Leiomyoma of the gallbladder. An extremely uncommon diagnosis. A case report.

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CASE REPORT

GENERAL SURGERY

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Abstract: Gallbladder mesenchymal neoplasms are rare, and the majority of these represent sarcomas of different types of histology. To our knowledge, the English literature has reported only a few patients with multiple Epstein-Barr virus (EBV)-associated gallbladder smooth muscle tumors in the immunodeficiency setting, but no single case of conventional leiomyoma has been well documented to date. A case of a healthy 41-year-old man with gallbladder leiomyoma is described here. Incidentally, the tumor was detected during a routine ultrasound examination, and was removed by simple cholecystectomy. At last follow-up the patient is alive and well, 6 months after surgery. Histology and immunohistochemistry were consistent with a very similar benign smooth muscle neoplasm to conventional uterine leiomyoma. For both EBV-encoded nuclear RNAs and latent membrane antigen EBV, the tumour was negative. The patient did not have any history other neoplasms or evidence of an immune deficiency. Leiomyoma should be included in the differential diagnosis of gallbladder spindle cell tumors and should be distinguished from leiomyosarcoma and the rare gastrointestinal stromal neoplasms recently reported at this unusual anatomical site.

Keywords
Gallbladder mesenchymal neoplasm, leyomioma, gallbladder leyomioma.

Introduction

Mesenchymal gallbladder neoplasms are sporadic; individually, very few gallbladder leiomyoma cases have been described, and all of them are diagnosed in patients with diseases of the immune system. A case report of a vesicular leiomyoma is described in a 41-year-old male patient without immune compromise. The patient lacked a significant past medical history and presented to the general surgery outpatient clinic with clinical data of acute cholecystitis and an ultrasound that reported cholelithiasis with no acute findings. It was sent to the emergency department for evaluation. Laboratory studies were performed, and the abdominal ultrasound was repeated, persistent symptoms were the primary indication to perform a laparoscopic cholecystectomy. He was discharged after clinical improvement after five days of hospital stay. Subsequent immunohistochemical study of the gallbladder specimen yielded the diagnosis of leiomyoma of the gallbladder fundus, with a positive SMA marker.

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Case report

41-year-old male patient, with no significant past medical history presented with intermittent right upper quadrant colic pain, occasionally accompanied by vomiting that spontaneously subsided. He was initially attended an outpatient general surgery clinic where an abdominal ultrasound was performed reporting non-acute cholelithiasis. It was decided to send him to the emergency department due to clinical exacerbation data with mild right upper quadran pain. Laboratory studies reported no relevant data. The patient was scheduled for an emergent cholecystectomy due to exacerbation of symptoms. Transoperative findings were consistent with thickened gallbladder wall, firm adhesions to the omentum, and the liver. **(Figure 1)** Puncture decompression of the gallbladder revealed purulent leakage material. Clinical deterioration with persistent tachycardia precluded an optimal postoperative evolution. A complete blood count listed a decrease in hemoglobin levels to 7 gr/dL. An abdominal ultrasound reported free fluid in the parietocolic gutter and in the space of Morrison. The patient was re-scheduled for surgical exploration. 500 cc of hemoperitoneum were found in the form of abundant clots, without an active bleeding site. Abdominal lavage with the placement of drains was performed. The evolution of the patient was

uneventful and he was discharged on the fifth day of hospital stay. The subsequent pathology analysis and immunohistochemical study of the surgical piece yielded the diagnosis of a 1.7 cm background gallbladder leiomyoma, with a positive SMA marker, KI-67, desmin, CD-34, and CD-117 negative. **(Figure 2)**

Discussion

The leiomyomas are benign tumors of smooth muscle, commonly originate in the female genital tract (95%), but can develop anywhere in the presence of smooth muscle fibers. Microscopic smooth muscle fibers with a fascicular pattern with eosinophilic cytoplasm and an elongated nucleus are observed microscopically. Immunohistochemically, the smooth muscle differentiation markers expressed are SMA, desmin, and h-Caldesmon, and are commonly negative for CD34, CD117, and S100 (1). In a study carried out in Chile by Torres-Q. et al., 5,699 biopsies were evaluated from 1998 to 2007, of which 4.9% reported benign tumor lesions, the most frequent being gallbladder polyp (45.5%) and a only one case of leiomyoma was found (0.4%) (2). So far, only one case of solitary vesicular leiomyoma has been described in the English literature in previous studies of benign gallbladder neoplasms, and only 3 cases in which a systemic leiomyomatosis in the context of infection by Epstein Barr virus-associated with immunodeficiency due to HIV, severe combined immunodeficiency and Wiskot-Aldrich syndrome (3).

The frequency of benign mesenchymal gallbladder tumors remains unknown. The incidence of gallbladder sarcomas is estimated to be around 2%. The study of the tissue by biopsy (usually after performing a cholecystectomy) is necessary to establish the definitive diagnosis and thus exclude the possibility of leiomyosarcoma and other sarcomas. Therefore, leiomyoma should be taken into account in the differential diagnosis of gallbladder tumors, not only in immunosuppressed patients but also in patients with normal immune function (3).

Gallbladder leiomyoma differential diagnosis involves leiomyosarcoma, and GIST (1). The second most common type of sarcoma found in the gallbladder is leiomyosarcoma (4). Unlike leiomyosarcoma, leiomyoma lacks nuclear atypia, pleomorphism, increased mitotic activity, and necrosis, so leiomyosarcoma differentiation isn't difficult. Epstein-Barr virus (EBV)-associated leiomyosarcoma / smooth muscle tumor occasionally involves the gallbladder in either an acquired or an iatrogenic immunosuppression setting (5,6). This type of tumor is microscopically characterized by the proliferation of oval to spindle-shaped mesenchymal cells, as well as positive and negative immunoreactivity for alpha-smooth muscle actin and desmin, respectively (6). EBER 1-positivity by in situ hybridization is also a characteristic feature (6). In addition, GIST of the gallbladder is rarely reported (4), and metastatic GIST should be also included in the differential diagnostic considerations,

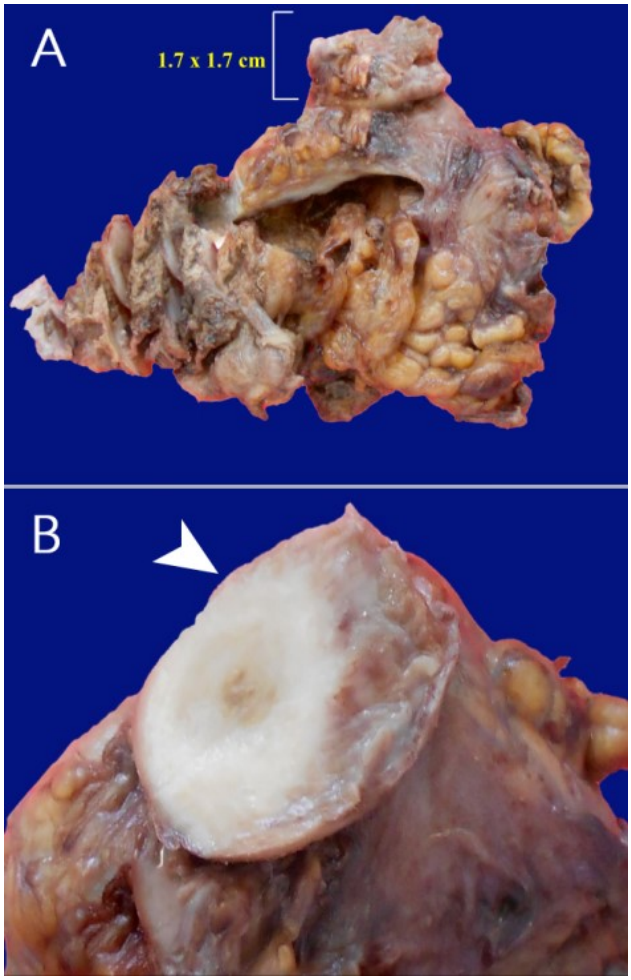

Figure 1: *A.* Gross pathology specimen of the gallbladder. A 1.7 cm diameter mass is found on the subserosal space. *B.* Sharply circumscribed, round, firm, grayish white, "raw silk" and whorled cut surface (white arrowhead).

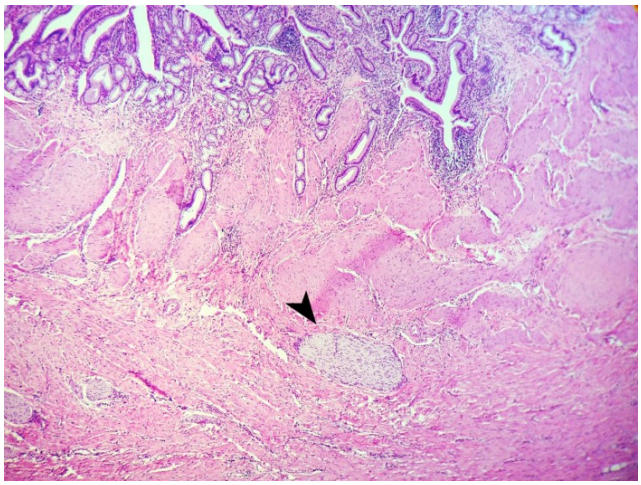

Figure 2: Whorled (fascicular) pattern of smooth muscle bundles separated by well vascularized connective tissue (black arrowhead). Elongated smooth muscle cells with eosinophilic or occasional fibrillar cytoplasm and distinct cell membranes

as in the present case. Although both leiomyoma and leiomyosarcoma may occasionally mimic GIST in morphology, the immunohistochemical characteristics of GIST can easily differentiate leiomyoma (CD117 negativity and desmin positivity) from GIST (CD117 positivity and desmin negativity) (6).

In conclusion, we report a case of leiomyoma of the gallbladder. Leiomyoma of the gallbladder is extremely rare; however, it may be an underrecognized entity. It is important to differentiate leiomyoma from GIST to avoid unnecessary long-term clinical follow-up and treatment.

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