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A case report

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Leiomyoma of the small bowel. A case report

Small bowel leiomyomas are very rare tumors and originate from muscularis mucosae, longitudinal or circular muscular layers. Furthermore, leiomyomas are most common benign tumors of the small intestine. The most frequent location is jejunum. Diagnosis usually is made by CT or endoscope. Tumours can be found accidentally during autopsies or occasionally induce abdominal pain, bleeding or intestinal obstruction and must be treated surgically. To avoid recurrence, wide resection is required.

KEY WORDS: Leiomyoma, Muscularis Mucosa

Introduction

Benign small bowel neoplasms account for 1-6% of all gastrointestinal neoplasms. Leiomyoma occupies the fourth place of small bowel tumors after lymphoma, leiomyosarcoma and adenocarcinoma [6]. Leiomyomas are also reported in Meckel diverticulum or appendix 24. Approximately 50% of cases are found in jejunum, followed by ileum in 30%. The peak incidence is in the fifth – sixth decade of life [1, 4]. Signs and symptoms of small bowel tumors are unspecific and vague. Symptoms are silent many years and can be abdominal pain (66%), bleeding, nausea, intestinal obstruction (8-13%) [6].
Leiomyomas of small intestine present in different age groups and are uncommon benign tumors of small bowel. We present a case of small intestine leiomyoma.

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Case Report

A 28 years old male, with a history of peptic ulcer disease presented to Vilnius University Santaros Clinics Hospital with pain in abdomen for 2 months. Physical examination showed normal limits, a man appeared 173 cm tall, weight was 92 kg, BMI 30,74 kg/m^2.
Gastroscopy showed no pathology. Ultrasonography revealed a large, well-defined, hypoechogenic cystic lesion in lower abdomen and few small irregularly shaped hyperechogenic lesions in the liver.
The patient went to emergency room after pain get worse. General physical examination showed no abnormalities and patient complained of intermittent intense abdominal pain and nausea. Laboratory analysis showed no deviations. Abdominal ultrasound was performed and showed intestinal gas, extended segment of the bowel and a well-defined, non-homogenous cystic lesion (Fig. 1). Abdominal CT revealed a large ~ 118 x 76 x 123 mm in size, solid mass which is intraperitoneally with accumulation of contrast material marginally (Fig. 2). The lesion retracted jejunum, mesentery seen slightly infiltrated and twisted, small amount of free intraperitoneal fluid found. Patient was admitted to Vilnius University Hospital Santaros Clinics (VULSK) and oper-

ation was performed by the Head of Abdominal and Oncosurgery Department prof. K. Strupas and Senior Resident doctor Z. Zidonis. Tumor was 3 metres from ileocecal valve. Proximal to tumor bowels were distended and it was not overgrown mesentery. Tumor was resected and small bowel anastomosis end-to-end was performed. Resected tumor was given for full histopathological examination. More than 6 months later, the patient remains well, with no further symptoms.

Pathology report

During the macroscopic dissection of the resected specimen a large circumscribed intramural 14x7,5x11 cm tumor with pushing border in contact with the ileal mucosa was found. Tumor had a whiteish color and a clearly fasciculated structure (Fig. 5). On microscopic examination an intact mucosa and loose submucosa is

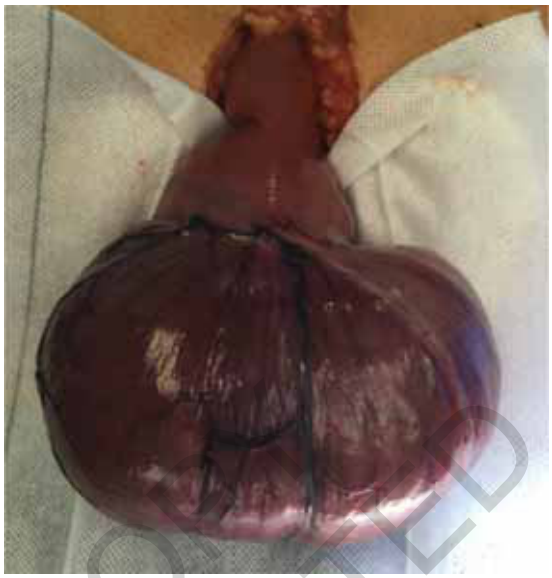

Fig. 3: Tumor in Operating Room.

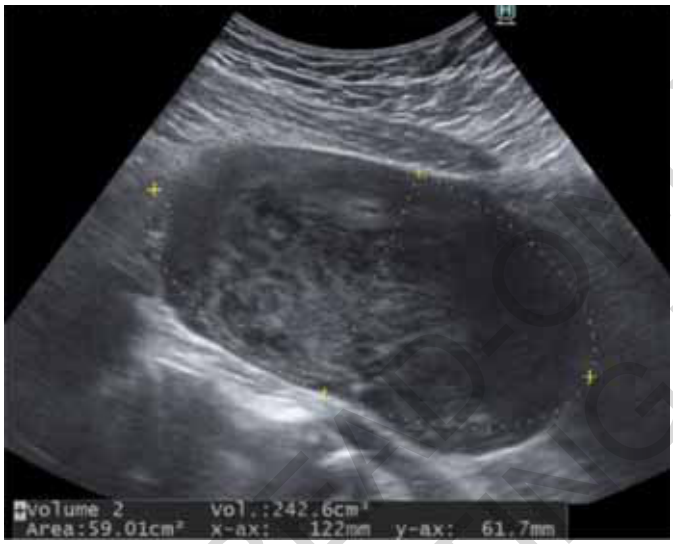

Fig. 1: Ultrasound view of tumor.

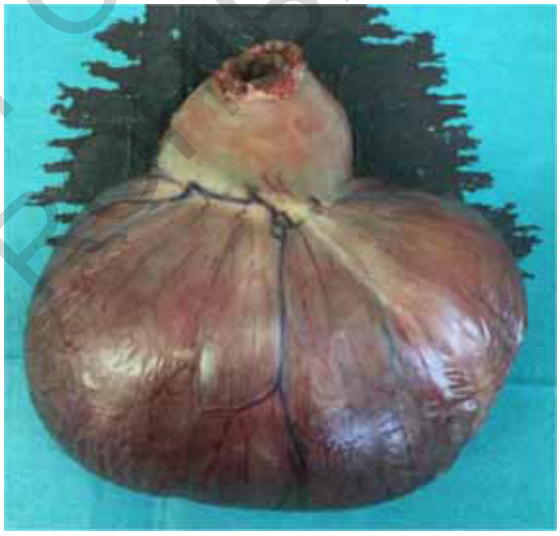

Fig. 4: Tumor after surgery before being sent for histopathological examination (14 x 7 x 11 cm in size).

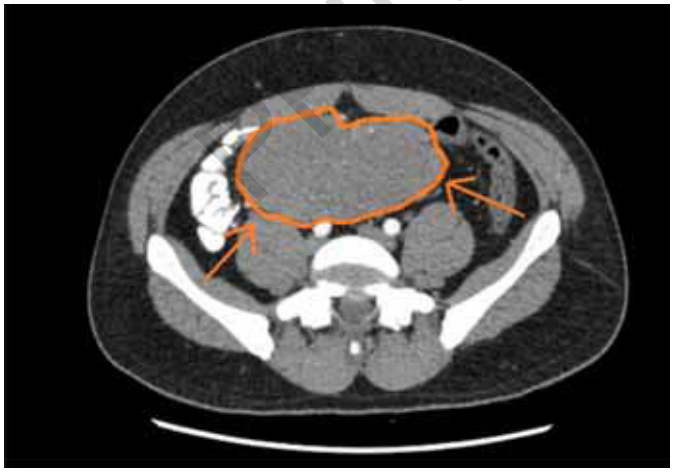

Fig. 2: CT scan (orange arrows show tumor).

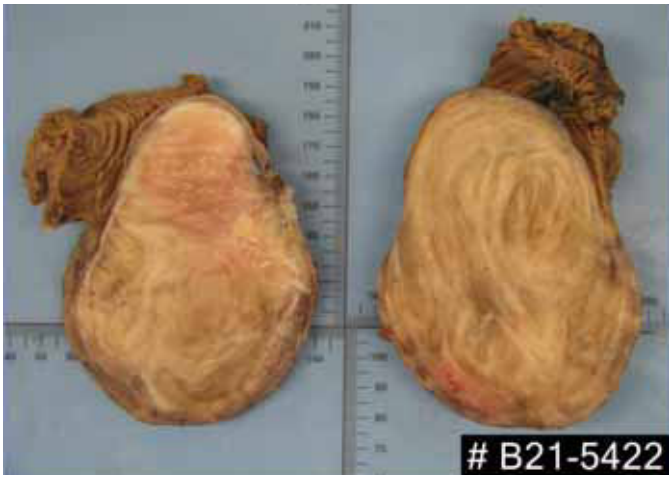

Fig. 5: Macroscopic appearance of the tumor.

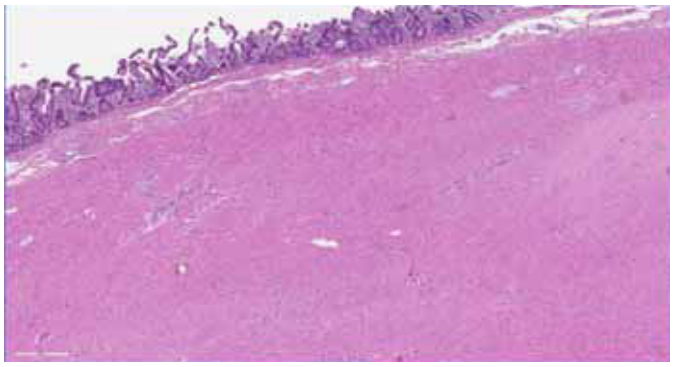

Fig. 6: Intact mucosa and submucosa covering the tumor (hematoxylin-eosin, 10X).

Fig. 8: Tumor is negative for both CD117 and DOG1 (immunohistochemistry, 10X).

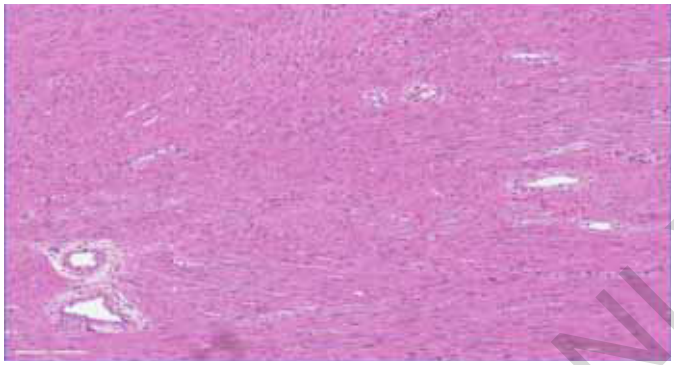

Fig. 7: Fascicles of bland spindle cells with elongated nuclei (hematoxylin-eosin, 20X).

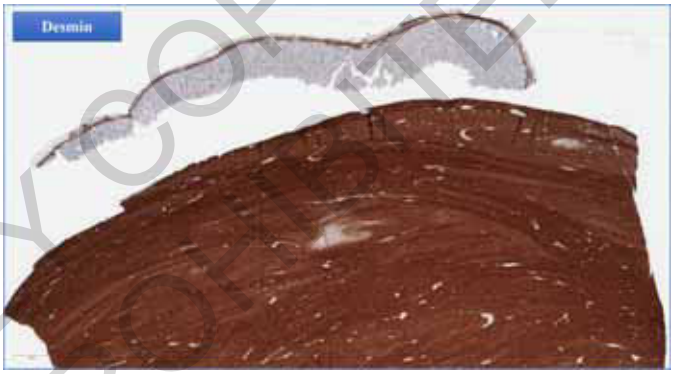

Fig. 9: Diffuse and strong positivity for Desmin (immunohistochemistry, 2X).

covering a low cellularity tumor (Fig. 6) with a well demarcated border, composed of sheets and swirls of bland spindle cells with scant eosinophilic cytoplasm and normochromic elongated nuclei (Fig. 7). There is no significant nuclear atypia or foci of necrosis, mitotic activity is very low (mitotic count: 1 per 50 HPF). Tumor had no reactivity (Fig. 8) with DOG1 and CD117 (that are immunohistochemical markers of gastrointestinal stromal tumors (GIST)), but had a stong and diffuse positivity (Fig. 9) for Desmin confirming myogenic origin of the tissue; Ki67 proliferative activity is very low, below 1%. A final diagnosis of an benign ileal leiomyoma was signed out.

Discussion

Small intestine neoplasms are uncommon in clinical practice. The first ever documented evidence of this kind of tumor in the small intestine dates to 1858 8. Benign small intestine tumors usually are asymptomatic. The stomach is the most common site for leiomyomas in gastrointestinal tract [13].

The most common site for leiomyomas in small intestine is jejunum, followed by ileum and duodenum [2]. These tumors can be intramural, intraluminal, extraluminal and bidirectional [6]. A leiomyoma should also be thought in the differential diagnosis of neoplastic diseases of small bowels if showing increased uptake on FDG-PET/CT [11].

Preoperative diagnosis is difficult because leiomyomas do not occur often and there is no pathognomonic signs. The signs and symptoms of leiomyomas that occur are similar to other gastrointestinal manifestations (e.g. ulcers, diverticulosis) [7], as a consequence of that these benign tumors often go untreated for prolonged periods [4] or frequently diagnosed after failed conservative treatment for a mistaken pathology [7].

The differentiation between small bowel leiomyomas and GISTs cannot be made based on size, anatomic location or gross morphology but instead, requires immunohistochemical and histological analysis [9]. Symptoms that occur are bleeding (59-70 %), abdominal pain (66 %), acute intestinal obstruction (8-13 %). Palpable mass has also been documented and some authors associate leiomyoma with intestinal atresia [3]. Work-up can include fecal occult blood tests, haemoglobin levels, cross-sectional imaging, and endoscopy (gastroscopy, colonoscopy, capsule endoscopy, or push endoscopy) [10].

Differential diagnosis is important before making a

choice of treatment. Barium imaging has limited role in GI tract tumors. Abdominal Ultrasound often shows large masses and heterogeneous reflectivity [17]. GIST's are better seen in computed tomography [18]. As for enhancement pattern, leiomyoma arising in the gastrointestinal tract shows a uniform enhancement pattern reaching a peak in the capillary phase on rapid intravenous contrast bolus CT [19].

In contrast to leiomyoma, leiomyosarcoma is characterized as showing a heterogeneous enhancement pattern and lesions are composed of a central low-density area and peripheral well-enhanced are [20]. Large benign leiomyomas may demonstrate heterogeneous enhancement [19]. Small GISTs usually appear on CT as smooth-walled, sharply margined, homogenous, involving the wall of small intestine and often presenting an intraluminal component [21]. Large GISTs mostly exhibits central areas of haemorrhage or necrosis and mural calcification rarely seen [22]. MRI is supplement to CT, especially in evaluation of large tumors [23]. The definitive diagnosis must be verified with an adequate histopathological study [12].

Immunohistochemical reactions must be performed to reliably differentiate leiomyoma from GIST. Leiomyomas are negative for CD117, but positive for desmin and smooth muscle actin [4] and we should be careful that a considerable number of GISTs, especially small bowel GISTs, are also positive for smooth muscle actin.

Over 90% of gastric GISTs are positive for CD34, but approximately half of GISTs other than gastric GISTs are negative for CD34.

A considerable number of IFPs are positive for CD34, and some of other GI mesenchymal tumors such as Schwannomas and leiomyomas might show partial and/or weak staining for CD34 [14]. Gastrointestinal stromal tumours (GIST) are identified by stain positive CD117 (C-Kit) and DOG1 [15,16].

The treatment for leiomyoma is surgery. Because of leiomyomas have tendency to recur locally, wide resection is mandatory [3]. Lymphadenectomy is not recommended because of a low risk of lymph node metastasis [12].

Prognosis of leiomyomas after surgery is excellent [25].

Riassunto

I leiomiomi dell'intestino tenue sono tumori molto rari e originano dalla muscolaris mucosae, strati muscolari longitudinali o circolari. Inoltre, i leiomiomi sono i tumori benigni più comuni dell'intestino tenue. La posizione più frequente è il digiuno. La diagnosi di solito viene effettuata mediante TC o endoscopia. I tumori possono essere trovati accidentalmente durante le autopsie o occasionalmente inducono dolore addominale, sanguinamento o ostruzione intestinale e devono essere trattati chirurgicamente. Per evitare recidive, è necessaria un'ampia resezione.

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