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

Rufa Wang  
Nanjing Children's Hospital: Children's Hospital of Nanjing Medical University

Tao Han  
Nanjing Children's Hospital: Children's Hospital of Nanjing Medical University

Xiaoguang Zhou (zxgnch@sina.com)  
https://orcid.org/0000-0002-1783-2845

Case report

Keywords: Spindle cell hemangiomatia, Intralesional curettage, Outcome, Vascular lesions

DOI: https://doi.org/10.21203/rs.3.rs-379859/v1

License: ©️ This work is licensed under a Creative Commons Attribution 4.0 International License.  
Read Full License
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 bula 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.

Funding

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.

Acknowledgements
The authors thank the patient’s parents for allowing us to publish his case.

Authors’ information

1 Department of Orthopedic Surgery, Children’s Hospital of Nanjing Medical University, No. 72 Guangzhou Road, Nanjing, Jiangsu 210008, China.

2 Department of Burns and Plastic Surgery, Children’s Hospital of Nanjing Medical University

3 Neonatal Medical Center, Children’s Hospital of Nanjing Medical University

References

1. Tosios KI, Gouveris I, Sklavounou A, Koutlas IG. Spindle cell hemangioma (hemangioendothelioma) of the head and neck: case report of an unusual (or underdiagnosed) tumor. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2008; 105:216–21.

2. Gao BQ, Zhou DK, Qian XH, Zhang W, Ying LX, Wang WL. Spindle cell hemangioma of the spleen: A case report. Medicine. 2019;98:e14555.

3. Liu H, Rao Y, Gu H, Yang X, Hu L, Sun Y, Chen H, Lin X. Characteristic appearance of spindle cell hemangiomatosis, often misdiagnosed as venous malformation: A retrospective study of 11 cases. J Dermatol. 2020;47:1424–31.

4. Marusic Z, Billings SD. Histopathology of Spindle Cell Vascular Tumors. Surg Pathol Clin. 2017;10:345–66.

5. Maclean FM, Schatz J, McCarthy SW, Scolyer RA, Stalley P, Bonar SF. Epithelioid and spindle cell haemangioma of bone. Skeletal Radiol. 2007;36(Suppl 1):50-7.

6. Vijayan S, Naik M, Rao S, Hameed S. Spindle cell hemangioma of femur. Clin Cancer Invest J. 2015; 4:469 – 72.

7. Miller DD, Gupta A. Histopathology of vascular anomalies: update based on the revised 2014 ISSVA classification. Semin Cutan Med Surg. 2016; 35:137 – 46.

8. Lekwuttikarn R, Chang J, Teng JMC. Successful treatment of spindle cell hemangiomas in a patient with Maffucci syndrome and review of literatures. Dermatol Ther. 2019;32:e12919.

9. Gupta V, Mridha AR, Khaitan BK. Unsatisfactory response to sirolimus in Maffucci syndrome-associated spindle cell hemangiomas. Dermatol Ther. 2019;32:e12851.

10. Murakami K, Yamamoto K, Sugiura T, Kirita T. Spindle Cell Hemangioma in the Mucosa of the Upper Lip: A Case Report and Review of the Literature. Case Rep Dent. 2018; 2018:1370701.

11. Tastemel Ozturk T, Suslu AE, Kavuncuoglu A, Gumeler E, Kosemehmetoglu K, Yalcin B. Spindle cell hemangioma of nasal passage and ethmoidal sinus in a 4-month old infant. Arch Argent Pediatr. 2021;119:e36–40.
12. Minagawa T, Yamao T, Shioya R. Spindle cell hemangioendothelioma of the temporal muscle resected with zygomatic osteotomy: a case report of an unusual intramuscular lesion mimicking sarcoma. Case Rep Surg. 2011; 2011:481654.

13. Duqing X, Zhaohong W, Gefei W. Multiple spindle cell hemangiomas in both lungs: a rare case report and review of the literature. J Cardiothorac Surg. 2019;14::86.

14. Vaseenon T, Saengsin J, Pattamapaspong N, Settakom J, Pruksakorn D. Spindle Cell Hemangioma of the Midfoot: A Case Report. J Orthop Case Rep. 2017;7:75–9.

15. Hakozaki M, Tajino T, Watanabe K, Yamada H, Kikuchi S, Hojo H, Ishida T, Konno S. Intraosseous spindle cell hemangioma of the calcaneus: a case report and review of the literature. Ann Diagn Pathol. 2012;16:369–73.

16. Tsukamoto S, Honoki K, Shimada K, Fujii H, Kido A, Takano M, Enomoto Y, Kasai T, Konishi N, Tanaka Y. Periosteal spindle cell hemangioma of the fibula: a case report. Skeletal Radiol. 2013;42:1165–8.

17. Guo C, Zhang X, Gao F, Wang L, Sun T. Surgical management of proximal fibular tumors: risk factors for recurrence and complications. J Int Med Res. 2018;46:1884–92.

18. Mostafa MF. Subperiosteal resection of fibular aneurysmal bone cyst. Eur J Orthop Surg Traumatol. 2015; 25:443–50.

19. Erler K, Demiralp B, Ozdemir MT, Basbozkurt M. Treatment of proximal fibular tumors with en bloc resection. Knee. 2004; 11:489 – 96.

20. Sheehan M, Roumpf SO, Summerlin DJ, Billings SD. Spindle cell hemangioma: report of a case presenting in the oral cavity. J Cutan Pathol. 2007;34:797–800.

21. Chavva S, Priya MH, Garlapati K, Reddy GS, Gannepalli A. Rare Case of Spindle Cell Haemangioma. J Clin Diagn Res. 2015;9::ZD19–21.

22. Perkins P, Weiss SW. Spindle cell hemangioendothelioma. An analysis of 78 cases with reassessment of its pathogenesis and biologic behavior. Am J Surg Pathol. 1996; 20:1196 – 204.

23. Patel SV, Bass FD, Niemi WJ, Pressman MM. Spindle cell hemangioendothelioma: a case presentation and literature review of a rare lower extremity tumor. J Foot Ankle Surg. 1996;35:309–11.

**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.