Case Report

Treatment of rectal leiomyoma by endoscopic resection

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SILVA RDP; SAAD-HOSSNE R; FERRAZ RA; MATSUSHITA MM; FALZONI R; TEIXEIRA FV. Treatment of rectal leiomyoma by endoscopic resection. **J Coloproctol,** 2011;31(4):382-386.

ABSTRACT: Leiomyomas of the rectum are rare, with low reported incidence in literature. In most cases, patients are asymptomatic, and are often incidental endoscopic findings. The difficult distinction from leiomyosarcomas, associated with the possibility of recurrence, implies the absence of a standard treatment. Endoscopic resection, if well indicated, may be a therapeutic option. In this study, we report two cases of asymptomatic leiomyoma of the rectum in two patients, discovered incidentally during a routine colonoscopy, removed by conventional polypectomy and discuss its diagnostic and therapeutic aspects based on a literature review.

Keywords: rectal neoplasm; leiomyoma; colonoscopy.

INTRODUCTION

Leiomyoma and leiomyosarcoma, tumors of smooth muscle, can occur in the whole gastrointestinal tract[1-3], affecting more frequently organs such as the stomach (65%) and small bowel (25%)[4] and rarely found in the rectum[1,5-14].

Leiomyoma of the rectum occurs in approximately 1 out of 2,000-3,000 rectal tumors[4,8] and, although such occurrence is rare, many cases have been reported in the literature[1,4-14] since its first histopathological confirmation described by Malassez in 1872[4].

These tumors are originated in the smooth muscle fibers of mucosa or muscular fibers of the circular and longitudinal layers of the rectal wall[4,8,12,15,16] or blood vessel walls[4,12,16]. They occur especially in the distal two-thirds of the rectum[13,15] and tend to present intraluminal growth[7].

Most cases are asymptomatic[1,9,10,17,18], and for this reason, found incidentally while performing endoscopic procedures[1,3,5,9,10,16,18,19].

The difficult distinction from leiomyosarcoma, associated with the possibility of recurrence, implies the absence of a standard treatment[9,11].

The purpose of this study was to report the authors' experience with endoscopic resection in two cases of leiomyoma of the rectum found incidentally during a colonoscopy and discuss its diagnostic and therapeutic aspects based on a literature review.

Study carried out at the Service of Endoscopy, Digestive Tract Surgery and Coloproctology at UNIGASTRO, Associação Beneficente Hospital Universitário (ABHU) – Marília (SP), Brazil.
Financing source: none.
Conflict of interest: nothing to declare.

Submitted on: 08//11/2010
Approved on: 10/27/2010

CASE REPORTS

Report 1

A 39-year-old male patient, was sent for investigation of a lower gastrointestinal bleeding that had started one year before. He reported that the bleeding was sporadic after evacuating, noticed during anal cleansing, with streaks of blood on the toilet paper, without any other associated symptom. General and abdominal examinations showed no alteration. The proctologic examination showed first-degree hemorrhoid. No tumor was evidenced with digital rectal examination. To better elucidate the situation, the patient was submitted to colonoscopy, which detected a sessile polypoid lesion, of smooth surface, around 4 mm diameter, with no alterations to the mucosa and no signs of lower rectal bleeding (Figure 1A). A loop polypectomy was performed, without complications. The patient was discharged one day after the procedure.

Report 2

A 56-year-old male patient came in with rectal pain at evacuation for 6 months. He reported burning sensation of moderate intensity, caused by evacuation and relieved after passing stool. He denied any history of bleeding or other associated symptoms. General and abdominal examinations showed no alteration. The proctologic examination showed a chronic anal fissure in the posterior midline. The digital rectal examination did not detect any tumor. Colonoscopy showed a sessile polyp of 5 mm diameter, smooth surface, no alterations to the mucosa and no signs of upper rectal bleeding (Figure 1B). The endoscopic resection of the lesion (loop polypectomy) was performed, without complications. The patient was discharged one day after the procedure.

The two dissected specimens were submitted to an anatomopathological analysis. Macroscopically, they presented uniform whitish color and consistency. No area with necrosis or cystic alterations were observed.

In both lesions, both showed in microscopy mucosa of colonic pattern without interruption. In the first, the glass plate showed proliferating smooth muscle cells, with relatively abundant cytoplasm, and arranged in crossed-over bundles. The nuclei were elongated and regular, with delicate and evenly distributed chromatin, without evidence of necrosis. The second presented crossed-over bundles of smooth muscle cells in the submucosa, individually uniform, with elongated nuclei and smooth margins, located in the cell nuclei, with no areas of necrosis. In both dissected specimens, the margins did not present neoplasm, and no mitosis was observed in ten high-resolutions fields.

At the immunohistochemical analysis, the tumors were positive for smooth muscle actin and desmin and negative for CD117 (c-kit) (Figures 2A-D).

Both patients are now receiving outpatient care, with no symptoms and signs of local recurrence, as well as no colon lesions in colonoscopy examinations made in the follow-up period.

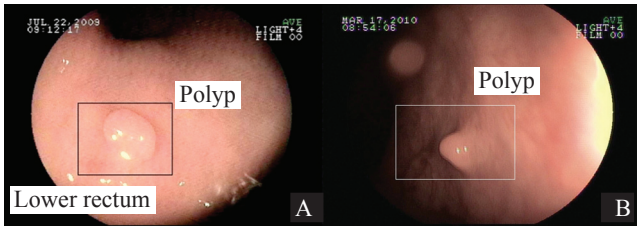

Figure 1. *Colonoscopy showing: (A) sessile polypoid lesion in the lower rectum, of smooth surface and around 4 mm diameter, with no alterations to the mucosa and no signs of bleeding; (B) sessile polyp in the upper rectum, of 5 mm diameter, smooth surface, with no alterations to the mucosa and no signs of bleeding.*

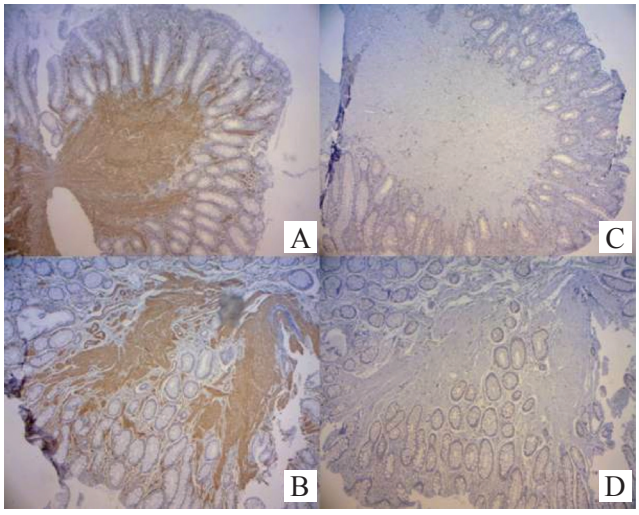

Figure 2. *Immunohistochemical blades of specimens endoscopically resected via rectum showing positivity for smooth muscle actin (case 1 - A and case 2 - B) and negativity for CD117 (c-kit) (case 1 - C and case 2 - D).*

DISCUSSION

Leiomyoma of the rectum represents only 3% of all gastrointestinal leiomyoma[1], and less than 0.1% of rectal tumors[1,4,8].

So far, the most important compilation of anorectal tumors of smooth muscle (432 leiomyoma and 480 leiomyosarcoma) found in worldwide literature has been that published by Hatch et al.[7], in 2000. In Brazil, few cases of smooth muscle tumors of the rectum have been described[15,16,18,20-24].

In general, these tumors occur more predominantly in individuals between 40 and 59 years old[7]. This study investigated two male patients and, in the case compilation made by Hartch et al.[7], the probability that men will develop anorectal benign and malign tumors of smooth muscle was a little higher than in women[7]. Regarding the wall location, there seems to be no significant difference when comparing leiomyoma and leiomyosarcoma[7].

The clinical manifestations vary with the size, location and direction of the tumoral growth[1]. Most patients are asymptomatic[1,9,10,17,18]; discomfort or local pain, associated or not with defecation, palpable mass, sensation of a strange body, changes in intestinal habit and rectal bleeding[1,3,9,15,16,18] are sporadically reported in the literature[1,9,25,26]. Usually, when symptomatic, the tumors are diagnosed within one year of symptom onset[7]. As in the cases reported in this study, most tumors are intraluminal and sessile[1,5,6] and may occasionally present a pedicle[1,5,10].

In both cases described here, the patients had proctologic complaints - the first was related hemorrhoid and the second to a chronic anal fissure -, and these findings are similar to those in the literature, where most lesions are incidentally diagnosed, through digital rectal examination or endoscopy[1,3,5,9,16-19], before the patients had any complaint[1,5,17].

Besides colonoscopy, some examinations can help in the lesion diagnosis and staging: opaque enema, colonoscopy, computed tomography, magnetic resonance and endoanal ultrasound[1,9,11,15,16,18]. Therefore, the histopathological analysis will determine the sure diagnosis and the distinction between benign and malign lesions[11,16,21].

Particularly when the lesion is in the rectum, the endoanal ultrasound can help define the lesion extent (size, number and layer it originated)[1,7,11,14,18,21]. It can also help distinguish benign from malign disease (rupture of normal tissue planes, cystic degeneration and lymphadenopathy)[7,11,14,19].

Differential diagnosis includes schwannomas, carcinoid tumors, neurofibroma, hemangioma, endometriosis, lipomas, melanoma, and especially gastrointestinal stromal tumors (GISTs) and leiomyosarcoma[6,16,27]. The distinction of these lesions is essential, as it affects the proper therapy selection[14,19,27], as each lesion has specific clinical and pathological characteristics[6,27].

Histologically, leiomyomas differ from GISTs due to their uniform positivity in immunohistochemistry for smooth muscle actin and desmin and their negativity for CD34 and CD117 (c-kit), unlike the GISTs[1,5,6,14,16,19,27].

On the other hand, differing leiomyomas from leiomyosarcomas is often difficult[2,7-9,13,14-16,19,21,25-28]. In histology, leiomyosarcomas present differentiated smooth muscle cells[6,14]; most of them of high-degree type, with focal pleomorphism and increased mitotic activity[6,27], and presenting the same immunohistochemical pattern as that of leiomyomas[6,27].

However, no criteria have been established to determine the malignity of these tumors[19,21]. Then, certain characteristics suggest the malignity degree, including the tumor size (>5 cm), histological aspect (necrosis of tumor cells, ulceration and cell atypia), and the number of mitotic figures per 10 high-power fields[1,8,14,15,18,19,27,28]. From these, the latter is the most important malignity criterion[7,14,16]; tumors of less than two mitoses per 10 fields offer good prognosis; however, if two or more mitoses are found, the tumor is usually considered as malign[7,19]. Therefore, a complete analysis of the resected specimens is essential for malignity diagnosis and correct selection of treatment plans[27].

Due to the difficult distinction between benign and malign tumors[2,7-9,13-16,19,21,27], possibility of recurrence[7-9,11-13,16,19,21,25,29] and insensitivity of these tumors to adjuvant therapies[7,8,14-16,18,19,28], most authors recommend the surgical removal of the tumor[3,7,8,11-19,29-31], either through open[15,16,18,21] or endoscopic procedure[1,2,5,10,12,21,32,33].

The surgical treatment should ensure tumor-free margins[1,18]. The options include transanal excision,

endoscopic resection, lower anterior resection or abdominoperineal amputation[7,9,15,19].

In both cases, endoscopic resection of the lesion was considered a therapeutic procedure, as the margins were free of neoplasm, and the tumors were smaller than 5 cm, without necrosis, ulceration and cell atypia, and the number of mitoses per 10 high-power fields was less than two[1,8,14-15,19,27,28].

Some cases using endoscopic resection of rectal leiomyoma have been described[2,10,12,32,33]. This approach is a valid alternative to invasive surgery when the complete removal of the lesion is ensured[5,32]. However, it may lead to hemorrhage and perforation[1,2,32,34].

Therefore, endoscopic resection is improper to leiomyosarcoma and leiomyoma of ≥2 cm diameter or originated in the *muscularis propria*, due to the risk of hemorrhage and perforation[32]. Thus, it is extremely important to determine the layer of lesion origin[32]. In this context, endoscopic ultrasound will help decide about the proper surgical procedure[1,7,11,14,18,34], as the distinction from the lesion origin layer is important for the proper surgical planning[2,11,14,34]. Usually, leiomyomas originated in the *muscularis mucosa* can be endoscopically resected, while for those originated in the *muscularis propria*, this procedure should be avoided[34]. Unfortunately, this procedure was not performed in the patients of this study.

Prognosis is uncertain for these tumors[25], due to the recurrence rate[7-9,11-13,16,19,21,25,29], and the short follow-up period for the cases reported above[5,7,15,21,25]. Then, extended follow-up is important to confirm a disease-free status[2,5,7,14,15,18,19,21]. The postoperative follow-up can include tomography, flexible digestive endoscopy and endoanal ultrasound[4,14,19,21].

We have concluded that leiomyoma in the rectum is rare, usually asymptomatic, and that it should be distinguished from GISTs and leiomyosarcoma. For this purpose, histological and immunohistochemical analyses of the whole specimen should be performed by a pathologist with experience in this type of lesion. Endoscopic resection is an therapeutic option, since it enables the complete removal of the tumor.

RESUMO: Os leiomiomas de reto são raros, com baixa incidência relatada na literatura; na maioria dos casos os pacientes são assintomáticos, sendo que em muitos casos são achados incidentais endoscópicos. A dificuldade de distinguí-los dos leiomiossarcomas, aliada a possibilidade de recorrência, implica na inexistência de um tratamento padrão. A ressecção endoscópica desde que bem indicada pode ser uma opção terapêutica. Reportamos dois casos de leiomioma de reto assintomáticos em dois pacientes, descobertos casualmente durante exame colonoscópico de rotina, removidos por polipectomia convencional e discutimos seus aspectos diagnósticos e terapêuticos, através de uma revisão da literatura.

Palavras-chave: neoplasia retal; leiomioma; colonoscopia.

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