Giant Pleomorphic Lipoma With Malignant Transformation in the Buttock: a Rare Case Report and Literature Review

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Case report

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Abstract

Background: Pleomorphic lipoma is a benign tumor, which is often located in the neck, shoulder, and other subcutaneous tissues. Pleomorphic lipoma with malignant transformation in the buttock is rarely reported.

Case Presentation: A 53-year-old male attended to our department with a large mass in his buttock that had been present for about one year, leading to difficulty with walking and defecation. Computed tomography (CT) showed a 10. 2 × 8. 3 cm mass with lumpy and mixed low to medium density shadows in the right side of the lower part of the tailbone. Color Doppler ultrasound showed that the skin and subcutaneous tissues of the right buttock were thickened with lymphedema. Pre-operative pathology revealed a large amount of adipose and fusiform fibrous tissues with obvious blood vessels. The patient underwent complete resection of the mass in the right buttock. Postoperative pathology indicated that the tumor was pleomorphic lipoma with malignant transformation. The patient recovered well and was discharged from the hospital at day 12 post-surgery. Follow up at 6 months did not find any symptoms of tumor recurrence.

Conclusions: Pleomorphic lipoma with malignant transformation is rare. CT, magnetic resonance imaging (MRI) and histopathology are the main methods used to diagnose pleomorphic lipoma. Complete resection of the tumor is vital to treat giant pleomorphic lipoma successfully.

Background

Pleomorphic lipoma is a benign soft tissue tumor that hardly ever occurs in the dermis. The lack of morphological demarcation means that pleomorphic lipoma is easily confused with other kinds of spindle cell lipomas [1, 2]. Pleomorphic lipoma was thought to be separated from spindle cell lipoma. However, other studies have proven that there are similarities in terms of clinical characteristics, histology, and cytogenetic findings between pleomorphic lipoma and spindle cell lipoma [2]. Thus, recently, it was proposed that pleomorphic lipoma and spindle cell lipoma are variants of the same disease [2]. Pleomorphic lipoma usually occurs in the subcutaneous tissues of the neck, shoulders, and back of males age 50–70 years old [2–4].

Pleomorphic lipoma is usually characterized by a slowly growing lesion or a rapid expansion of a previous stable mass [4]. Malignant transformation of pleomorphic lipoma is rarely seen in the clinic. In most cases, lipomas are painless and slow growing; however, they sometimes infiltrate the muscle tissues, which requires surgical treatment [5].

Here, we report a very rare case of pleomorphic lipoma with malignant transformation in the buttock. We also summarize the features, diagnosis, and surgical management of this disease.

Case Presentation

A 53-year-old male attended to our department with a large mass in the buttock that had been present for about one year, leading to difficulties with walking and defecation. In the past two months, the mass had become obviously swollen and the defecation difficulty was aggravated gradually; however, no mucopurulent bloody stool was found.

The patient received heart stent surgery one year ago. He denied any other history of hypertension, diabetes, blood transfusions, or drug and food allergies. A 15 × 10 cm mass was observed on the right side of the hips. The mass was tough, with poor mobility and no tenderness. Compression by the mass had moved the anus to the left and upper direction.

Biochemical examination indicated normal levels of carcinoembryonic antigen (CEA), carbohydrate antigen 19 − 9 (CA 19 − 9), alpha fetoprotein (AFP), total leukocytes, and hemoglobin. Abdominal computed tomography (CT) scan showed a 10. 2 × 8. 3 cm mass with lumpy and mixed low to medium density shadows in the right side of the lower part of the tailbone (Fig. 1A). The wall of the mass was thick, with soft tissue density. The boundary between the left side of the mass and the anus was not clear. Multiple enlarged lymph nodes were observed in the bilateral inguinal region. Color Doppler ultrasound showed that the skin and subcutaneous tissues of the right buttock were thickened with lymphedema. An irregular mass of 15.3 × 7.4 × 11.1 cm was observed (Fig. 1B). Under the guidance of ultrasound, the mass was punctured and biopsied for pathology. The preoperative pathology found a large amount of adipose and fusiform fibrous tissues with obvious blood vessels. The mass was considered to be a mesenchymal tumor.

The patient underwent complete resection of the mass in the right buttock. The skin was incised longitudinally (Fig. 2A). During surgery, we observed that the capsule of the tumor was intact. The upper margin of the tumor was closely related to the levator ani muscle. The tumor was separated completely along the skin (Fig. 2B and C). A drainage tube was retained in the residual cavity, which was then sutured and fixed. The resected tumor was sent for intraoperative rapid pathological examination and was diagnosed as lipoma. The operation was performed smoothly and the patient received anti-inflammatory treatment.

Postoperative pathology indicated that the tumor was pleomorphic lipoma. Some cells of the tumor were heteromorphic and overlapped morphologically with atypical lipoma, which indicated malignant transformation of the tumor (Fig. 3). Immunohistochemical staining showed positive staining for CD34, CD99, CDK4, murine double minute2 (MDM2), P16, and S-100 (Fig. 4).

Mild edema of the incision occurred after surgery (Fig. 2D). Active dressing changes were performed to eliminate the edema. The patient recovered well and was discharged on day 12 after the surgery. During a follow-up time of 6 months, we did not observe any signs of tumor recurrence.

Discussion And Conclusions

Most cases of pleomorphic lipoma are benign [1, 4, 6], and malignant transformation of pleomorphic lipoma is very rare. Pleomorphic lipoma usually occurs in the hands, feet, thighs, shoulders, arms, knees, and buttocks [7, 8]. Lipomas are termed giant when they are larger than 5 cm, and 75% of cases are symptomatic [9]. Pleomorphic lipoma accounts for 1.5% of all lipomas [10]. In our case, the diameter of the pleomorphic lipoma was more than 10 cm. A typical clinical presentation of pleomorphic lipoma is important for diagnosis. Pleomorphic lipoma usually presents as a painless soft tissue lesion. Large pleomorphic lipoma is characterized by swelling and expansion, which might cause pain when it compresses a nerve. In contrast, small pleomorphic lipomas are usually asymptomatic [11, 12]. In this case, pleomorphic lipoma occurred in the buttock and had relatively specific symptoms, such as walking inconvenience and defecation difficulty.

Pleomorphic lipoma is very difficult to diagnose, and is not easy to distinguish from highly specialized liposarcoma [10]. The diagnosis of pleomorphic lipoma is mainly based on CT, magnetic resonance imaging (MRI), and histopathology. The imaging findings of pleomorphic lipoma are variable because of the proportion of fat to non-fat components [13, 14]. On CT, pleomorphic lipoma is a relatively clear and inhomogeneous low-density mass [15]. Pleomorphic lipoma usually has clear boundaries and sometimes has local calcifications [12]. Occasionally, it reveals a mildly lobulated, irregular soft tissue mass [16]. On MRI, pleomorphic lipoma usually presents with a heterogeneous hypointense signal on T1 and a heterogeneous hyperintense signal on T2 [13, 15]. Contrast-enhanced MRI indicated heterogeneous enhancement that corresponded to a hypointense signal on T1-weighted images [13]. Heterogeneous enhancement within the tumor in CT findings might be a sign of malignant transformation of pleomorphic lipoma [17]. In our case, although there was no clear sign of infiltration on MRI, the surrounding area of the lesion seemed to be blurred, which is a suspicious radiological finding [6]. At the same time, some imaging features have been proven to increase the risk of malignant transformation of pleomorphic lipoma, such as a thick septum, a size larger than 10 cm, a nodule or mass like area, and less than 75% macroscopic fat composition [18]. In this case, we reported a large mass with lumpy and mixed low to medium density shadows on CT. The wall of the mass was thick, with soft tissue density. The boundary between the left side of the mass and the anus was not clear. The pre-operative biopsy histopathology only found a large amount of adipose and fusiform fibrous tissues within the tumor and the CT findings revealed low to medium density shadows; therefore, we did not suspect malignant transformation. However, the post-surgical histological and immunohistological examinations indicated malignant transformation of lipoma. As a result, according to this case, mixed low to medium density shadows and an unclear boundary on CT may be an indicator for the diagnosis of malignant transformation of pleomorphic lipoma. However, this needs to be further confirmed by subsequent studies.

Preoperative pathology may be effective; however, if the tissue is not large enough, it may be misdiagnosed. In this case, the preoperative pathology showed that the mass was mesenchymal tumor. However, the postoperative pathology indicated that the tumor was a pleomorphic lipoma with malignant transformation. The preoperative diagnosis was not clear. Frequently, there is no significant difference in imaging findings between malignant transformation and benign lipoma. Preoperative pathology did not

indicate malignancy, possible because the tumor volume was too large and part of the tissue necrosis reduced the accuracy of pathological diagnosis. Preoperative pathology through biopsy is sometimes imprecise because atypical cells may be concentrated and easily missed [19]. Histological and immunohistochemical examinations are the “gold standard” for the diagnosis of pleomorphic lipoma. Pleomorphic lipoma is usually characterized by multinucleated giant cells and rough collagen fibers [6]. Unique giant cells with overlapping nuclei and edge arrangement could also be found in pleomorphic liposarcoma [20, 21]. In immunohistochemistry, MDM2 and CDK4 are positive, which are helpful to distinguish benign lipoma from other lipomas [13, 20, 22]. CD34, B-cell lymphoma-2 (BCL-2), and CD99 are always strongly positive, while S-100 protein and signal transducer and activator of transcription 6 (STAT6) are usually negative [11, 14, 23, 24]. However, in a few cases, S-100 and desmin are positive in pleomorphic lipoma [13]. In this case, the immunohistochemistry showed positive staining for CD34, CD99, CDK4, MDM2, P16, and S-100.

The malignant transformation of pleomorphic lipoma is very rare in the clinic. In our opinion, if the margin of the pleomorphic lipoma is malignant, a further wider resection is needed. In addition, pleomorphic lipoma with malignant transformation might require further postsurgical treatment, such as local radiology. However, the patient in this case did not receive any treatment after surgery. The definitive prognosis of pleomorphic lipoma with malignant transformation has not been reported. As a result, close follow-up is necessary. A second surgery or other treatment methods might be needed if any tumor recurrence is found.

Complete resection of the tumor is vital to treat giant pleomorphic lipoma. Postoperative recurrence of pleomorphic lipoma is rare [10]. It is important to be familiar with the association between the lipoma and nearby organs before surgery [4]. In this case, the giant pleomorphic lipoma was located near the anus. The operation required the maximum preservation of anal function and complete resection of the lesion. The incision was large, and the postoperative recovery period was relatively long. The patient received active dressing changes and was placed on a low slag diet to prevent infection of the incision.

Abbreviations

CEA: carcino-embryonic antigen; CA 19-9: carbohydrate antigen 19-9; AFP: alpha fetoprotein; CT: computed tomography; MRI: magnetic resonance imaging; MDM2: murine double minute2; BCL-2: B-cell lymphoma-2; STAT6: signal transducer and activator of transcription 6.

Declarations

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Not applicable.

Authors' contributions

FL wrote the first version of the article. FL, XW, GMX, HYH, WL, GLL, and JNL participated in the conception and design of the study and drafting the article. GLL and JNL supervised the study and reviewed the article critically for intellectual content. All authors reviewed and approved the final version of the article.

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Availability of data and materials

All data generated or analyzed are included in this published article.

Ethics approval and consent to participate

This study was approved by the Ethics Committee and Institutional Review Board of the Second Hospital of Jilin University, Changchun, China. Written informed consent was obtained from the patient.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Competing interests

All authors declare that they have no competing interests.

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Figures

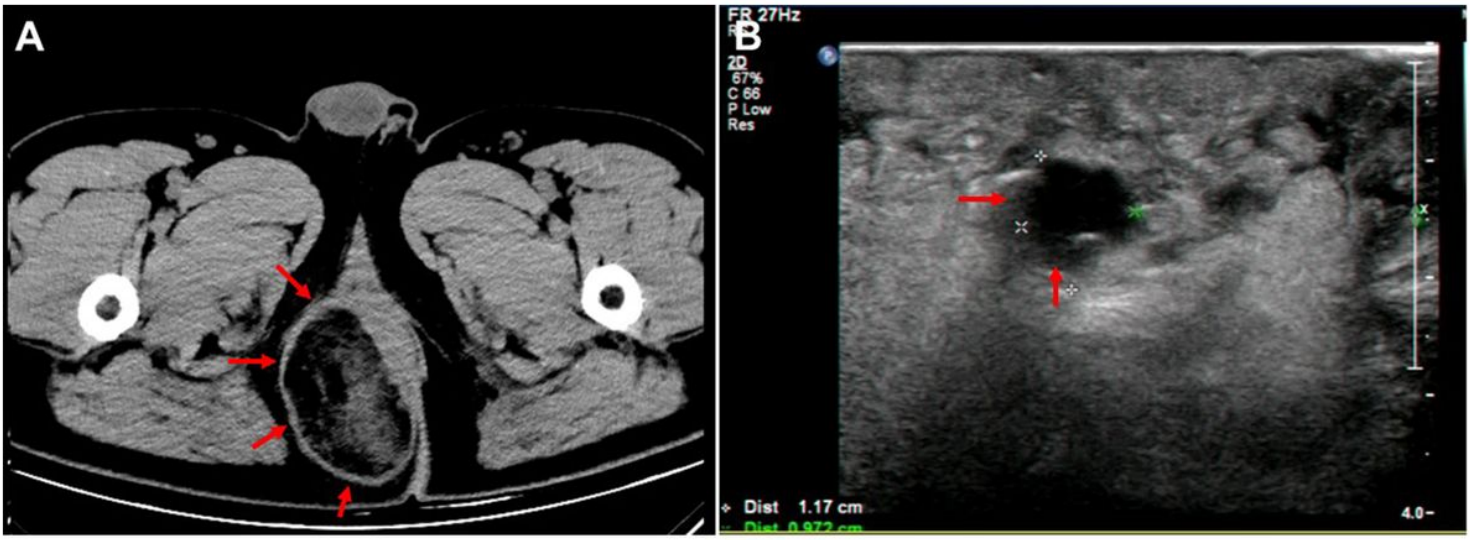

Figure 1

CT and Color Doppler ultrasound examination. (A) CT showed a 10. 2× 8. 3 cm mass (red arrows) with lumpy and mixed low to medium density shadows in the right side of the lower part of the tailbone. (B) The ultrasound examination showed an irregular mass (red arrows), which was 15.3 × 7.4 × 11.1 cm in size.

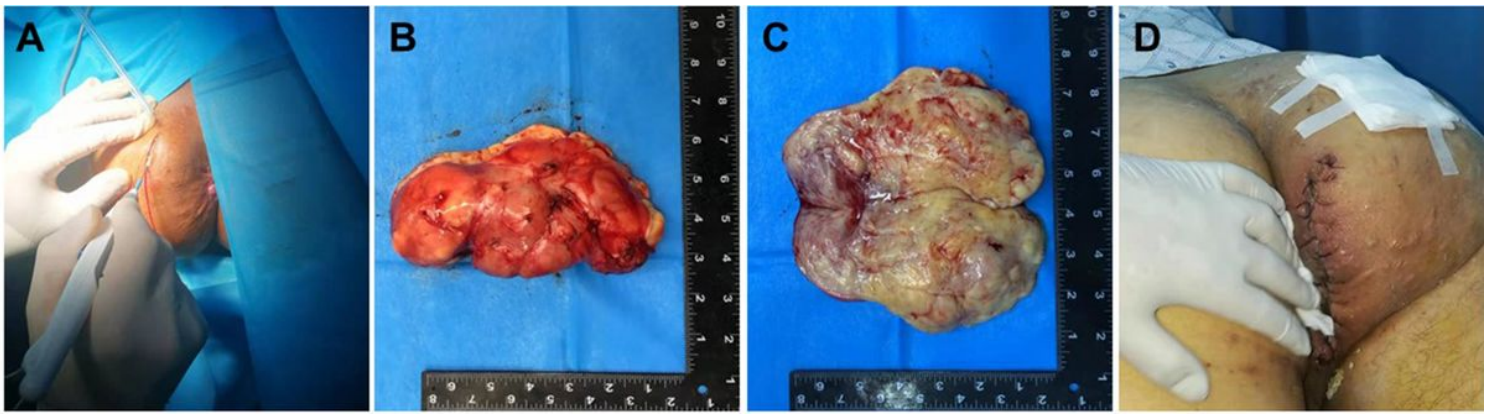

Figure 2

The macroscopic images of the skin incision, the resected tumor, and post-surgical incision. (A) The skin was incised longitudinally along the tumor. (B) The tumor was resected completely. (C) The macroscopic image of the cross section of the tumor. (D) Mild edema of the incision at day 5 post-surgery.

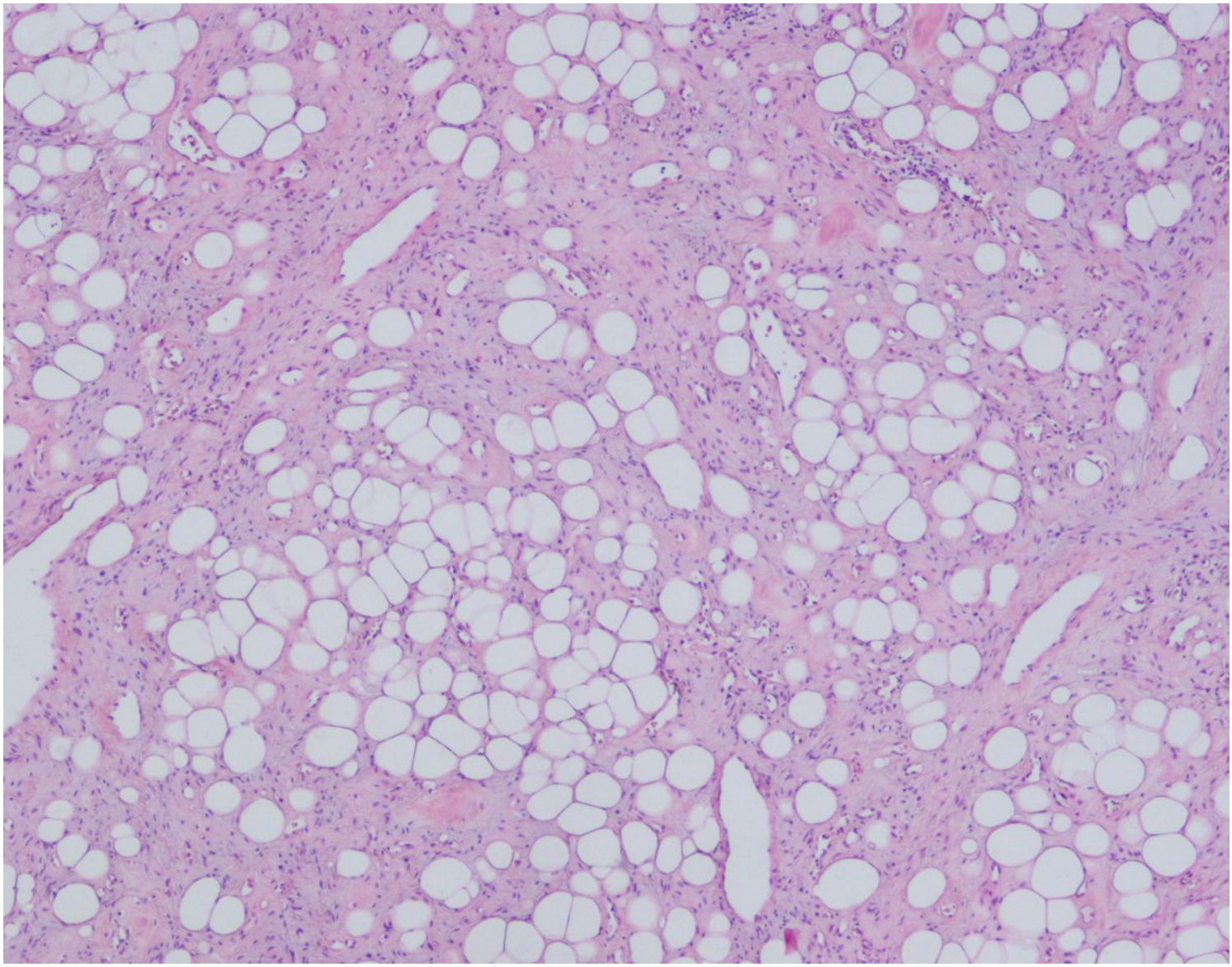

Figure 3

Postoperative pathology indicated that the tumor was pleomorphic lipoma with malignant transition.

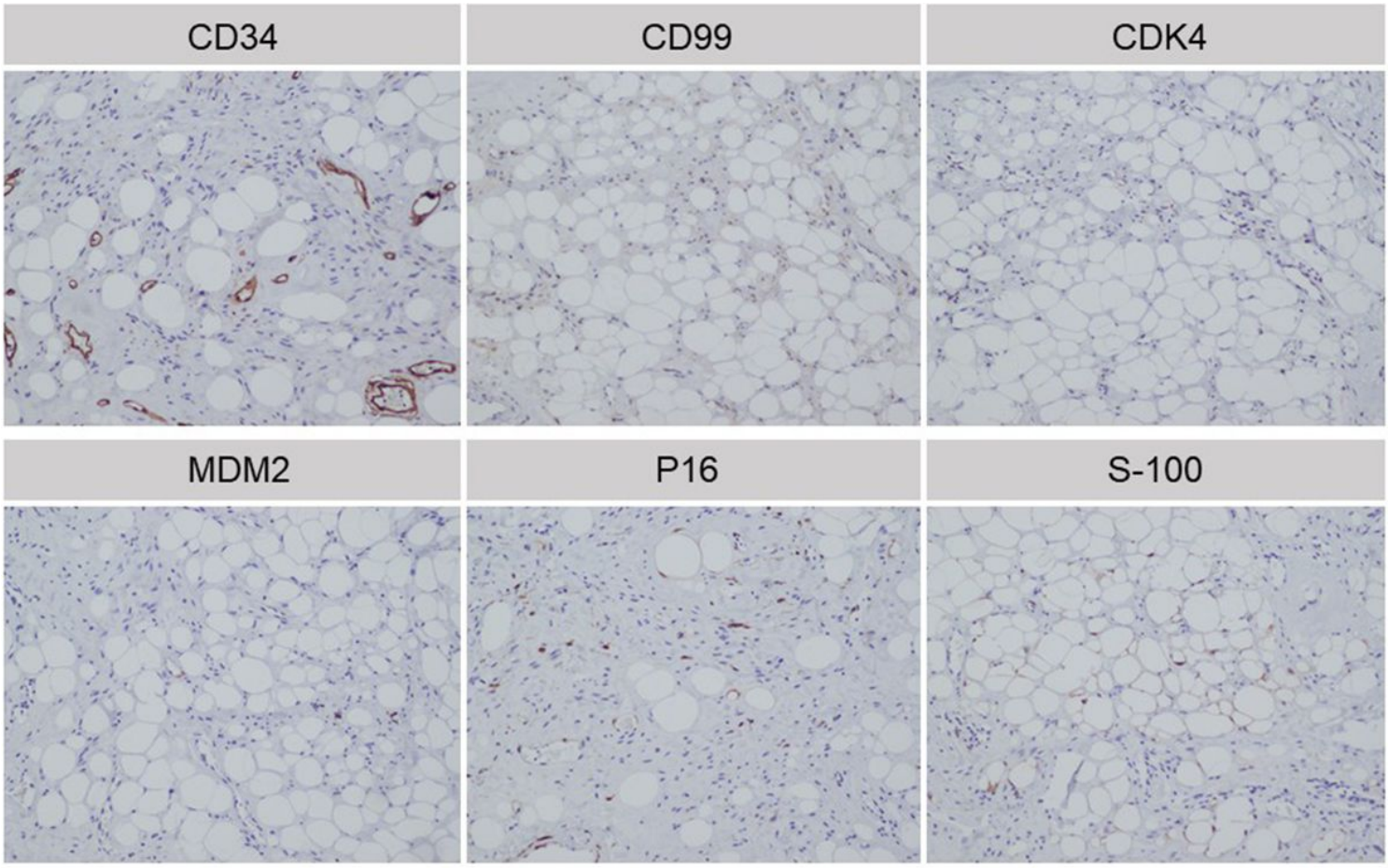

Figure 4

Immunohistochemical pathology of the tumor.