International Journal of Surgery Case Reports 99 (2022) 107643

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr

Case report

# Intraosseous schwannoma of distal femur: A case report

Muhammad Wahyudi [a,*], Ramadan Premiarto Clevfirstarachma [b], Mardiani Djailani [c]

[a] Department of Orthopaedic and Traumatology, Fatmawati General Hospital, Jakarta, Indonesia
[b] Fatmawati General Hospital, Jakarta, Indonesia
[c] Department of Anatomical Pathology, Fatmawati General Hospital, Jakarta, Indonesia

ARTICLE INFO

*Keywords:*
Case report
Femur
Intraosseous
Primary
Schwannoma
Tumor

ABSTRACT

*Introduction and importance:* Intraosseous schwannoma is a rare benign tumor, which mostly occurred in head and neck region. In this report we aimed to describe a unique case of intraosseous schwannoma in the distal femur without any other clinical finding aside from pain and tenderness.

*Case presentation:* 19-year-old female presented with persistent pain on her left thigh for 4 years. Aside from tenderness on her left thigh, her physical examination was unremarkable. Plain radiographic of left femur showed a small geographic osteolytic cortical lesion with sclerotic rim in the distal region. Further evaluation with MRI showed eccentric lytic lesion with an isointense signal on T1-weighted images and a hyperintense signals on T2-weighted images. Patient then temporarily diagnosed with osteoblastoma. Because there were no signs of malignancy, the patient underwent a curettage of the mass followed by synthetic bone graft application. Histopathological findings were consistent for schwannoma. Further immunohistochemical examination showed positive S100 staining, confirming the final diagnosis of intraosseous schwannoma. There were no signs of early complication on 3 months post-operation. The patient was further scheduled for follow up on 6 months and then routinely every year post-operation to evaluate any signs of complication or recurrence.

*Clinical discussion:* It is difficult to make an accurate initial diagnosis of intraosseous schwannomas. Because the clinical presentation was most likely not specific as such in this case and there many other tumors of the bone with similar radiographic finding which are more common. Curettage of the mass followed by synthetic bone graft application was performed as there were no sign of malignancy making more invasive option deemed to cause more harm than good to the patient.

*Conclusion:* The possibility of intraosseous schwannoma should have been taken under consideration in the differential diagnosis of osteolytic lesion with pain in long bone.

## 1. Introduction

Schwannomas (neurilemmoma) is a benign tumor originating from the Schwann cells. Comprising approximately 5 % of all benign soft tissue tumors, schwannoma is an uncommon condition. As Schwann cell function is to form the myelin sheath on peripheral nerve, schwannomas can virtually develop in almost any body region. But the most common predilection sites of schwannomas are around head and neck region as those area contains rich innervation from either cranial or spinal nerve [1,2].

Because nerve fibers pass alongside with blood vessels inside of canals of the bone, Schwannomas can also develop from inside the bone (intraosseous). But intraosseous schwannoma is a very rare case which accounts for only 0.2 % of all primary bone tumors. Mandible is the most commonly affected bone. Though schwannoma can also develop from the long bones such as tibia, femur, humerus, radius and ulna such cases were rarer than in the mandible [2–4].

It is difficult to make an accurate initial diagnosis of intraosseous schwannomas. First, because the clinical presentation was most likely not specific as most patients with intraosseous schwannomas are generally asymptomatic while others complained pain with or without edema. Second, because it is highly uncommon condition, it is most likely misdiagnosed as other tumors of the bone with similar radiographic finding which are more common [2–4]. Hence in this publication, we are reporting a case of intraosseous schwannoma in distal femur.

This case report has been reported in line with the SCARE Criteria [5].

* Corresponding author.
*E-mail address:* rama.premiarto@gmail.com (M. Wahyudi).

https://doi.org/10.1016/j.ijscr.2022.107643
Received 3 July 2022; Received in revised form 15 August 2022; Accepted 9 September 2022
Available online 13 September 2022

## 2. Case report

A 19-year-old female was referred by primary care center to Orthopedic Unit at Fatmawati General Hospital for persistent pain on her left thigh for about 4 years. The pain was relieved by analgesics such as meloxicam, but recurred few moments after. The patient felt that her pain worsened for about three months, which had led to difficulties of walking.

The patient stated that she previously had sought for medical attention to another hospital and was diagnosed with bone tumor, but she didn't continue the treatment protocol. Aside from the tumor, she denied any previous history of medical condition, trauma, and surgery. Her medication history were only analgesics such as meloxicam, which she didn't use routinely. The patient was a student and she denies any routine activity that may lead to chronic microtrauma. Family history of tumor was denied by the patient.

On physical examination patients vital sign were normal and her BMI was 17,04 kg/m$^2$. There was tenderness on palpation at distal region of femur, but there was no sign of mass, deformity, atrophy, and swelling. Left knee range of motion was also normal. There were no sign of lymph node enlargement or any other abnormalities to be found on physical examination (Fig. 1).

Plain radiograph of the left femur for initial assessment showed a small geographic osteolytic lesion with sclerotic rim inside the anterior cortex of the distal femur. Further evaluation with MRI revealed an eccentric lytic lesion with clearly demarcated but irregular border. On T1-weighted images the lesion was isointense with hypointense margin and hyperintense on T2-weighted images. These findings still suggest several possibilities such as a non-ossifying fibroma and simple bone cyst (Figs. 2 and 3).

Based on clinical and radiological examination, the initial working diagnosis of osteoblastoma was established. As most of possible differential diagnoses (such as non-ossifying fibroma and simple bone cyst) were benign condition the patient surgical procedure was planned with curettage the lesion and synthetic bone graft application. The procedure was done by orthopaedic oncology consultant in Fatmawati general hospital which was a teaching hospital. 2 g of ceftriaxone was used preoperatively as surgical prophylactic antibiotic. Spinal anesthesia was used for procedure. The surgery went from incision directly from anterior approach because of the tumor location, followed by curettage of the lesion, collecting samples for histopathological analysis and synthetic bone graft application. The procedure was done without any intraoperative complication and the patients was discharged the day after operation and instructed for follow up 3 months after.

Histopathological analysis after surgery showed biphasic lesion of Antoni A and Antoni B cell. Cells are narrow and elongated with palisading arrangement (verocay body) which were consistent for schwannoma. Further immunohistochemical examination showed positive staining for S100, and showed negative staining for CD34, GFAP, EMA, Desmin and SMA, confirming the final diagnosis of intraosseous schwannoma. Three months after the surgery, the patient had no complain, no post-operative complications and was able to walk normally without pain. The patient was further scheduled for follow up on 6 months and then routinely every year post-operation to evaluate any signs of complication or recurrence (Figs. 4–6).

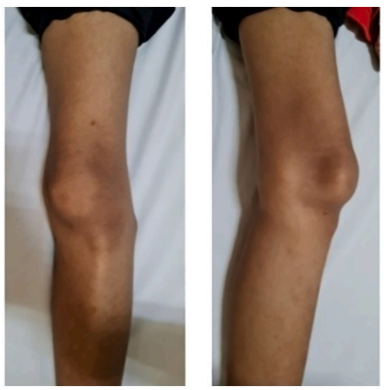

**Fig. 1.** Photograph of patients left extremity.

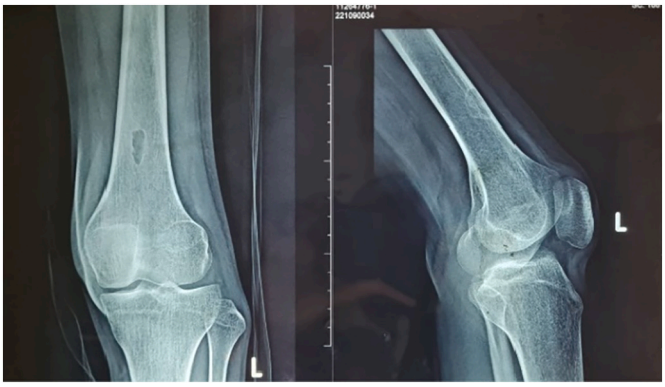

**Fig. 2.** A plain radiograph of the left distal femur showed a lytic lesion with sclerotic rim. (Left) anterior view. (Right) lateral view.

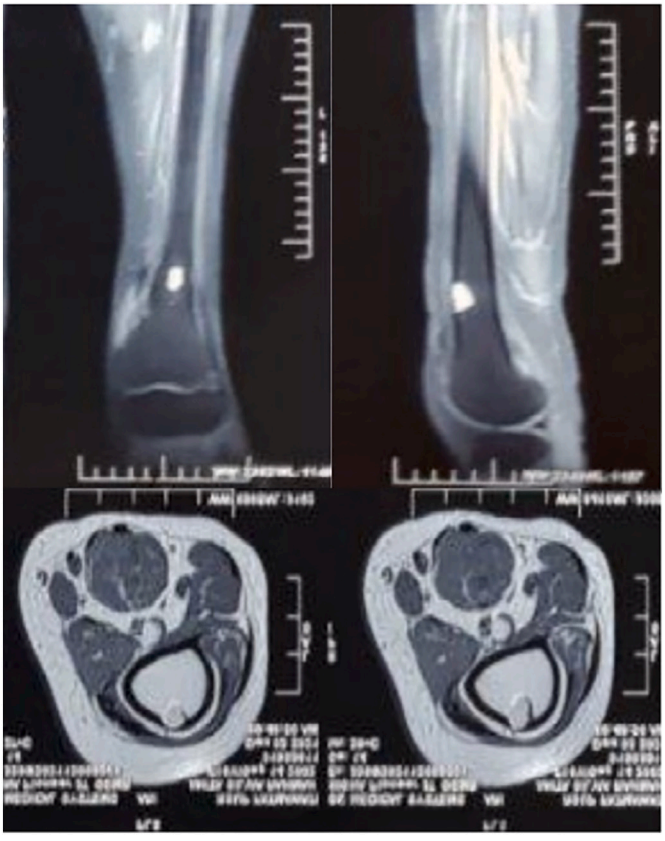

**Fig. 3.** T2 weighted MRI images of the distal femur showed hyperintense signal on both coronal and sagittal view. T1 weighted MRI images axial view showed isointense lesion with hypointense margin.

## 3. Discussion

Schwannoma is a slow growing benign tumor derived from Schwann cells, sheaths that cover myelinated sensory nerve fibers. Patients are usually asymptomatic and swelling is the common complain at the presentation, though pain might also appear in some cases. Head and

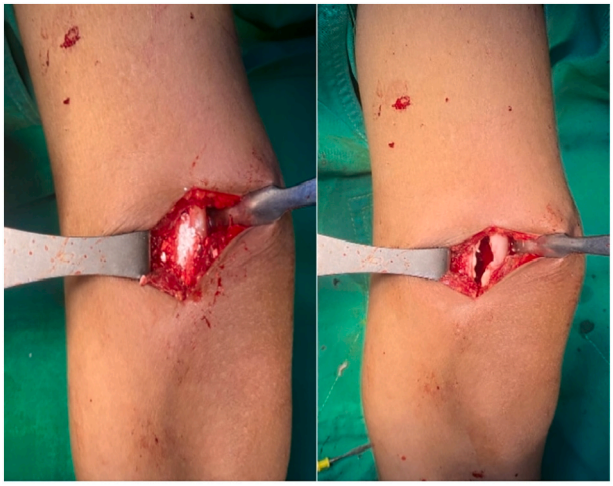

**Fig. 4.** Intraoperative picture of the curettage and then fill it with synthetic bone graft.

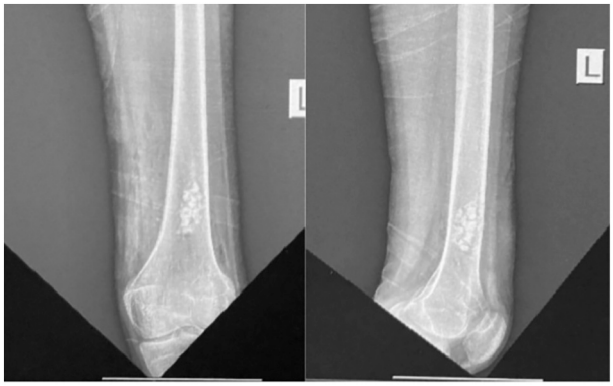


**Fig. 5.** Follow up X-ray 3 months after the surgery showing no signs of post-operative complication.

neck are the most common predilection for schwannomas. Intraosseous schwannomas are the rare variants of schwannomas that accounts for approximately less than 0.2 % cases among the primary bone tumors. Majority of intraosseous schwannomas appears on the mandible, but it can also occur in the long bones (tibia, femur, humerus, radius and ulna). Kito and his colleagues suggested that intraosseous schwannomas may involve bones tissue through 3 mechanisms: first, intramedullary by producing rarefaction of the bone. Second, may be in the nutrient canal. And third, extraosseous which erodes the bone [4,6,7].

Intraosseous schwannoma shows frequent predilection to myelinated nerves, especially sensory nerves. Thus, the frequent appearances in mandible were concluded because the mandible canals consist of sensory nerves of the trigeminal canal origins. While on the long bones intraosseous schwannoma is suggested to have arisen through the nutrient canals [4,6,8].

Plain radiograph examination of intraosseous schwannoma manifest as a well circumscribed lytic bone lesion, with a sclerotic rim, which occasionally destroys the bone cortex. There is no calcification or bone tissue formation within the lesion. MRI examination usually shows solid lesion mostly to be isointense to skeletal muscle on T1 weighted image and hyperintense and/or heterogenous on T2 weighted image. Cortical involvement of the lesion also could be seen [9,10]. These radiological descriptions match our imaging result for this patient. However, these imaging finding are nonspecific. The differential diagnosis includes benign bone lesions such as bone cyst, osteoblastoma, fibrous dysplasia, non-ossifying fibroma. The final diagnosis was not made until histologic examination of tissue obtained.

The evaluation of the biopsy sample under the microscope is the most important evaluation in establishing a definitive diagnosis for intraosseous schwannomas. The histological findings of intraosseous schwannomas are similar to the soft tissue schwannomas with two major components which consists of: a highly ordered cellular component (Antoni A) and a loose myxoid component (Antoni B), and some cases present what is called a verocay bodies which is form from a nuclear palisade alternating with fibrillar areas. Then the diagnosis is supported by the findings for S100 protein on immunohistochemical evaluation [1,2,6,8].

Intraosseous schwannoma is treated by a procedure of intralesional curettage and then fills the cavity with bone cement (PMMA) or bone graft. Bone graft is preferable than the use of bone cement. Recurrence is very rare if the lesion is completely removed, and no malignant transformation has ever been reported. For this patient we performed a curettage and the use of synthetic bone graft application [7,8].

## 4. Conclusions

Intraosseous schwannoma of the distal left femur is a very rare case. Which is one of some reason explaining difficulty to make an accurate initial diagnosis of intraosseous schwannomas. Hence it is important to perform histopathological analysis. In this case curettage of the mass followed by synthetic bone graft application was performed as there were no sign of malignancy making more invasive option such as excision or resection deemed to cause more harm than good to the patient.

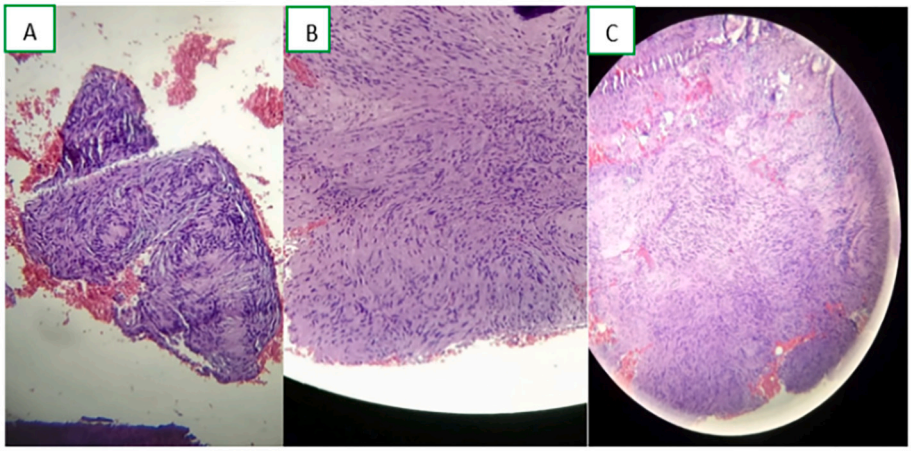


**Fig. 6.** Histopathology result of the lesion showing verocay body (A), Antoni A (B) and Antoni B (C) which were consistent for schwannoma.

## Consent for publications

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

## Provenance and peer review

Not commissioned, externally peer reviewed.

## Ethical approval

N/A.

## Funding

This publication did not receive any funding from agencies, commercials nor not-profit sectors.

## Guarantor

Muhammad Wahyudi is the sole guarantor of this submitted article.

## Research registration number

N/A.

## CRediT authorship contribution statement

Muhammad Wahyudi: Conceived and designed the publication, Contributed data
Ramadan Premiarto Clevfirstarachma: Collected the data, Wrote the paper
Mardiani Djailani: Contributed data.

## Declaration of competing interest

None.

## References

[1] F. Al-Lhedan, Schwannoma of the femur: a rare case report, J. Bone Oncol. 1 (8) (2017 Sep) 1–3.
[2] G.C. Drumond, S.A. Nakagawa, F.D. Costa, M.Y. Souza, J. Comunello, W.T. Chung, Schwannoma intraósseo: relato de caso e revisão da literatura, Rev. Bras. Ortop. 15 (55) (2020 May) 258–262.
[3] B. Haberal, D. Turkbey Simsek, E.K. Simsek, Intraosseous schwannoma of the calcaneus: a rare tumor of the bone, Case Rep. Orthop. 2 (2018 Oct) 2018.
[4] J. Huajun, Q. Wei, W. Yuxuan, Y. Jingjing, Intraosseous schwannoma of the proximal humerus with pathologic fracture, Eur. J. Med. Res. 26 (1) (2021 Dec) 1–8.
[5] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
[6] M. Kito, Y. Yoshimura, K.I. Isobe, K. Aoki, T. Momose, H. Kato, Intraosseous neurilemmoma of the proximal ulna, Int. J. Surg. Case Rep. 5 (12) (2014 Jan 1) 914–918.
[7] P. Reyniers, H. Wafa, F. Sinnaeve, P. Debeer, R. Sciot, Intraosseous schwannoma of the glenoid: case report and literature review, SICOT-J. 7 (2021).
[8] K. Suzuki, T. Yasuda, K. Watanabe, M. Kanamori, T. Kimura, Association between intraosseous schwannoma occurrence and the position of the intraosseous nutrient vessel: a case report, Oncol. Lett. 11 (5) (2016 May 1) 3185–3188.
[9] X.J. Wang, K. Hartley, G.E. Holt, O. Fadare, J.M. Cates, Intracortical schwannoma of the femur, Skelet. Radiol. 43 (5) (2014 May) 687–691.
[10] T. McAleese, K. Clesham, D. Moloney, A. Hughes, N. Faheem, K. Merghani, Intraosseous schwannoma of the femur in a patient with monoclonal gammopathy of undetermined significance, Int. J. Surg. Case Rep. 1 (72) (2020 Jan) 494–498.