An Abdominal-sacral Approach With Preoperative Embolisation For Vulvar Solitary Fibrous Tumour: A Case Report

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Case report

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Abstract

Background: Solitary fibrous tumours (SFTs) in the female genital tract are uncommon. Resection of these tumours is controversial because it can cause life-threatening haemorrhage. We report a case of vulvar SFT that was excised in a combined abdominal-sacral approach after preoperative embolisation.

Case Presentation: At another hospital, an inoperable intrapelvic tumour was diagnosed in a 34-year-old woman. Computed tomography and magnetic resonance imaging showed that the uterus, urinary bladder and rectum were compressed laterally by a pelvic tumour with a maximum diameter of 11 cm. This mass was hypervascular and had a well-defined border. Transperineal biopsy was performed, and immunostaining revealed that the mass was an SFT. The tumour was supplied by feeding vessels from the right iliac arteries. First, we embolised the feeding vessels. Second, we performed surgical resection in a combined abdominal-sacral approach; no blood transfusion was necessary, and no perioperative complications occurred. The final pathological diagnosis was SFT that was positive for CD34 and signal transducer and activator of transcription 6 according to immunohistochemical staining.

Conclusion: During a year of follow-up, the disease did not recur. Treatment of pelvic SFT should aim at complete resection through various approaches after careful measures are taken to prevent haemorrhage.

Background

Solitary fibrous tumours (SFTs) were first described by Klemperer and Rabin in 1931 as mesenchymal tumours of the pleura [1]. Although SFTs are commonly considered intrathoracic tumours, approximately 30% of them arise in various extrapleural sites [2, 3]. Of the extrapleural SFTs, those in the female genital tract are rare. This tumour is also characterised by low potential for malignant transformation and by abundant blood vessels. Surgical excision with curative intent is generally recommended for the management of this tumour, but controlling bleeding during the operation is often difficult [4, 5]. Surgical methods are controversial because tumour resection sometimes causes life-threatening haemorrhage.

We succeeded in complete en bloc resection of vulvar SFT, without morbidity or the need for blood transfusion, by a combined abdominal-sacral approach after embolisation of the vessels supplying blood to the tumour.

Case Presentation

A 34-year-old woman was referred to our hospital to evaluate an asymptomatic pelvic mass detected with transvaginal ultrasonography in a private clinic, which she had visited for treatment of infertility. Computed tomography revealed a 112 × 62 × 58 mm hypervascular mass with a well-defined border. This mass compressed the bladder, uterus and rectum in the peritoneum (Fig. 1a). Subsequent contrast medium–enhanced and fat-suppressed T1-weighted magnetic resonance imaging (MRI) then revealed tensity, and T2-weighted images showed a mixture of

isointensity in the muscles and high intensity of the tumour, as well as compression of the bladder, uterus and rectum (Fig. 1b and 1c). Laboratory data revealed no abnormalities such as squamous cell carcinoma antigen, carcinoembryonic antigen, cancer antigen 125 or carbohydrate antigen 19 – 9. We performed a transperineal biopsy, the results of which established the diagnosis of SFT.

Before surgery, we performed embolisation of the feeder vessels to reduce intraoperative blood loss because this SFT was hypervascular, supplied primarily by the right obturator artery and the internal pudendal artery, according to angiography (Fig. 1d). The next day, with the patient in the supine position, we separated the tumour from the right side of the rectum and uterus through a transabdominal approach, which would have been challenging in a narrow and deep pelvis. Laparotomy was performed through a midline incision. We approached the paravesical space and confirmed that the tumour invaded the retroperitoneal cavity. After the wound was closed, the patient was repositioned into the jackknife position for resection through the sacral approach. We made a paramedian skin incision and easily identified the elastic but hard tumour (Fig. 2). We ensured the adequacy of surgical margins to prevent local recurrence and minimise bleeding. The tumour was completely excised over a period of 223 minutes with 250-mL blood loss, and no blood transfusion was required.

Postoperative pathological study showed that the tumour was encapsulated and elastic but hard, and the cut surface was greyish-white (Fig. 3. Microscopic study revealed that the tumour consisted of proliferating, relatively small oval and spindle cells with prominent branching and a hemangiopericytoma-like vascular pattern. Cytological atypia was not significant. Immunohistochemical staining revealed that the tumour cells were positive for CD34 and for signal transducer and activator of transcription 6 (Fig. 4). The final diagnosis was also SFT. The postoperative course was uneventful; no adjuvant treatment was given because complete surgical resection was achieved. In the year since surgery, the patient has shown no evidence of tumour recurrence.

Discussion And Conclusions

In this case, a solitary fibrous tumour invading the retroperitoneum from the vulva was treated successfully by a combined abdominal-sacral approach after embolisation, without a need of blood transfusion. To our knowledge, this is the first case report of the resection of a vulvar SFT through a combined abdominal-sacral approach.

SFTs are rare soft tissue tumours that commonly arise in the pleura [1]. Such tumours rarely arise from the female genital tract, although they have been reported in various other organs. Nine percent of SFTs occur in the female genital tract, and only 42 cases, including several in the retroperitoneum, have been reported so far [5, 6]. Furthermore, only 11 cases of vulvar SFTs have been reported [7]. The management of vulvar SFTs is controversial: The prognosis depends on complete resection of both extrapleural and pleural SFTs [5], but surgery is difficult because of frequent intraoperative heavy bleeding, which occurs because SFTs in the pelvis are usually supplied with blood by multiple vessels, such as the branches of

the inferior mesenteric artery or the internal iliac arteries [8, 9]. Therefore, it is necessary not only to ensure a sufficient blood supply but also to control bleeding during surgery.

Because the tumour extended from the vulva into the pelvis, we performed the surgery through a combined abdominal-sacral approach out of concern about the difficulty in establishing an appropriate surgical field deep inside the pelvis by laparotomy. For that reason, we first separated the tumour from the right side of the rectum and uterus through a transabdominal approach and then successfully resected the tumour through a transsacral approach. Most patients with pelvic SFTs have undergone laparotomy, but some patients suffer heavy bleeding, which is difficult to control [9, 10]. In one report, massive bleeding was not avoided even with the transperineal approach [11]. Katsuno et al. reported that the transsacral approach was useful for complete resection of pelvic SFTs [12]; however, this approach carries a high risk of postoperative complications, such as surgical site infection and anal dysfunction [13]. In another report, surgery with a combined abdominal-sacral approach was performed for five cases of giant presacral tumours, and complete resection without massive bleeding was achieved. The advantages of this approach are that complications are minimised and it allows for complete resection of a tumour that may be difficult to remove through other approaches [14]. Thus a combined abdominal-sacral approach can be an option for resecting tumours deep in the pelvis.

We embolised feeder vessels to the tumour before surgery to reduce intraoperative bleeding. Preoperative percutaneous arterial embolisation allows for safe and complete resection in cervical, thoracic and lumbar locations in the spinal cord [15, 16]. Embolisation for pelvic SFT has been reported; Soda et al. reported that a tumour was resected after blood flow block was achieved by an intraoperatively inserted aortic balloon catheter, and the resulting blood loss was 13,660 mL [9]. On the other hand, in other reports, the feeder vessels of SFTs were selectively embolised before operation, which resulted in less intraoperative blood loss without the need for blood transfusion [11, 17, 18]. In addition, in two reports, preoperative embolisation did not have the effect of shrinking the tumour [17, 18]. In our case, it was possible to complete surgery without blood transfusion by performing preoperative embolisation. Therefore, embolisation may control intraoperative bleeding, but it is not effective in reducing tumour volume. In addition, selective embolisation of the feeding vessels is more appropriate than intraoperative aortic occlusion.

We completely resected a vulvar SFT without blood transfusion. This tumour is very rare, nonmetastatic and characterised by abundant blood vessels. The main treatment for SFTs is surgical resection. However, pelvic SFTs carries the risk of massive bleeding and organ damage, and inadequate tumour resection can lead to local recurrence. Preoperative embolisation of feeder arteries reduced intraoperative bleeding in our patient. In addition, use of the abdominal-sacral approach can reduce perioperative complications. This combination thus has potential in the treatment of pelvic SFTs.

List Of Abbreviations

MRI – Magnetic resonance imaging

SFT = Solitary fibrous tumours

Declarations

Ethics approval and consent to participate

Consent was obtained from the patient for participation in this study.

Consent for publication

We obtained the patient's consent for publication of this case report.

Availability of the data and materials

All the data are available in the patient's medical record.

Competing interests

The authors have no conflicts of interest relevant to this article.

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Not applicable.

Authors' contributions

AT was responsible for this patient's operation, conducted a literature search and drafted with the manuscript. RK, TA, HN, MD and FY were involved in the gynaecological management of the patient. SM was involved in the pathological diagnosis of the mass. FK and TM contributed to the manuscript review. AT and RK wrote the final version of the manuscript. All authors read and approved the final manuscript.

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Figures

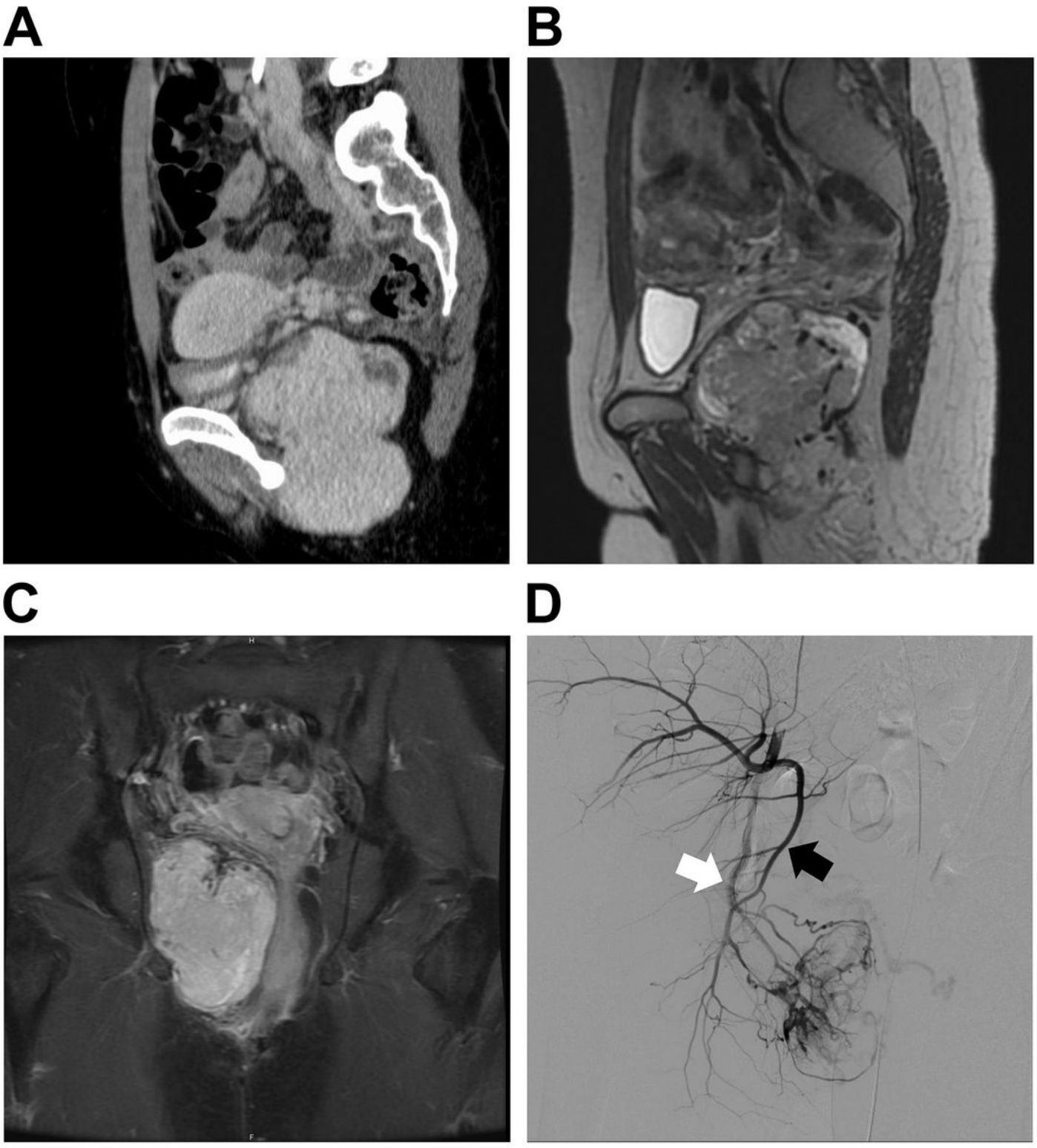

Figure 1

Preoperative imaging findings (a) Preoperative contrast medium–enhanced sagittal computed tomography showed a pelvic mass lesion, measuring 112 × 62 × 58 mm. (b) T2-weighted sagittal magnetic resonance imaging (MRI) showed a mass with heterogeneous intensity in the pelvic cavity. (c) Fat-suppressed contrast medium–enhanced T1-weighted coronal MRI showed a tumour with a relatively avity. (d) On angiography of the right iliac artery, the solitary

fibrous tumour was found to be supplied by the right obturator artery (black arrow) and the right internal pudendal artery (white arrow).

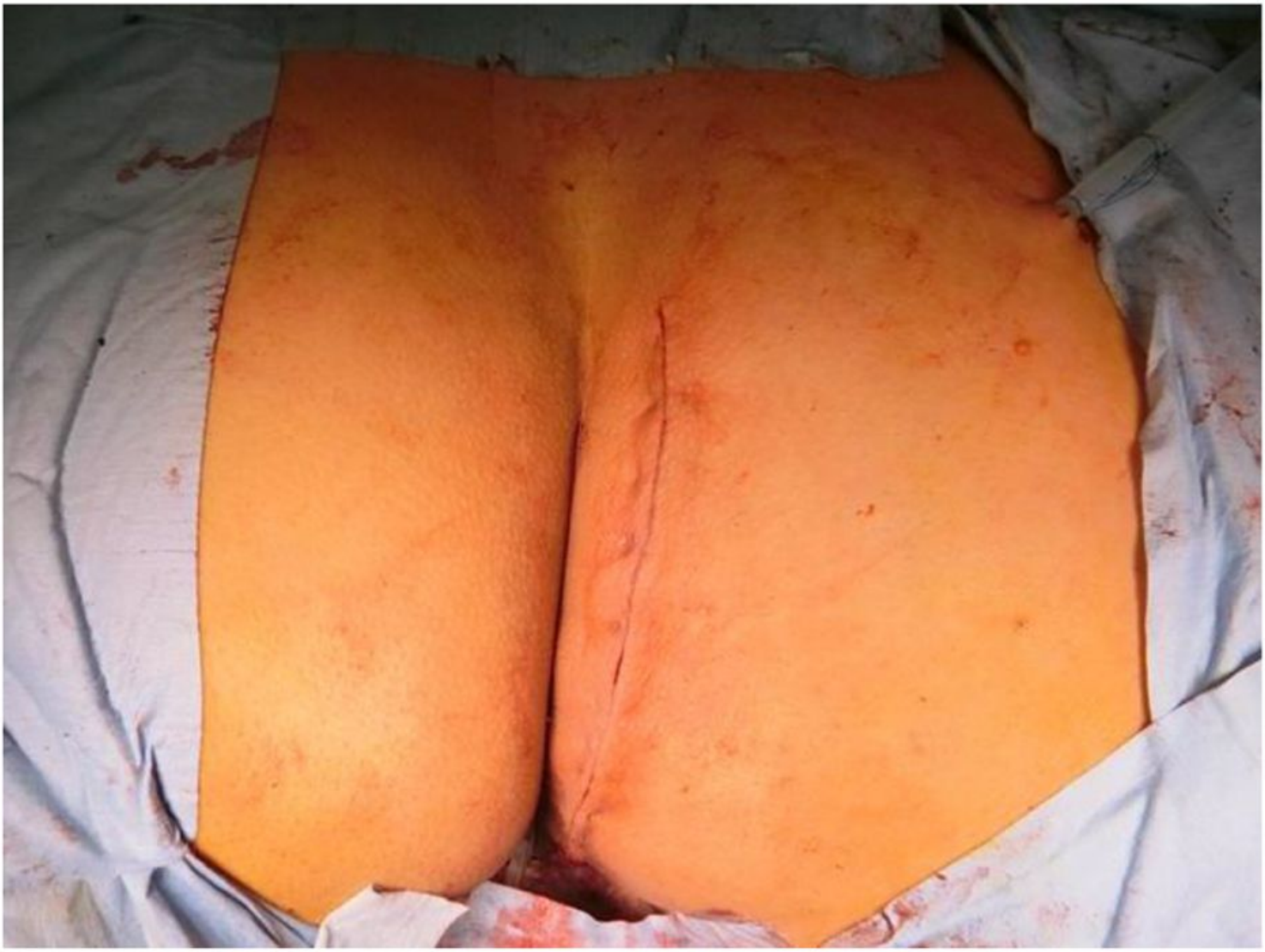

Figure 2

Skin incision For tumour resection, the patient was in a jackknife position, and the lateral paramedian incision was made in the skin.

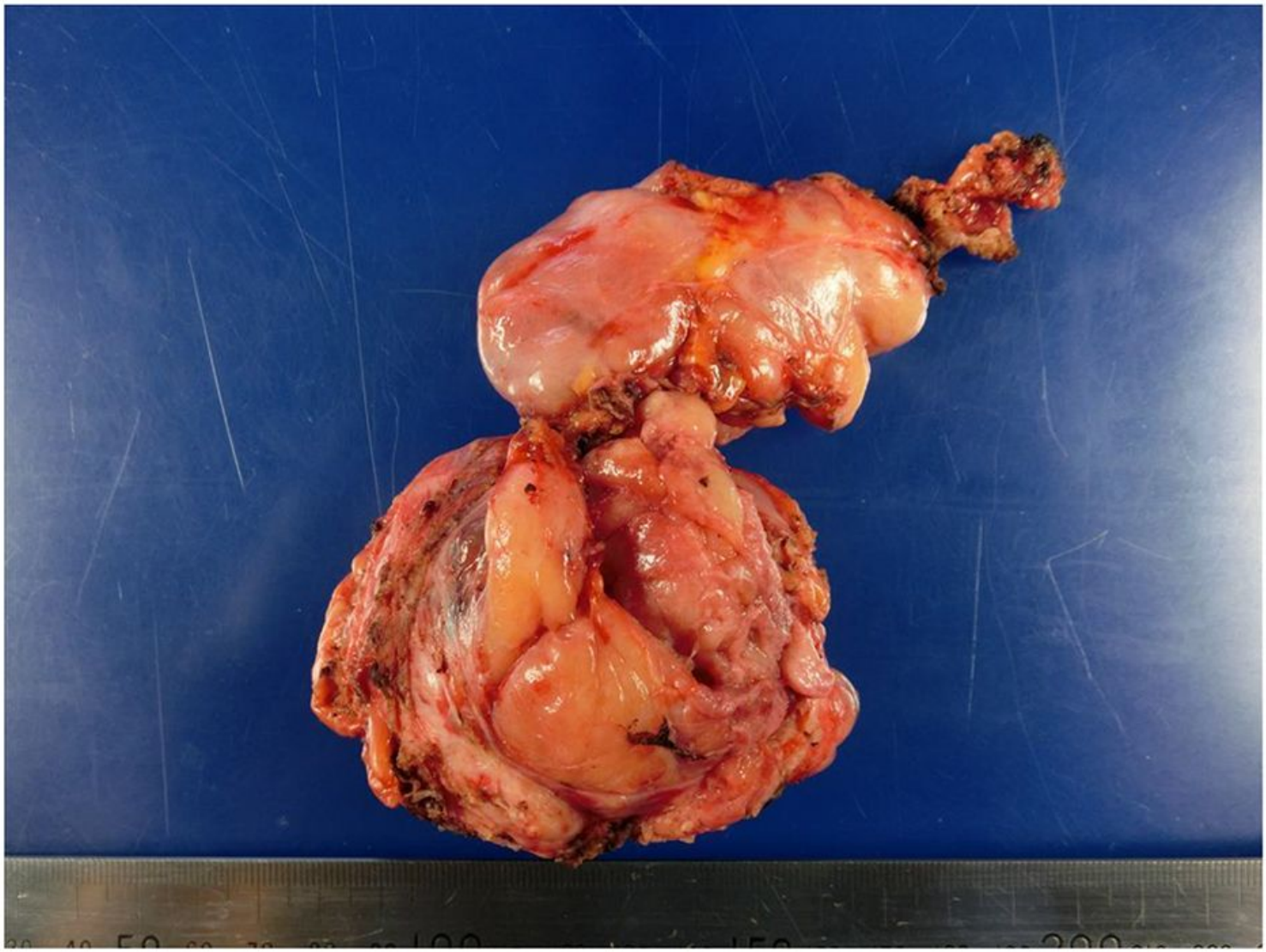

Figure 3

Gross of solitary fibrous tumour Photograph of the tumour. Macroscopically, the tumour was elastic but hard, with an intact capsule and the cut surface was greyish-white.

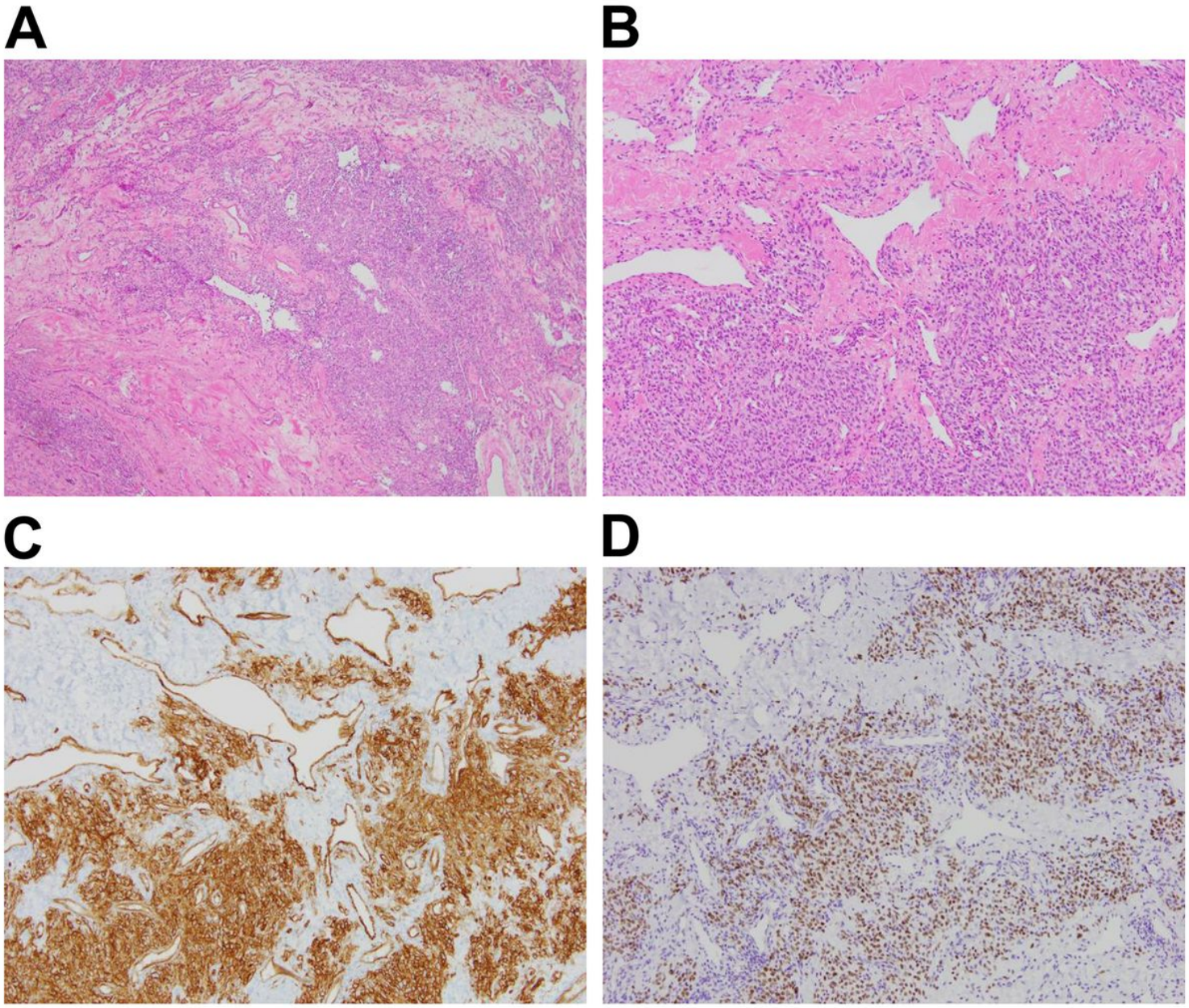

Figure 4

Microscopic histological findings (a, b) Microscopic findings showed spindle cells with a patternless growth arrangement and enlarged blood vessels. ((a)Magnification, ×40, (b)Magnification, ×100 ; haematoxylin and eosin stain.) (c) The tumour cells were strongly positive for CD34. (Magnification, ×100.) (d) The tumour cells were positive for signal transducer and activator of transcription 6. (Magnification, ×100.)

Supplementary Files

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