the ulcer, specifies its location in relation to the rectal walls and allows biopsies to be taken in order to eliminate any cancer. The classic endoscopic appearance is that of a flat or superficial ulcer, usually suspended, resting on a sclerotic-looking base with congestive peri-ulcerative mucosa limited to the edge of the ulcer. The lesion sits on the anterior face of the rectum between 4 and 12 cm from the anal margin [16, 25].

In our series, the most common lesion was a single clean-bottom ulceration. The AlGhulayqah AI series [17] noted a single ulcer in 50% of cases and polypoid lesions in 55%. The other authors also found results close to ours, Chiang JM *et al.*, [23] found a single ulcer in 55.1% and polypoid lesions in 24% of cases. For Madigan MR *et al.*, [2] he reported in his study a single ulcer in 70% of patients and 30% were multiple ulcerations.

It is important to point out that all its elements are inconstant. The ulcer may be absent or multiple. The lesion can sit on any side of the rectum or be circumferential.

The anatomopathological study is a criterion allowing the diagnosis of SRUS and to eliminate a tumoral proliferation, an ulceration of inflammatory, radiation, iatrogenic, or ischemic proctitis. Biopsies should be performed in the perilesional area to increase the chances of finding the most typical architectural changes of the rectal wall [15, 13].

Complementary examinations, dynamic pelvic explorations and anorectal manometry, aim to identify a rectal static disorder and/or anismus. They make it possible to specify the physiopathology of SRUS and to guide management. The most common anomaly is internal prolapse of the rectum or intra-rectal or intra-anal intussusception of the rectal mucosa (up to 40% of cases according to studies) [26, 27].

Anorectal manometry sometimes proves essential for the study of sphincter function in search of hypertonia or associated incontinence of neurogenic origin [28-30]. Although these additional examinations are important in the care of patients, they have not been carried out systematically in all our patients due to a lack of means and/or the non-accessibility to these techniques. In our series, anorectal manometry was performed in 77% of cases (n=20) and objectified dyssynergia anorectal bone and abnormal sphincter tone.

Soudan D [25] noted that the frequency of dyssynergia anorectal can reach 60% to 80% in the case of SRUS [4, 31] and should ideally be reproducible on two examinations or identified clinically.

On the therapeutic level, very recently in 2020 an expert consensus on the management of SRUS was published [9]. The authors recommend management of SRUS according to the severity of the symptoms and the existence or not of a rectal prolapse [5, 6, 8, 9, 25, 32, 33].

The primary therapeutic objective is to improve symptoms and the quality of life of patients. Obtaining healing of the ulcer is uncertain and therapeutic escalation should not continue in an asymptomatic patient [9, 25].

A distinction is made between conservative medical treatment offered as first intention, endoscopic treatment and surgical treatment [6, 9].

Conservative treatment is based on the regularization of transit (fibers and laxatives), defecatory hygiene measures with learning of the correct thrust and ano-perineal rehabilitation by



biofeedback in the event of anismus. The effectiveness of biofeedback in this indication varies according to the studies from 60 to 70% [25, 29, 34].

Endoscopic treatment by argon plasma coagulation (APC) may be proposed in the presence of a bleeding ulcer. Its effectiveness has been demonstrated by two studies by Somani S. K *et al.*, [35] and Zergani FJ *et al.*, [36], who showed more than 70% bleeding control by APC versus 30% by medical treatment alone.

Surgical treatment is indicated in case of complete rectal prolapse and/or in case of failure of conservative treatment [5, 7, 8, 37]. The reference intervention in the correction of rectal static disorders is ventral rectopexy, in the event of internal prolapse of the rectum. It improves the symptoms of defecatory obstruction in 80% of cases [38]. The effect of this intervention on the healing of a SRUS has been evaluated in two studies which showed efficacy on the improvement of symptoms in more than 70% of cases and on ulcer healing in 90 to 100% of cases [39, 40].

In our study, medical treatment was recommended for all our patients (100%). This treatment was associated with ano-perineal rehabilitation by biofeedback in 53.8% (n=14). Recourse to surgery was indicated by the failure of medical treatment and/or biofeedback and was observed in 19.2% of cases (n=5). The surgical techniques performed are rectopexy in 15.4% (n=4) and resection with recto-anal anastomosis in 3.8% of cases (n=1). The postoperative course was simple with no major complications. The small number of our sample and the nature of the retrospective study constitute a limitation of our study because all aspects of long-term follow-up could not be analyzed.

According to Meurette G *et al.*, [11] it seems necessary to monitor patients and follow them over the long term, since significant alterations in their quality of life may occur. In particular, the recurrence rate is high and is estimated at 30% after surgery. Gouriou C *et al.*, [9] suggested annual clinical assessments during follow-up to assess symptoms, mucosal healing status, and to adjust medical therapy.

## V. CONCLUSION

SRUS is a rare, chronic and benign clinical entity defined by its endoscopic and histological appearance, manifesting mainly by rectal bleeding. Its etiopathogenesis remains incompletely identified, but the association of internal rectal prolapse and a mechanical anal obstacle to evacuation contributes to the genesis of mucosal lesions. The search for contributing factors must be systematic by anorectal manometry and defecography. The first-line treatment is conservative medicine associated with ano-perineal

rehabilitation by biofeedback. Surgical treatment is indicated in case of failure of medical treatment.

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