Successful outcome after intralesional curettage for spindle cell hemangioma of fibula: a case report

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Case report

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Abstract

Background

Spindle cell hemangioma (SCH), a non-neoplastic reactive vascular lesion, rarely locates in bones. We herein report a successful case of intralesional curettage for an infant with SCH of fibula.

Case presentation

A 11-month-old male patient was admitted to our center with a painless mass in right proximal calf. Preoperative digital radiograph demonstrated a massive vascular lesion with an irregular bone destruction of proximal fibula. The lesion was removed via the intralesional curettage approach and pathologically diagnosed as SCH. After the surgery, the patient gained bone structure recovery of right proximal fibula. Two years after the surgery, he experienced no local recurrence.

Conclusion

For the management of SCH of fibula with partial bone destruction, we suggest early-stage intralesional curettage as its safety and effectiveness.

Background

Spindle cell hemangioma (SCH), characterized by cavernous blood vessels and spindle cell proliferation, has recently been considered as a non-neoplastic reactive vascular lesion[1, 2]. SCH often occurs at early age with high risk of recurrence after surgery, due to its uncertain border with surrounding tissue. It commonly arises in the dermal or subcutaneous tissue of the distal extremities[3, 4]. Reports on SCH cases involving bone are rare, most of which focus on histopathological description, but lack sufficient clinical and follow-up data[5, 6]. Herein, we present a case of SCH in proximal fibula that was managed successfully by intralesional curettage, and moreover, discuss its clinical characteristics and long-term surgical outcome.

Case Presentation

A 11-month-old male patient presented to our center with a 2-month history of painless mass in right proximal calf. The mass had been noted to be slowly enlarging in 3 weeks after presentation. No significant symptom was found in this patient. Initial workup performed included radiograph, 3-dimensional computed tomography (3d CT) reconstruction, and magnetic resonance imaging (MRI). Radiographs of the right tibia and fibula indicated an irregular bone destruction of proximal fibula (Fig. 1a, b), and the lytic bone destruction was confirmed by 3d CT reconstruction (Fig. 1c, d). MRI revealed a massive vascular tumor with surrounding soft tissue hyperplasia, and involvement of the proximal fibular epiphyseal plate (Fig. 1e, f).

Considering partial bony structure of proximal fibula was normal, intralesional curettage was performed on the right proximal fibula under general anesthesia. After lesion exposure, a 4.0×2 .0 cm-sized vascular mass was identified with extension to proximal fibular. Gross examination showed a reddish spongious solid mass, containing topical hemorrhage, partial thrombosis, and irregular bone destruction (Fig. 2a). With protection of common peroneal nerve and peripheral vessels, complete curettage of lesion was performed to normal fibular surface (Fig. 2b, c). Histologically, the lesion was characterized by the fissure-like vessel lumens lined with flattened endothelial cells among the spindle cells (Fig. 3a). The spindle shaped cells arranged in fascicular pattern in solid area, with similar cell morphology and no atypia (Fig. 3b, c). Immunohistochemically, the endothelial cells lining the vessel spaces stained positive for CD31, CD34, and ERG. Therefore, with standard of international society for study of vascular anomalies (ISSVA) classification[7], the diagnosis of spindle cell hemangioma was made in this patient according to the clinical and histopathologic manifestations. On postoperative follow-up, this patient was asymptomatic without any evidence of recurrence (Fig. 4). Two years after this surgery, he returned to hospital for outpatient review. Radiographs showed the reformation of the cortex of the proximal fibula (Fig. 5a, b), and both uniform bone mineral density and continuous cortical of right proximal fibula were confirmed by 3d CT reconstruction (Fig. 5c, d). Besides, MRI demonstrated remarkable regression of lesion without any signs of tumor growth through the fibula (Fig. 5e, f).

Discussion

This case is rare in comparison with majority of reported SCH cases and merits discussion on following points: location of lesion, selection of surgical intervention, histopathologic characteristics, and long-term postoperative follow-up. SCH is a benign vascular lesion which generally locates in the subcutis at the distal extremities and presents as solitary and multifocal masses. It also can be associated with several clinical syndromes, among which Maffucci syndrome is the most common[8, 9]. In several uncommon cases, SCHs have been found in lips, nasal passage, temporal muscle, and even in lungs and spleen[2, 10–13]. In comparison, the reported cases of SCH arising in bones are even more unusual so far[14–16]. In our case, a solitary lesion of SCH involved the proximal fibula with surrounding soft tissue hyperplasia, while the superficial skin and tissues were normal.

To date, the main treatment choice for fibular tumor is segmental or subperiosteal resection, in case of local recurrence at surgical site[17–19]. Given that preoperative digital radiograph indicated that the vascular mass on fibula was solitary, and part of both cortex and cancellous fibula were not involved, intralesional curettage was selected as the surgical intervention in this case for achieving the maximum retention of healthy bony structure. During the operation, complete curettage was performed to the normal fibular surface without residual lesion.

The histologic appearance in this case consisted of the fissure-like vessel lumens lined with flattened endothelial cells among the spindle cells, which arranged in fascicular pattern in solid area. Subsequent immunohistochemical analysis revealed positive staining for CD31, CD34 and ERG in the majority of spindle cells, consistent with the diagnosis of SCH[20, 21]. Metastasis of SCH is rare, although local

recurrence may occur[22, 23]. On the most recent imaging examination, 2 years after the initial surgery, our patient was still disease-free and found to experience entire reformation of bone structure of right proximal fibula. This indicates the safety and effectiveness of intralesional curettage for the management of this case.

Conclusions

For SCH of fibula with partial bone destruction, intralesional curettage renders a safe and efficient intervention at early stage.

Abbreviations

SCH, Spindle cell hemangioma; 3d CT, 3-dimensional computed tomography; MRI, Magnetic resonance imaging.

Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

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

Availability of data and materials

All data generated during this report are included in this published article.

Competing interests

The authors declare that they have no competing interests.

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None.

Authors' contributions

RW performed the surgery and conducted the data analyses. TH wrote sections of the article and edited the figures. XZ participated in reviewing literature and designed the study. All authors read and approved the final manuscript.

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Figures

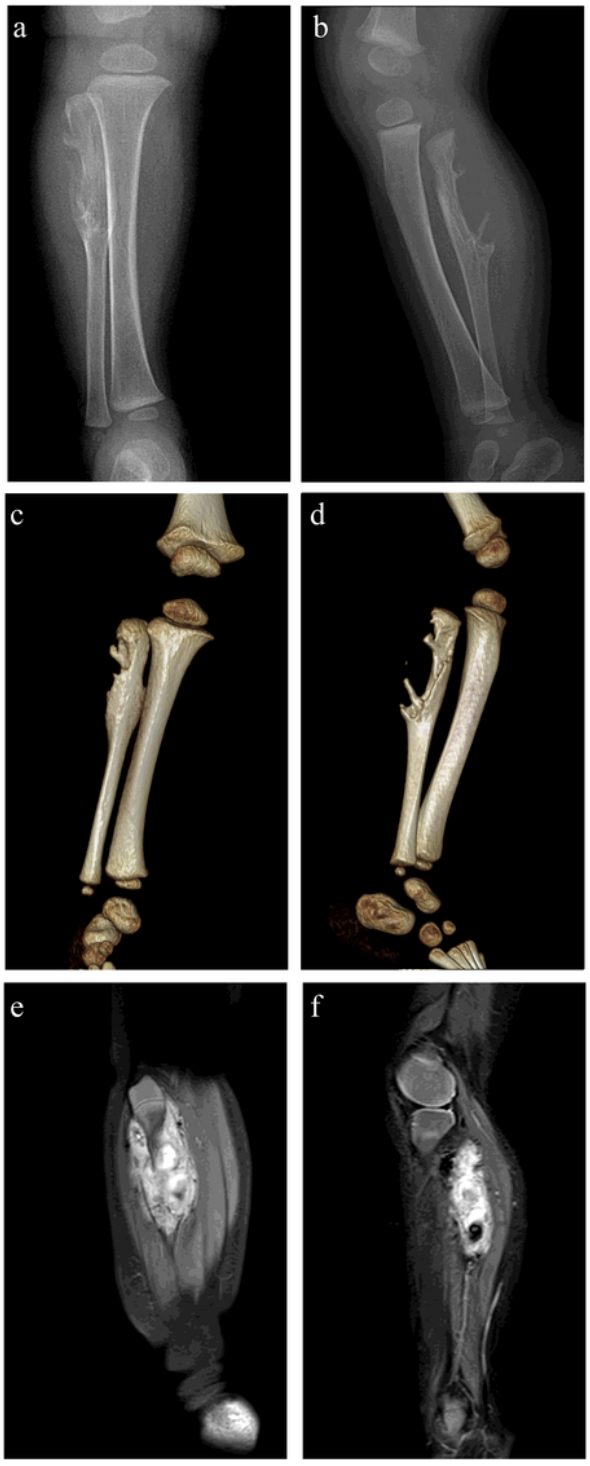

Figure 1

Digital radiograph preoperatively: a, b radiographs of the right tibia and fibula showing an irregular bone destruction of proximal fibula; c, d 3d CT reconstruction demonstrating lytic bone destruction of right proximal fibula; e, f MRI revealing a massive vascular tumor with surrounding soft tissue hyperplasia, and involvement of the proximal fibular epiphyseal plate.

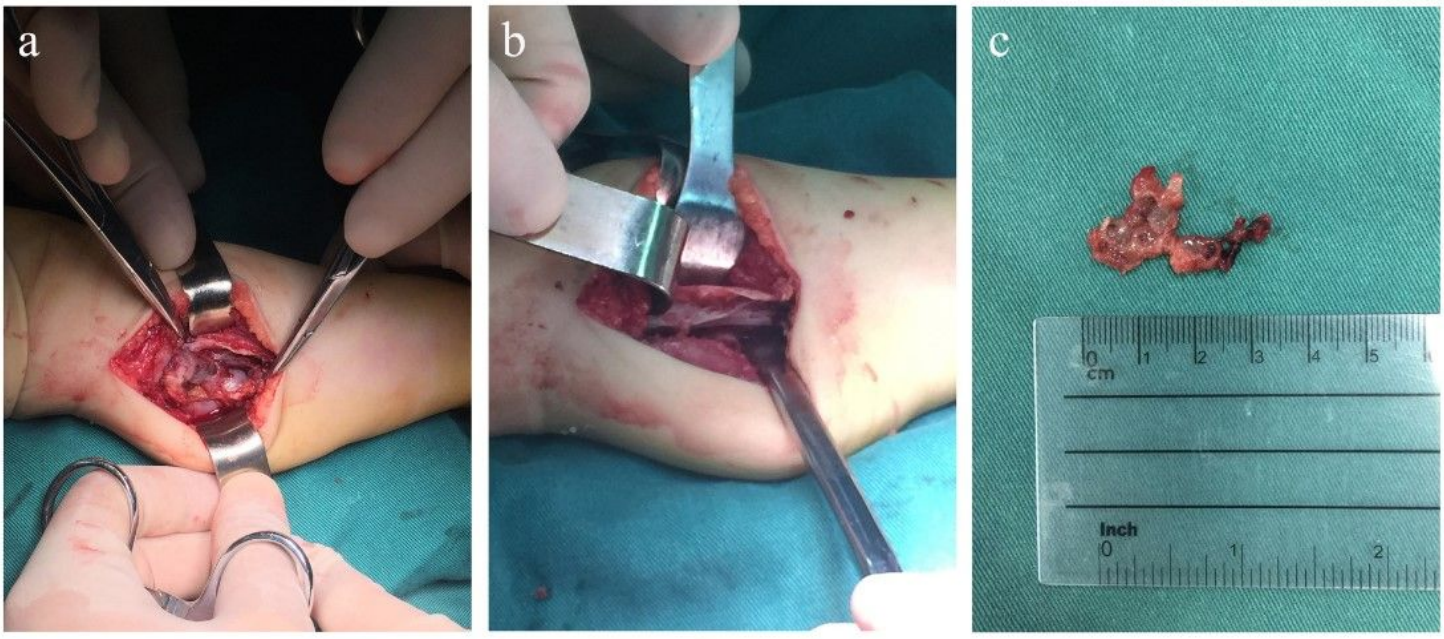

Figure 2

The photograph during the surgery: a intraoperative image of the surgical finding of a vascular mass attached to proximal fibula; b complete curettage of lesion to normal fibular surface; c macroscopic appearance of the excised lesion.

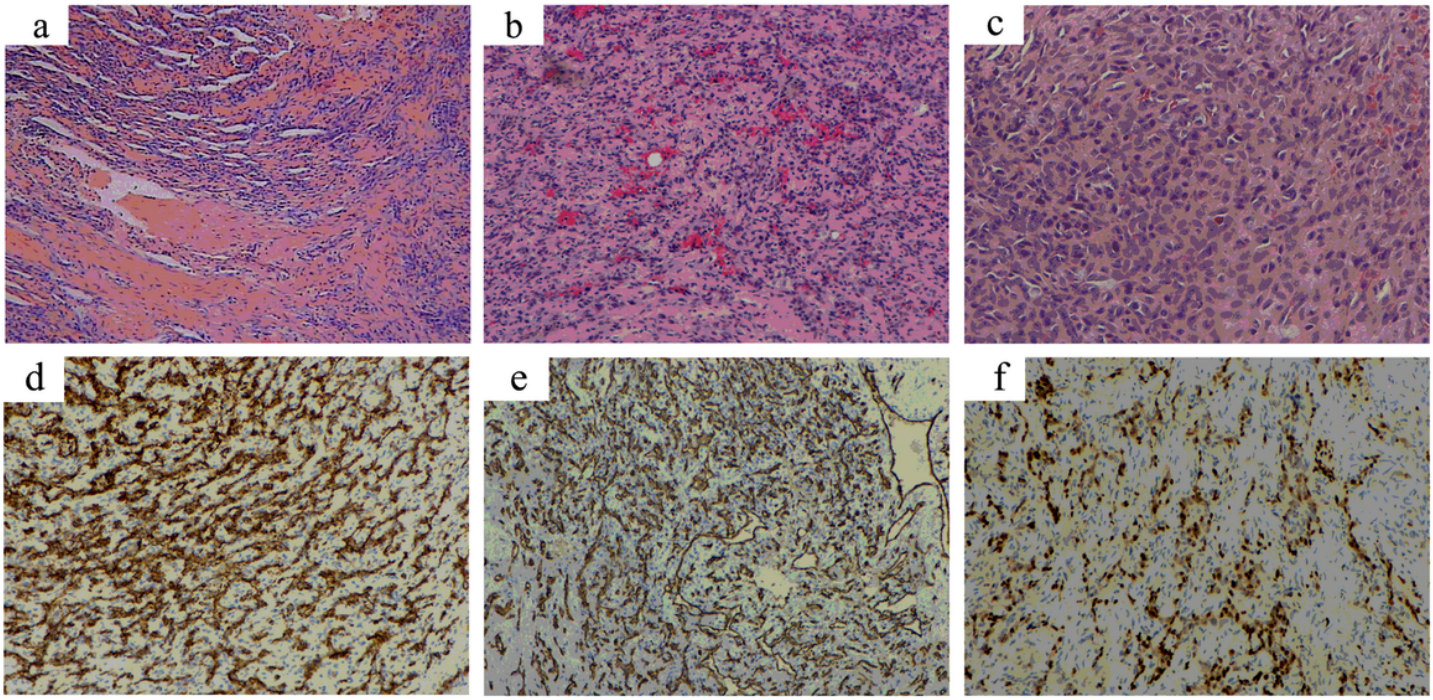

Figure 3

Histopathological features: a (HE, ×40) the fissure-like vessel lumens lined with flattened endothelial cells among the spindle cells, b (HE, ×100), c (HE, ×200) the spindle shaped cells arranging in fascicular

pattern in solid area. Immunohistochemical analysis revealing positive staining for d CD31 (×100), e CD34 (×100) and f ERG (×100) in the majority of spindle cells.

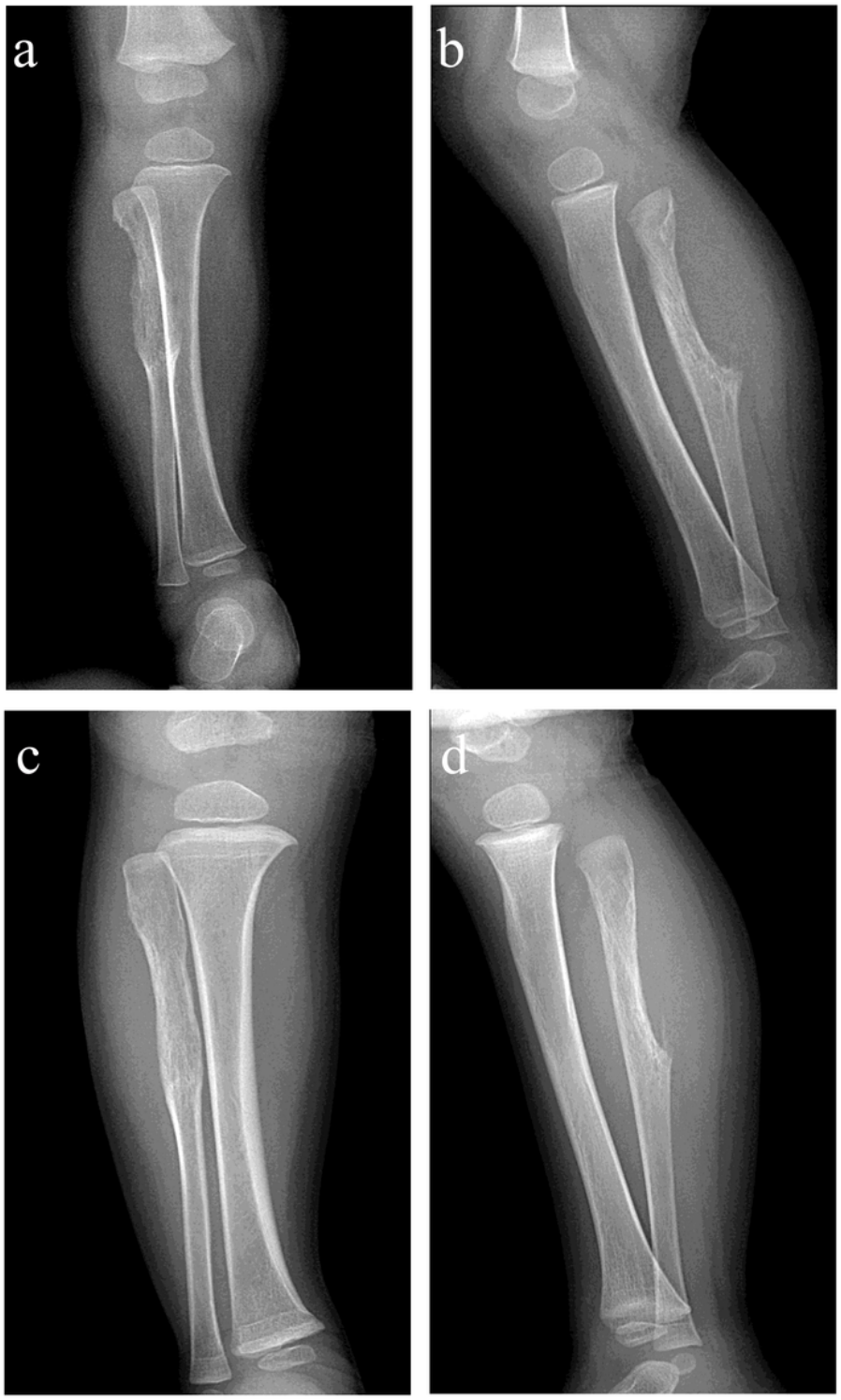

Figure 4

a Digital radiograph at 1 month follow-up, b 6 months postoperatively.

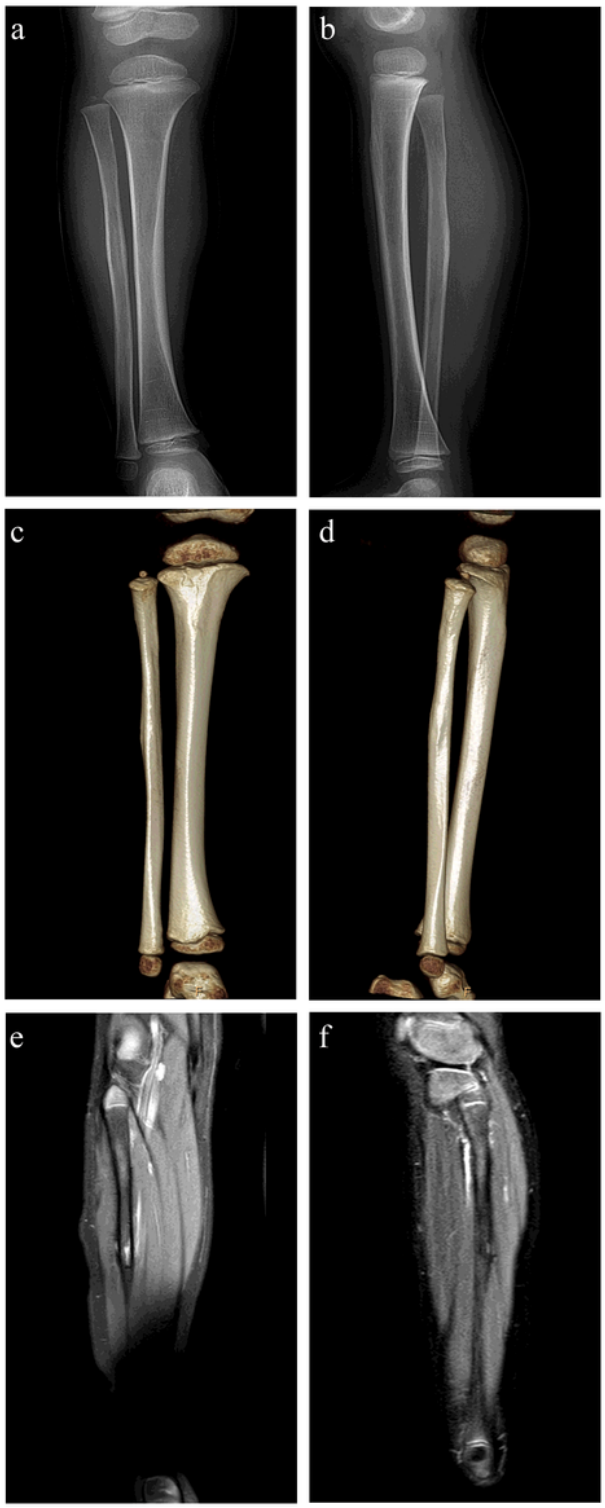

Figure 5

Digital radiograph at 2 years postoperatively: a, b radiographs showing reformation of the cortex of the proximal fibula; c, d 3d CT reconstruction demonstrating both uniform bone mineral density and continuous cortical of right proximal fibula; e, f MRI revealing remarkable regression of lesion without evidence of local recurrence.