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CASE REPORT

Groin synovial sarcoma with intraluminal femoral sheath involvement☆

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Summary Synovial sarcomas are highly aggressive soft tissue malignant tumors that occur primarily in young adults. They arise from mesenchymal tissue and typically in or near tendons and tendon sheaths. They can develop at any site but the majority (80–95%) of synovial sarcomas are located in the extremities and mostly at the lower extremities. Groin synovial sarcoma is rare. About 10% of lower limb soft tissue sarcomas have major vessel involvement. In the past, amputation was the standard surgical procedure for lower limb soft tissue sarcoma with major vessels involvement. However, using modern surgical techniques, we can successfully perform a limb-preserving operation with a tumor-free margin. A 17-year-old girl presented with left inguinal synovial sarcoma with femoral artery and vein encasement. The patient received radical tumor resection with major vascular structures. Segmental femoral vessel defects were reconstructed with polytetrafluoroethylene grafts. After revascularization, we designed a pedicled anterolateral thigh fasciocutaneous flap for coverage of the wound and reconstructed vessels simultaneously. The circulation of the lower extremity was good and the girl recovered well after the surgery without morbidity. Synovial sarcoma would potentially be associated with local recurrence and distant metastasis. The presences of bone and neurovascular bundle invasion are adverse predictors of recurrence and mortality. Surgery remains the mainstay of treatment. In the advance of current surgical technique, limb-preserving surgery is possible even for tumor-involved major vessels.

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

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1. Introduction

Soft tissue sarcomas account for approximately 1% of malignant tumors. Most of them arise from the extremities, followed by the trunk wall, retroperitoneum, and head and neck.[1] Synovial sarcoma is an aggressive subtype[2] that occurs primarily in young adults.[3,4] The term "synovial" is derived from its morphologic origin and its histological resemblance, and is easily misinterpreted to mean that the tumor comes from synovium.[5] However, it is a mesenchymal tumor.[2]

The majority of the tumors appear in the extremities, with two-thirds being located in the lower extremities.[2,5] The most common site is within 10 cm of the knee. Extra-articular is dominant and true intra-articular tumor is very rare.[2] It presents as a slow-growing, frequently painful tumor.[2,3] The current standard treatment is wide resection of the tumor, followed by irradiation with or without chemotherapy.[5] Initial adequate radical ablation surgery is still the most important tumor-free prognosis factor.

When soft tissue sarcomas in the extremities involve major vessels, it may become a challenge to achieve a clear margin. In the past, amputation was the mainstay to obtain a cancer-free margin. Here, we report a case of groin synovial sarcoma that encased the femoral vasculature. We cooperated with vascular surgeons for a tumor wide excision and vascular segmental resection followed by vascular reconstruction and pedicled anterolateral thigh flap reconstruction for vessel and soft tissue defect.

2. Case report

A 17-year-old girl was in a healthy condition previously. She presented to our outpatient clinic due to a small palpable mass in the left groin area noticed for 2 months, which enlarged gradually. She denied recent trauma to the affected extremity. Physical examination revealed an elastic, firm, and fixed tumor, which was 10 cm in diameter. Its palpation provoked minimal discomfort without numbness or tenderness. Her lower extremity range of motion and muscular strength were normal. Magnetic resonance imaging also revealed a heterogenous tumor both on T1WI and T2WI images at the groin area with contrast enhancement. It also revealed septation and central necrosis. Femoral vessels were wrapped by the tumor (Fig. 1). An incisional biopsy was then performed and the final histologic diagnosis was synovial sarcoma. Further oncological surveillance to evaluate distant metastasis, such as bone scan, chest computed tomography, and chest X-ray all ruled out metastasis. A tumor wide excision was then planned and possible vascular resection will be needed. Due to the indispensability of the femoral artery for the lower extremity, further vascular reconstruction was necessary.

An oncological resection with a cutaneous peripheral safe margin of 2 cm was designed (Fig. 2). Deep plane as far as the rectus femoris muscle fascia was included, sent for frozen section intraoperatively, and revealed to be free of tumor cells. The tumor encased the femoral artery and veins at the arterial bifurcation site, and the femoral nerve was spared (Fig. 3). The involved and estimated resection vascular length measured 11 cm. The vascular surgeon decided to use a synthetic graft with polytetrafluoroethylene (PTFE) for vascular reconstruction. A 6 mm PTFE graft was first used to make a Y-shaped graft in the back table for arterial reconstruction. We did tumor *en bloc* resection with femoral artery and vein segmental resection. The lower extremity became cold and poorly circulated, so the vascular surgeon started revascularization. The Y-shaped PTFE graft is anastomosis from the femoral artery to the deep femoral artery and superficial artery. Another 8 mm graft was used for vein reconstruction from the proximal femoral vein to distal femoral vein (Fig. 4). No intraoperative heparin was administered. After revascularization, the lower extremity became warm and well circulated. An ipsilateral pedicled anterolateral thigh flap was then designed for resultant inguinal soft tissue defect reconstruction. The pedicled anterolateral thigh flap with skin paddle sized 15 cm $\times$ 9 cm with two musculocutaneous perforators, based on the lateral circumflex branch of femoral artery, was harvested (Fig. 4A–C). Partial tensor fascial lata was also included. The flap was rotated through the subcutaneous tunnel to reach the inguinal defect (Fig. 4D). A fascial lata was used for inguinal deep fascia reconstruction and repaired with 1-0 Surgilon (Covidien). The flap donor site was covered with a split-thickness skin graft harvested from the medial thigh.

Neither anticoagulant agent nor antiplatelet therapy were given postoperatively. The circulation of the flap and lower extremity were good. The patient was discharged uneventfully.

2.1. Outcome

The biopsy specimen grossly revealed a tumor cell intramural invasion (Fig. 5). The final pathology report confirmed synovial sarcoma, biphasic type, with free margins and local regional lymph nodes were negative of malignancy. Pathological pictures revealed spindle-shaped neoplastic cells and focal epithelial components with glandular formation. Vascular invasion and emboli were also noted. Immunohistochemically, both the spindle and epithelial components were positive for epithelial membrane antigen (Fig. 6). Adjuvant radiotherapy and chemotherapy were suggested, but the patient and her parents refused radiotherapy. Therefore, chemotherapy with epirubicin and cisplatin were started 1 month after the operation. Six courses of chemotherapy were applied until abandoned at the patient's wish. During follow-up for 14 months, neither lower limb swelling, synthetic graft thrombosis, nor wound infection were detected (Fig. 7).

3. Discussion

Synovial sarcomas are malignant soft tissue sarcomas comprising approximately 6–9% of all adult soft tissue sarcomas. There is a wide age range at presentation, but young adults are affected most often.[6] There is no sex predilection. It can develop in any anatomical site; however, the extremities are the most common sites and the majority occur in the lower extremities. It mostly develops