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Diagnosis and management of a pelvic solitary fibrous tumor in a postmenopausal woman – a case report

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ABSTRACT

Background: Solitary fibrous tumors, previously known as hemangiopericytomas, originate from mesenchymal tissue and can occur at many body sites, such as the thorax, head and neck, retroperitoneal space and abdomen. These tumors are generally rare and pelvic location is extremely uncommon. Consequently, pelvic solitary tumors could be mistaken for ovarian cancer in menopausal women. This report presents a case of pelvic solitary tumor to highlight the importance of considering this diagnosis in a postmenopausal woman presenting with a solid pelvic mass, normal tumor markers and no ascites.

Case: A 54-year-old woman presented with amenorrhea, abdominal pain, constipation, nausea, vomiting, and frequency of urination. On examination she had a pelvic mass of approximately 20–24 weeks in size. Ultrasound and computed tomography imaging showed a well-defined, round, centrally hypodense, irregular thick and peripheral, enhancing solid mass originating from the left ovary. Carcinoembryonic antigen, carbohydrate antigen-125, and carcinoembryonic antigen 19–9 were all normal. Intraoperatively the tumor was attached to the peritoneum and mesentery. Part of the large bowel, including the sigmoid colon, were attached to it. The exact origin of the tumor could not be ascertained during surgery. The tumor was successfully excised, and specimen sent for histology and immunochemistry analysis. The definitive diagnosis was confirmed with immunochemistry. The patient had an uneventful postsurgical course and was discharged on day 4 after surgery for routine gynecological follow-up.

Conclusion: Solitary fibrous tumor is very rare; however, the diagnosis should be considered in a postmenopausal woman with solid pelvic mass, normal tumor markers and no ascites.

1. Introduction

Solitary fibrous tumors (SFTs) are fibroblastic tumors that originate from mesenchymal tissues. They can be found anywhere in the body, but most commonly in the thorax, head and neck, and retroperitoneal space abdomen. These tumors are rarely diagnosed in the female genital tract [1] [2]. The first SFT was reported by Klemperer and Rabin in 1931 [3]. Since then, several case reports have been published; however, there are very few reports on pelvic STFs in postmenopausal women. While considered to be benign, recurrence can occur in approximately 60% of patients after complete excision [4]. Furthermore, SFTs have malignant potential, although the reported risk of malignancy is very wide ranging, from 7% to 60% [5]. The risk of malignancy increases with larger and recurrent tumors [6] [7]. This is a case report about a postmenopausal woman who presented with a symptomatic large pelvic mass which was later confirmed to be an SFT. The case demonstrates the need to consider this diagnosis in a postmenopausal woman presenting with a solid pelvic mass, normal tumor markers and no ascites, as well as the importance of requesting immunochemistry analysis in such situations.

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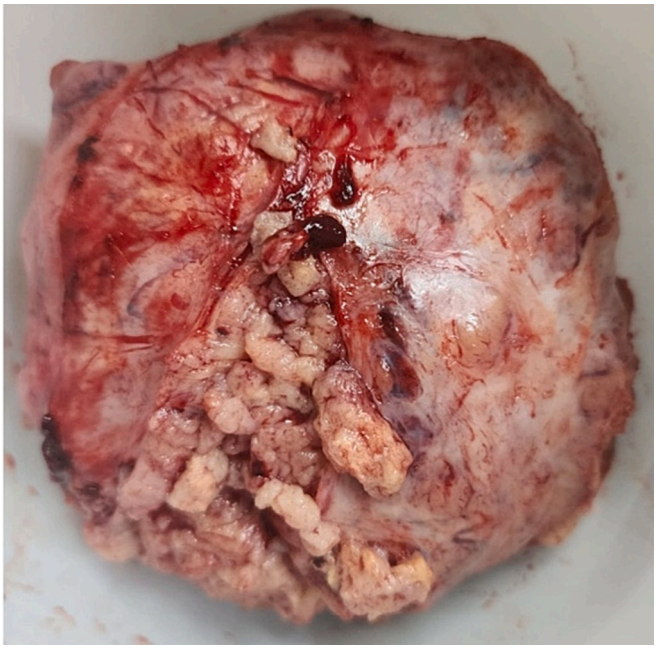

Fig. 1. Macroscopic appearance of the tumor.
Macroscopically, the mass, weighing 1045 g, is encapsulated with a yellowish-white solid cut surface and measured. The tumor is a well-circumscribed mass and is surrounded by a fibrous capsule.

2. Case Presentation

A 54-year-old woman was admitted with a pelvic-abdominal mass that required surgery. The patient complained of lower abdominal pains, frequent urination, nausea and vomiting, constipation, bloating, progressive abdominal distention with pains and a five-year history of amenorrhea. These symptoms had been present for over 12 months. She had no personal or family history of ovarian or breast cancer but her father had had colon cancer. Physical examination revealed a mass approximately 20–24 weeks in size coming out of the pelvis. The mass was felt to be pressing on the rectum on rectal examination.

Ultrasound and computed tomography imaging suggested that the mass originated from the left ovary and measured 121 mm × 120 mm × 138 mm. There were no calcifications, septae or ascites. The mass was described as round, well defined, irregularly thick, peripheral enhancing, with a centrally hypodense, mildly progressive centripetal filling on the delayed phase. Both uterus and right ovary were reported as normal, and a PAP smear done in the gynaecology outpatient department prior to admission was normal.

The working clinical diagnosis was of a left ovarian mass possibly due to malignant ovarian germ cell tumors (choriocarcinoma, dysgerminoma, yolk sac tumor) and malignant epithelial ovarian neoplasm; however, in the absence of septae and ascites, the latter was considered to be less likely. Tumor markers, carcinoembryonic antigen (CA), carbohydrate antigen-125 and CA 19–9 were normal (1.3 g/L, 14kU/L < 1kU/L respectively). Luteinizing hormone (LH), follicle-stimulating hormone (FSH) and estradiol were 42.4. IU/L, 58.4 IU/L and < 19 pmol/L, in keeping with the history of menopause. Intraoperatively the mass was found to have been attached to the peritoneum, mesentery large bowel and sigmoid colon; however, the exact origin of the tumor could not be established as both ovaries and uterus appeared normal. The tumor was completely excised, and specimen sent for histopathology and immunochemical analysis.

Macroscopically (Fig. 1), the mass weighed 1045 g, measured 1.10 mm x119mm x 105 mm, was surrounded by a fibrous capsule and had a yellowish-white solid cut surface. On microscopic examination (Fig. 2, A to D), the tumor was made up of haphazardly arranged plump spindle cells with densely cellular and looser, hypocellular areas. The tumor cells had small, hyperchromatic nuclei with moderately pale eosinophilic cytoplasm. Mild nuclear pleomorphism was also noted. There were four mitotic figures per ten high-power fields (HPF). A single area of tumor necrosis was visible. The stroma was collagenous and contained prominent branching vessels. Some of the vessels had a "stag

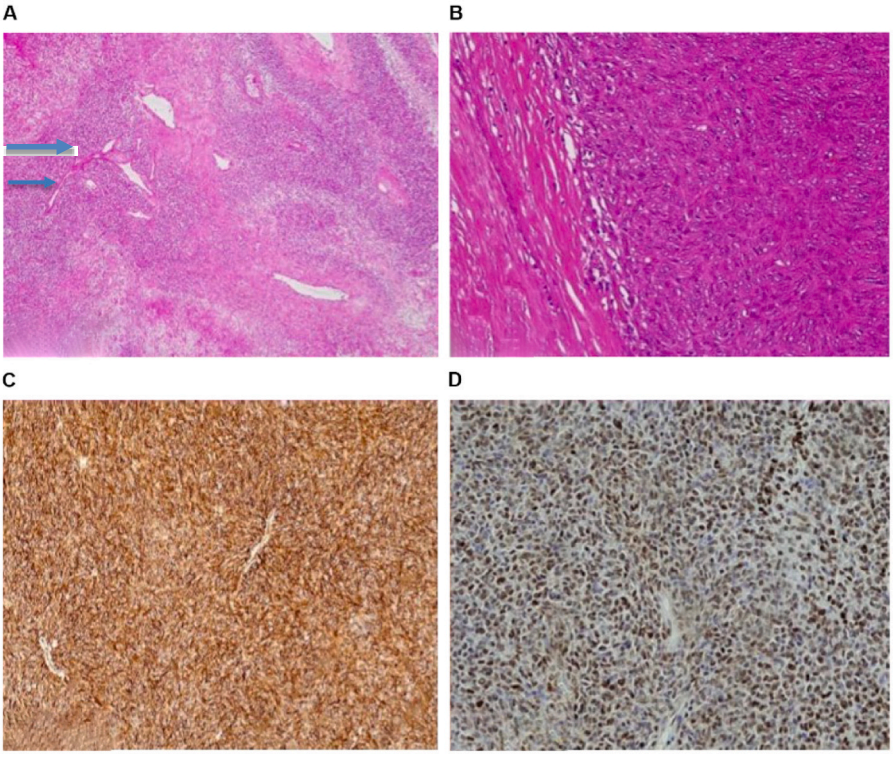

Fig. 2. Histopathology images.
A: Photomicrograph showing the haphazard arrangement of tumor cells with prominent staghorn vasculature (arrow) [haemotoxylin and eosin (H&E), 40×].
B: Photomicrograph of tumor at a high magnification showing an encapsulated spindled cell proliferation with uniform vesicular nuclei and eosinophilic cytoplasm (H&E, 200×).
C: Immunohistochemical staining for CD34 showing strong membranous staining (100×).
D: Immunohistochemical staining for STAT-6 showing diffuse, strong nuclear staining (200×).

horn" appearance. There was no heterologous differentiation, lymphovascular or perineural invasion. On immunohistochemistry, the tumor tested positive for both CD34 and STAT6 and negative for DOG-1, CD117, SMA, S100, desmin, and calretinin. The above features were in keeping with the diagnosis SFT with a score of 4–5 (equivalent to intermediate risk). Based on the score, the patient was considered to have been fully treated. She had an uneventful postsurgical course and was discharged on day 4 after surgery for follow-up at the gynaecology outpatient.

3. Discussion

SFTs are spindle cell tumors of mesenchymal cell origin. They are generally considered benign. These tumors are very rare and represent <2% of all soft-tissue tumors [6]. Because of their mesenchymal origin they can occur at any part of the human body; however, the majority of these tumors occur in the chest [8]. Pelvic tumors are extremely rare and when they occur in postmenopausal women they can be mistaken for ovarian malignancy. This could result in a staging laparotomy and consequently increased morbidity and long hospital stay.

Preoperative diagnosis of SFT is very difficult because of a lack of specific symptoms, radiological features and tumor makers. SFTs can be confused with ovarian malignancy especially when they present in the genital tract of a postmenopausal woman. Similarly, patients with ovarian cancer also present with a pelvic mass and non-specific symptoms. However, as in this case, the diagnosis of ovarian malignancy can be excluded if tumor markers are normal together with intraoperative findings of normal ovaries and uterus. The definitive diagnosis of STF is based on distinctive immunohistochemical features, specifically, the expression of CD34 and STAT6 by the tumor cells [9] [10]. Of the two markers, CD34 is the more common and is expressed in 79% of cases [11,12]. However, STAT6, a nuclear marker, has shown high sensitivity (98%–100%) and specificity (100%) for SFT independent of anatomical location and tumor morphology, hence the need to request both markers when sending specimens for histopathology and histochemistry [13].

4. Conclusion

This case illustrates the importance of including SFT in the differential diagnosis of a solid pelvic mass in postmenopausal woman with normal tumor markers and no ascites. Furthermore, the case highlights the importance of requesting histochemistry analysis, specifically CD34 and STAT6, in order to arrive at a definitive diagnosis. Surgical excision is the mainstay of treatment; however, these patients require close follow-up because of the risk of recurrence.

Contributors

Constant Ndjapa-Ndamkou was involved in the diagnosis and management of the patient, and wrote and reviewed the manuscript.

Sharol Ngwenya conducted histological and immunochemistry analysis, diagnosed the solitary fibrous tumor, submitted microscopic images, checked the pathology sections, and reviewed the manuscript.

Dimitrije Mamontov conducted histological and immunochemistry analysis, diagnosed the solitary fibrous tumor, submitted microscopic images, checked the pathology sections, and reviewed the manuscript.

Langanani Mbodi was involved in the management of the patient, writing of the initial draft, and reviewed the manuscript.

Logie Govender was involved in writing of the draft, and reviewed the manuscript.

Thifhelimbilu Luvhengo was involved in the diagnosis and management of the patient, writing of the first draft, and reviewing the manuscript.

Lawrence Chauke was involved in the diagnosis and management of the patient, and revised and reviewed the manuscript.

All authors saw and approved the final manuscript.

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Patient consent

The patient provided written informed consent for the publication of this case report. The case report received ethic clearance from Human Research Ethics Committee (HREC) of the University of the Witwatersrand (Clearance certificate No. M230383).

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This article was not commissioned. Peer review was directed by Professor Margaret Rees Editor-in-Chief, independently of Lawrence Chauke, an editorial board member of *Case Reports in Women's Health*, who was blinded to the process.

Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

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