



CASE REPORT / ПРИКАЗ БОЛЕСНИКА

Rare case of an anorectal leiomyoma

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Introduction Leiomyoma is a kind of benign tumors of smooth muscle origin, occurring mostly throughout the genitourinary system. They are rarely found in the anorectal region, where they represent less than 0.1% of the tumors of the rectum. Due to this exceptional occurrence, we report a case of leiomyoma arising in the anorectal region.

Case outline A 43-year-old otherwise healthy woman presents with the big mass in analorectal region. Major complain is pain and discomfort while sitting. Magnetic resonance imaging is done and pathohistology report confirmed large leiomyoma. Abdominoperineal resection of the rectum was performed due to the size of the tumor. Postoperative course was uneventful.

Conclusion There is no gold standard technique, and decision about the best surgical approach for all pathohistological types of tumors in the anorectal region. The decision sometimes has to be made according to size and major complains.

Keywords: anal canal; leiomyoma; surgery

INTRODUCTION

Leiomyoma are benign tumors of smooth muscle origin occurring throughout the genitourinary system [1]. They are usually present as an intrauterine tumor, but may occasionally be extrauterine. The leiomyomas, which evolve to lose their attachment from the uterus and become adherent to other organs (such as the broad ligament, omentum, or retroperitoneal connective tissue) and acquire blood supply from them, are called parasitic leiomyomas. They can be found anywhere in the abdomen and pelvis. Extrauterine leiomyoma can be classified according to the place of origin as a piloleiomyoma (derived from erectile muscles of hair follicles), an angioleiomyoma (from blood vessel smooth muscle wall), or a genital leiomyoma (derived from) tunica dartos of the scrotum [2]. In the digestive tract, the most common presentations are in the stomach, followed by the small intestine, but are rarely found in the anorectal region, where they represent less than 0.1% of the tumors of the rectum, with a rare presentation in soft parts, mainly in the perianal topography [3, 4]. Literature shows that the anal leiomyoma is a rare finding: isolated cases have been reported, and the most common presentation is a painless tumor in this area [4]. Due to this exceptional occurrence, we report a case of leiomyoma arising in the anorectal region.



Figure 1. Clinical presentation of the anorectal leiomyoma

and uneasiness while sitting. She noticed a bump in the anal region, which got bigger in size through the course of one year. This was her first visit. She denied other symptoms except pain during defecation. Her medical family history was unremarkable. Physical examination revealed a large mass protruding from the anal canal and the anus itself (Figure 1). On rectal examination, the anus was passable for a fingertip; endoscopy or endoanal ultrasonography could not be performed due to extreme discomfort and size of the lesion. The laboratory and biochemical results were within the referential values, findings on abdominal ultrasonography were normal. The magnetic resonance imaging (MRI) was done followed by a biopsy. Pelvic MRI showed an expansive tumor of the rectum of the anus without the uterus and the bladder infiltration (Figure 2). Pathohistology report confirmed leiomyoma. Due to the size of the tumor and infiltration of muscles, abdominoperineal resection was performed (Figure 3). The postoperative course

CASE REPORT

A 43-year-old otherwise healthy woman came to the office because of anal region disorders

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Figure 2. Magnetic resonance imaging finding of the large anorectal leiomyoma

was uneventful. One year after the surgery, control examination results were good, and the patient gave written consent for this case presentation, including the accompanying images, case history, and data.

DISCUSSION

In 1845, the first description of leiomyoma was made by Virchow, who defined it as a benign tumor of mesenchymal origin developing from smooth muscle fibers. The leiomyoma can develop in any place where smooth muscle is present. In the previous classification, it belonged to the so-called gastro-intestinal stromal tumor (GIST). Nowadays, the GIST is considered a soft tissue tumor, as well as the leiomyoma, but it has its own identity based on specific immunohistochemical pattern, while many tumors previously defined as leiomyoma are now classified as GIST and even have a different type of treatment [5, 6]. The leiomyoma have exhibited positivity for smooth muscle actin and negativity for CD117 and CD34 (c-Kit). Desmin may be absent, or it may be expressed later [4, 7]. Histological patterns are similar to those of leiomyosarcoma, but in leiomyosarcoma, a higher degree of cellular atypical activity with local pleomorphic and increased mitotic activity is expressed. There are no well-established criteria to determine malignancy in these tumors, with some characteristics suggesting a malignant behavior: tumor size (> 5 cm), histological appearance (necrosis, ulceration, or cells with atypical) and increased number of mitoses (> 2 mitoses per field, with a 10-fold increase) [4, 8]. In the case presented here, the growth and the size of the tumor suggested its malignant behavior.

It is classified as superficial or deep. The latter is further divided in somatic and retroperitoneal [2]. The superficial variant usually affects the extremities with the same incidence in both sexes whereas the retroperitoneal generally involves the pelvic region in fertile women, like in this case



Figure 3. Specimen after abdominoperineal resection for anorectal leiomyoma

[2, 9]. The organs mainly involved in gastrointestinal tract are the stomach and small bowel, less frequent regions are esophagus, colon, and anorectal localization which is fairly uncommon [5, 10]. In the anorectal region, they represent only 3% of leiomyomas of the gastrointestinal tract and less than 0.1% of rectum tumors, rarely found in soft tissues, mainly in the perianal area, and 3.8% incidence of all soft tissues benign tumors [11].

Considering their growth type, these tumors are divided into three variants: intraluminal, extraluminal, and intramural. Intraluminal leiomyomas are usually located in the posterior wall of the distal part of the rectum as in the case presented here, and they may be sessile or pedunculated. On the other hand, extraluminal leiomyomas generally grow from the colonic wall inside the abdomen and they often resemble a GIST [12]. Sometimes the tumors grow in both directions, forming an “hour glass,” which could be said for this case according to the MRI [5, 6].

Preoperative diagnosis is difficult because most patients are asymptomatic and these tumors are commonly identified during endoscopy and imaging examination. The symptoms related to the presence of leiomyoma vary widely. They may include pain, rectorrhagia, tenesmus or they can be confirmed by a biopsy and the treatment depends on their alteration, which is confirmed by medical examination. Biopsy is often non-informative because it does not involve the entire tumor, including the full evolution of histological characteristics [3, 6]. The radiological imaging, such as MRI or 360° tridimensional transrectal ultrasound, are useful to identify and localize the mass, to provide information on its relationship with the adjacent structures such as the anal sphincter or uro-gynecological structures and to indicate the operative strategy [5].

The treatment of perianal leiomyoma consists in the complete surgical resection, ensuring tumor-free margins [3]. Since local resection was impossible due to the size of the lesion and sphincter involvement, abdominoperineal resection was performed as suggested in other cases reported. The recurrence rate is low when complete local excision is performed, but the extended postoperative follow-up with clinical and complementary examinations is important to

confirm the absence of the disease or to detect any recurrences and/or malignant transformation [12, 13]. All perianal lesions need a final diagnosis, leiomyoma should be considered in differential diagnosis of tumors in anorectal region. The best surgical approach depends not only on

nature of the tumor, but sometimes decision has to be made according to size and major complains.

Conflict of interest: None declared

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Редак случај аноректалног лејомиома

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САЖЕТАК

Увод Лејомиоми су бенигни тумори порекла глатких мишића који се углавном јављају на генитоуринарном тракту. Ретко се срећу у аноректалној регији, где чине мање од 0,1% свих тумора ректума. У раду је приказан изузетно редак случај лејомиома аноректалне регије.

Приказ болесника Четрдесеттворогодишња, иначе здрава жена јавила се због велике масе у аноректалној регији. Жалила се на бол и нелагодност током седења. Магнетна

резонанца уз патохистолошки налаз показала је велики лејомиом. Због величине тумора и локализације изведена је абдоминоперинеална ресекција ректума. Постоперативни ток протекао је без компликација.

Закључак Не постоји златни стандард који се тиче хируршког лечења за све патохистолошке типове тумора у аноректалној регији. Одлука се понекад доноси у односу на величину тумора и главне тежбе болесника.

Кључне речи: анални канал; лејомиом; хирургија