Could the endoscopic resection of a large rectal leiomyoma be an effective and safe technique?

Emanuele Sinagra^{1,2,3}*, Giovanni Tomasello^{3,7}*, Giancarlo Pompei⁴, Massimo Menozzi⁵, Stefano Mandalà⁶, Francesca Rossi¹, Guido Martorana⁶, Marco Messina⁸, Aroldo Gabriele Rizzo⁴, Marco Calvaruso⁶, Gaetano Cristian Morreale¹⁰, Georgios Amvrosiadis¹⁰, Cappello Francesco^{3,7}, Carini Francesco⁷, Dario Raimondo¹

¹ Fondazione Istituto S. Raffaele - G. Giglio, Gastroenterology and Endoscopy Unit, Cefalù, Italy; ² Phd Course in Surgical Biotechnology and Regenerative Medicine, School of Medicine, University of Palermo, Italy; ³ Euro-Mediterranean Institute of Science and Technology (IEMEST), Palermo, Italy; ⁴ Fondazione Istituto S. Raffaele - G. Giglio, Pathology Unit, Cefalù, Italy; ⁵ Fondazione Istituto S. Raffaele - G. Giglio, Radiology Unit, Cefalù, Italy; ⁶ Fondazione Istituto S. Raffaele - G. Giglio, Surgery Unit, Cefalù, Italy; ⁷ Department of Experimental Biomedicine and Clinical Neuroscience, Section of Human Anatomy, (BIONEC), University of Palermo, Italy; ⁸ Fondazione Istituto S. Raffaele- G. Giglio, Oncology Unit, Cefalù, Italy; ⁹ Ospedali Riuniti V. Sofia-V.Cervello, Pathology Unit, Palermo, Italy; ¹⁰ Ospedali Riuniti V. Sofia-V.Cervello, Gastroenterology Unit, Palermo, Italy

Summary. Rectal leiomyomas are a rare conditions, with low reported incidence in literature and constitute about 0.1% of rectal tumours; in fact rectal leiomyomas occur in approximately 1 out of 2000-3000 rectal tumors. We report on a patient with a 3 cm semi-pedunculated colonic leiomyoma, which was successfully removed by endoscopic polypectomy after normal saline-epinephrine submucosal injection. When we encounter a tumor during a colonoscopic examination, we usually evaluate the tumor carefully and perform an endoscopic resection when we judge it is appropriate. When a symptomatic smooth muscle tumors smaller than 2 cm are incidentally found on colonoscopy, surgical resection is unnecessary. Furthermore, if a tumor can be lifted with a snare and it is either pedunculated or semi-pedunculated, endoscopic resection might be a safe option. For those tumors with wide-based or exoluminal growth, endoscopic removal should be avoided due to the higher risks of bleeding and perforation. The histological findings of the resected tumor are important. If there is any malignant element that can not be completely eradicated, we would suggest surgical treatment. We believe our process allows to avoid unnecessary surgery and reduces medical costs.

Key words: rectal leiomyoma, endoscopic resection, gastrointestinal stromal tumors

Introduction

Rectal leiomyomas are a rare conditions, with low reported incidence in literature. Most gastrointestinal leiomyomas are found in the stomach (1, 2), but they may also occur in the esophagus (3), small intestine (4), colon (5), and rectum (6-17). Only 3% of these smooth

muscle tumours arising from colon are gastrointestinal leiomyomas and constitute about 0.1% of rectal tumours; in fact rectal leiomyomas occur in approximately 1 out of 2000-3000 rectal tumors (7, 11) and, although such occurrence is rare, many cases have been reported in the literature since its first histopathological confirmation described by Malassez in 1872 (7, 18).

^{*} Sinagra Emanuele and Tomasello Giovanni contributed equally to this work

These tumors are originated in the smooth muscle fibers of mucosa or muscular fibers of the circular and longitudinal layers of the rectal wall (7, 11, 16, 19, 20) or blood vessel wall (7, 19, 20). They occur especially in the distal two-thirds of the rectum (17, 19) and tend to present intraluminal growth (11).

Rarity of this tumour delays early diagnosis but once identified it should be resected to avoid complications like bleeding and mechanical obstruction. Generally, large leiomyomas are believed to be best treated by surgical resection, because conventional colonoscopic resection of large and deep-seeded tumors poses a high risk of perforation (21). We report on a patient with a 3 cm semi-pedunculated colonic leiomyoma, which was successfully removed by endoscopic polypectomy after normal saline-epinephrine submucosal injection.

Case report

A 70-year-old male patient, with the evidence, at the computed tomography (CT) scan (performed for the occurrence of a symptomatic thrombosis of the left common femoral vein), of a thickening of the anterior rectal wall, was admitted to our Endoscopy Unit to perform a colonoscopy.

There was no history of hematochezia, abdominal cramping, fever, chills, nausea, vomiting or body weight loss. His haemoglobin level was 14 g/dL (normal range 12-16 g/dL), with a normal haematocrit and mean corpuscolar volume. The clinical examination of the patient was unremarkable. The per-rectal examination did not reveal any mass or blood. The colonoscopic examination, performed through a standard colonoscope (CF-Q165I, Olympus), revealed a 3 cm semi-pedunculated tumor located at 10 cm from anal verge, with no alterations to the mucosa and no signs of lower rectal bleeding. (Fig. 1) To remove the large tumor safely, firstly, 10 milliliters of normal saline-epinephrine was injected into the base and the vicinity of the tumor with a local injection needle. With the mucosa bulging after the normal saline injection, we could prevent hemorrhage. Second, the endoscopic resection of the lesion (loop polypectomy) was performed, without complications. However, a hemoclip (Resolution® Clip, Boston Scientific)

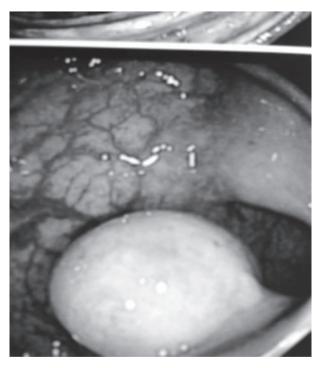


Figure 1. Colonoscopic examination: (performed through a standard colonoscope - CF-Q165I, Olympus), revealed a 3 cm semi-pedunculated tumor located at 10 cm from anal verge, with no alterations to the mucosa and no signs of lower rectal bleeding.

was then applied to prevent the occurrence of late bleeding. Patient had a good and uneventful postoperative recovery, and was then discharged in the same day of the procedure.

Gross examination revealed a semi-pedunculated polypoid lesion with a maximum diameter of 3 cm, with smooth surface (covered by non-ulcerated mucosa), of gray color; the cut surface looked whitish, with a spindle aspect, and a fibrous texture. Histological assessment revealed diffuse well-defined spindle-shaped smooth muscle cells with no mitosis. Immunohistochemical staining was positive for smooth muscle actin (SMA) and desmin (DESM), but negative for CD34, S100 and CD117. The tumoral growth were inserted on the superficial aspect of the muscularis mucosae, completely merging with it; the overlying epithelium presented alterations consisting with mild pressure atrophy (oedema, vasal congestion and mild crypt distortion); no superficial ulceration was noted. The resection margins consisted in small band of smooth muscle fibers with orderly arrangement, parallel to the superficial epithelium (remnants of muscularis mucosae). Based on these findings the lesion was classified as leiomyoma, completely resected. At the endoscopic follow-up the patient was free of disease at three months.

Discussion

Leiomyoma of the rectum represents only 3% of all gastrointestinal leiomyoma (6), and less than 0.1% of rectal tumors (7, 11). Smooth muscle neoplasms are the second most common mesenchymal neoplasms of gastrointestinal tract gastrointestinal stromal tumors (GIST).

In general, these tumors occur more predominantly in individuals between 40 and 59 years old. According the case compilation performed by Hartch and coworkers (10), the probability that men will develop anorectal benign and malign tumors of smooth muscle was a little higher than in women (10).

The clinical manifestations vary with the size, location and direction of the tumoral growth (6). Most patients are asymptomatic (6, 12, 13, 22, 23); discomfort or local pain, associated or not with defecation, palpable mass, sensation of a strange body, changes in intestinal habit and rectal bleeding (6, 12, 19, 20, 23, 24) are sporadically reported in the literature (6, 25, 26). Usually, when symptomatic, the tumors are diagnosed within one year of symptom onset (10). Most tumors are intraluminal and sessile (6, 8, 9) and may occasionally present a pedicle (6, 8, 13) as in the case reported in this article.

Leiomyomas are similar, at the macroscopic appearance, to gastrointestinal stromal tumors (GISTs) under light microscopy. With the advent of immunohistochemistry and electron microscopy, we have learned that they are of distinct origins. GISTs are believed to grow from gastrointestinal pacemaker cells, "Cajal cells", and usually react with c-kit (27). On the other hand, leiomyomas and leiomyosarcomas are myogenic in origin, and usually react positively for smooth muscle actin and negatively for c-kit (28). The tumor resected in our case was positive for smooth muscle actin and desmin, and negative for CD34, CD117 and S-100. No mitosis was seen on microscopic examination, and no evidence of distal metastasis was found.

Therefore, this tumor was clearly defined as a leiomyoma.

Up to now, several cases regarding endoscopic resection of rectal leiomyoma have been described (13, 15, 29-31). This approach is a valid alternative to surgical intervenction, when the complete removal of the lesion is ensured. However, it may lead to complication, as hemorrhage and perforation (6, 30-32).

Therefore, endoscopic resection is considered improper to leiomyosarcoma and leiomyoma of ≥2 cm diameter or originated in the *muscularis propria*, due to the higher risk of hemorrhage and perforation. Thus, it is extremely important to determine the layer of lesion origin. In this context, endoscopic ultrasound will help decide about the proper treatment procedure, as the distinction from the lesion origin layer is important to decide the most suitable surgical planning. Usually, leiomyomas originated in the *muscularis mucosa* can be endoscopically resected, while for those originated in the *muscularis propria*, is better to avoid this procedure. Unfortunately, endoscopic ultrasound was not performed in the patients of this study.

The prognosis of these tumors is still uncertain. Then, an extended follow-up is important to confirm a disease-free status. The postoperative follow-up could include CT, flexible digestive endoscopy and endoanal ultrasound.

In conclusion, we present a rare case of rectal leiomyoma successfully removed endoscopically with no post procedure complications. When we encounter a tumor during a colonoscopic examination, we usually evaluate the tumor carefully and perform an endoscopic resection when we judge it is appropriate. When a symptomatic smooth muscle tumors smaller than 2 cm are incidentally found on colonoscopy, surgical resection is unnecessary. Furthermore, if a tumor can be lifted with a snare and it is either pedunculated or semi-pedunculated, endoscopic resection might be a safe option. For those tumors with wide-based or exoluminal growth, endoscopic removal should be avoided due to the higher risks of bleeding and perforation.

The histological findings of the resected tumor are important. If there is any malignant element that can not be completely eradicated, we would suggest surgical treatment. We believe our process allows to avoid unnecessary surgery and reduces medical costs.

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Address: Giovanni Tomasello

Euro-Mediterranean Institute of Science and Technology (IEMEST), Palermo, Italy

E-mail: giovanni.tomasello@unipa.it