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Rare case of an anorectal leiomyoma

Редак случај анаоректалног лејомиома

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SUMMARY

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 forty-three years 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. Abdominoperianal 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

САЖЕТАК

Увод Лејомиоми су бенигни тумора порекла глатких мишића који се углавном јављају на генитоуринарном тракту. Ретко се срећу у анаоректалној регији, где чине мање од 0.1% свих тумора ректума. У раду је приказан изузетно редак случај лејомиома анаоректалне регије.

Приказ случаја Иначе здрава жена, стара 43 године, јавља се због велике масе у анаоректаној регији. Жали се на бол и нелагодност током седења. Магнетна резонанца уз патохистолошки налаз показује велики лејомиом. Због величине тумора и локализације изведена је абдомино-перинеална ресекција ректума. Постоперативни ток протиче без компликација.

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

Кључне речи: анални канал; лејомиом; хирургија

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 may 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 scrotum) [2]. In the digestive tract, most common presentations are in the stomach, followed by the small

intestine, but being 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 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.

CASE REPORT

A forty-three year old, otherwise healthy woman comes to the office because of anal region disorders and uneasiness while sitting. She noticed a bump in anal region which got bigger in size through the course of one year. This was her first visit. She denies other symptoms except pain during defecation. She has no family history. Physical examination revealed a big mass protruding from anal canal and anus itself (Figure 1.). On rectal exam: the anus is 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, finding on abdominal ultrasonography was normal. The magnetic resonance imaging (MRI) was done followed by 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 abdomino-perineal resection was performed (Figure 3). Postoperative course was uneventful. One year after surgery, the control examination was a good, and patients gave written consent for case presentation.

DISCUSSION

The first description of leiomyoma was made in 1845 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 the 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 have even 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 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 10x 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, as in this case [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 posterior wall of the distal part of the rectum as in 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 not 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 give its relationship with the adjacent structures such as 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 abdomino perineal 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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REFERENCES

1. Goldman HB, McAchran SE, MacLennan GT. Leiomyoma of the urethra and bladder. *J Urol* 2007; 117:1890. DOI: [10.1016/j.juro.2007.02.017](https://doi.org/10.1016/j.juro.2007.02.017) PMID: 17437842
2. Bhargava P, Haque K, Veja R, Turbat-Herrera E, Chu Q, Sangester G, D'Agostino H. Parasitic leiomyoma presenting as an inguinal hernia in a postmenopausal woman: a case report. *Radiol Case Rep* 2018;13:767–71. DOI: [10.1016/j.radcr.2018.04.014](https://doi.org/10.1016/j.radcr.2018.04.014) PMID: 29887930
3. Garacía-Santos EP, Ruescas-García FJ, Estaire-Gómez M, Martín-Fernández J, González-López I. Anorectal leiomyoma: a case report and literature review. *Ver Gastroenterol Max.* 2014;79:58–66. DOI: [10.1016/j.rgmx.2013.05.002](https://doi.org/10.1016/j.rgmx.2013.05.002) PMID: 24629571
4. Fernandes B, Rodrigues L, Dadalto P, Boteli F, Formiga G, Costa A. Perianal leiomyoma: a case report. *J Coloproctol (RIO J).* 2017;238–41. DOI: [10.1016/j.jcol.2017.03.003](https://doi.org/10.1016/j.jcol.2017.03.003)
5. Sturale A, Fabian B, Naldini G. A rare case of leiomyoma of the internal anal sphincter: a case report. *Int J Surg Case Rep* 2016;23:4–7. DOI: [10.1016/j.ijscr.2016.03.048](https://doi.org/10.1016/j.ijscr.2016.03.048) PMID: 27078867.
6. Dagmura H, Daldal E, Akbaş A, Daşiran Fatih. A rare anal mass: anal leiomyoma presented as perianal fistula. *J Surg Case Rep* 2019;2,1–3. DOI: [10.1093/jscr/rjy351](https://doi.org/10.1093/jscr/rjy351)
7. Sunkara T, Then EO, Culliford A, Gaduputi V. Rectal leiomyoma, rare entity. *Clin Pract* 2018;21;8(2):1053. DOI: [10.408/cp.2018.1053](https://doi.org/10.408/cp.2018.1053) PMID: 29657702
8. Saunders RN, Pattenden C, Agarawal PK. Heavy rectal bleeding secondary to the passage of a rectal leiomyoma per anus. *Ann R Coll Surg Engl* 2004; 86(6):W44–6. DOI: [10.1308/147870804100](https://doi.org/10.1308/147870804100) PMID: 16749966
9. Misusmi S, Irie T, Fukuda K, [Tada S](#), [Hosomura Y](#). A case of deep soft tissue leiomyoma: CT and MRI findings. *Radiat Med* 2000;18:437–43. PMID: 11247002
10. Sun P, Ou H, Huang Sh, Wei L, Zhang S, Liu J, Geng Sh, Yang K. Perianal atypical leiomyoma: case report. *Medicine (Baltimore)* 2018;97(13). PMID: 29595693
11. De Palma GD, Rega M, Masone S, et al. Lower gastrointestinal bleeding secondary to rectal leiomyoma. *World J Gastroenterol* 2009;15:1769–70. PMID 19360922
12. Dasari BVM, Khosraviani K, Irwin TS, Scott M. Perianal leiomyoma involving the anal sphincter. *Ulst Med J.* 2007;76:173–4. PMID:17853650
13. Kock KS. Leiomioma retal: relation de casoe revisao de literature. *Verbras Coloproct* 2004;24:170–3.



Figure 1. Clinical presentation of anorectal leiomyoma

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Figure 2. Magnetic resonance imaging finding of large anorectal leiomyoma



Figure 3. Speciment after abdominoperineal resection for anorectal leiomyoma