




Intraosseous Lipoma of the Calvaria in the Early Stage Resembling Normal Fatty Marrow

Shunsuke Nagase¹  Kanako Ogura¹ Karin Ashizawa¹ Asumi Sakaguchi¹ Shiori Hotchi¹
Makoto Hishii² Masaharu Fukunaga³ Toshiharu Matsumoto¹

¹ Department of Diagnostic Pathology, Juntendo University Nerima Hospital, Tokyo, Japan

² Department of Neurosurgery, Juntendo University Nerima Hospital, Tokyo, Japan

³ Department of Pathology, Shin-Yurigaoka General Hospital, Kanagawa, Japan

Address for correspondence Shunsuke Nagase, MD, Department of Diagnostic Pathology, Juntendo University Nerima Hospital, 3-1-10 Takanodai, Nerima-ku, Tokyo 177-8521, Japan
(e-mail: s-nagase@juntendo.ac.jp).

J Neurol Surg Rep 2022;83:e29–e32.

Abstract

Intraosseous lipoma (IOL) is a benign bone tumor that usually arises from the lower limb and rarely arises from the skull. Radiological diagnosis of a typical case is not problematic due to its characteristic calcification and marginal sclerosis. Here, we report a case of calvarial IOL in the early stage lacking conventional radiopathological features. The patient is a 7-year-old girl who presented with a slow-growing protuberance on the vertex of the head. Computed tomography displayed a low-density mass without calcification that was continuous with the surrounding diploe. The mass was resected piece by piece for diagnostic and cosmetic reasons. Histologically, the specimen consisted of bony trabeculae and intertrabecular adipose tissue, which resembled normal fatty marrow. However, adipose tissue was considered neoplastic since it lacked hematopoietic elements. The final diagnosis of IOL was made by radiopathological correlation. This case suggests that IOL should be included in the differential diagnosis of diploic expansion, even if calcification is absent. The histology of an early-stage IOL resembles normal fatty marrow, but recognizing the absence of hematopoietic elements aids the diagnosis. Also, our literature review indicates that such cases are likely to be encountered in the calvaria than cranial base.

Keywords

- early
- intraosseous
- lipoma
- skull
- calvaria
- diploe

Introduction

Intraosseous lipoma (IOL) is a rare benign tumor consisting 0.1 to 2.5% of all primary bone tumors.^{1,2} Common sites are the calcaneus, femur, and tibia, and only 4% of the IOLs occur in the skull.³ IOLs of the skull are subdivided into three groups according to their location: the calvaria, cranial base, and facial skeleton (► **Fig. 1**). The common radiological features are calcification (57%), marginal sclerosis (74%),

and cyst formation (67%). Although IOL shows various histological changes such as atrophic bony trabeculae, fat necrosis, myxoid stroma, and calcification,¹ most typical cases are diagnosed by imaging alone without invasive procedures such as biopsy and resection.^{3,4} Here, we report a case of calvarial IOL in the early stage, presenting only an expansion of the diploe without conventional radiopathological features. We also review the literature and probe into the characteristics of calvarial IOLs.

received
February 12, 2021
accepted
October 19, 2021

DOI <https://doi.org/10.1055/s-0042-1747972>.
ISSN 2193-6358.

© 2022. The Author(s).

This is an open access article published by Thieme under the terms of the Creative Commons Attribution-NonDerivative-NonCommercial-License, permitting copying and reproduction so long as the original work is given appropriate credit. Contents may not be used for commercial purposes, or adapted, remixed, transformed or built upon. (<https://creativecommons.org/licenses/by-nc-nd/4.0/>)

Georg Thieme Verlag KG, Rüdigerstraße 14, 70469 Stuttgart, Germany

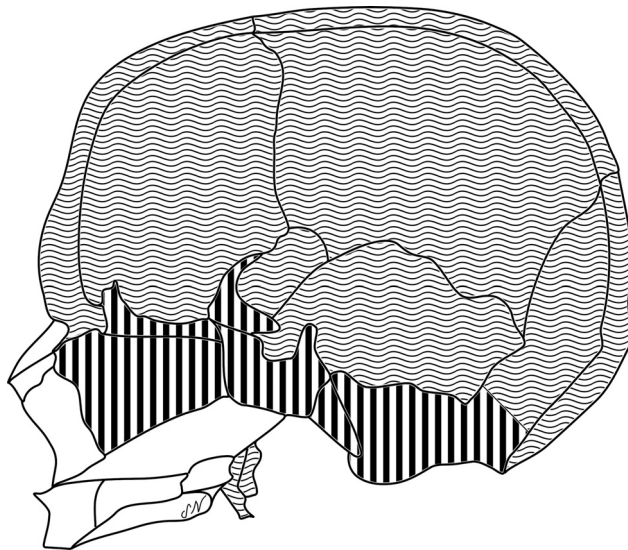


Fig. 1 Illustrated midsagittal section of the skull. Calvaria is filled with horizontal wave pattern; cranial base is filled with vertical stripe pattern. Facial skeleton is unfilled.

Case Report

The patient is a 7-year-old girl who was followed-up for a firm protuberance at the vertex of the head. She initially presented to our hospital at the age of two when her mother realized that the hump had been growing slowly in the past 1 year. The patient had no other medical history or known family history. On examination, the hump was as firm as a bone while the skin was intact.

Plain skull radiography displayed a radiolucent mass at the vertex of the skull (►Fig. 2A, B). Computed tomography (CT) illustrated a spindle-shaped low-density mass that was continuous with the surrounding diploe (►Fig. 2C, D). On magnetic resonance imaging, the mass was 34 mm in diameter and 8 mm in thickness showing homogeneous high signal intensity on T1- and T2-weighted images (►Fig. 2E, F). Calcification was not evident in any modality. Since the mass continued to expand slowly, surgery was considered at the age of seven for diagnostic and cosmetic reasons.

Intraoperative findings revealed that the surface of the outer table was intact, yet the diploe was thickened (►Fig. 3). The lesion displayed an indistinct border with a yellow cut-surface resembling a normal cancellous bone. It was resected piece by piece by a diamond drill, and all specimens were submitted for a pathological study. The internal plate was preserved as it appeared to be normal.

Microscopically, most areas (95%) consisted of trabeculae of lamellar bone and intertrabecular adipose tissue reminiscent of normal fatty marrow (►Fig. 4A, B). However, the adipose tissue seemed monotonous and lacked hematopoietic elements. There were also focal areas (5%) exhibiting a transition to a preexisting marrow, which included hematopoietic elements (►Fig. 4C, D). These findings indicated the neoplastic feature of the intertrabecular adipocytes. The final diagnosis of IOL was made by radiopathological correlation.

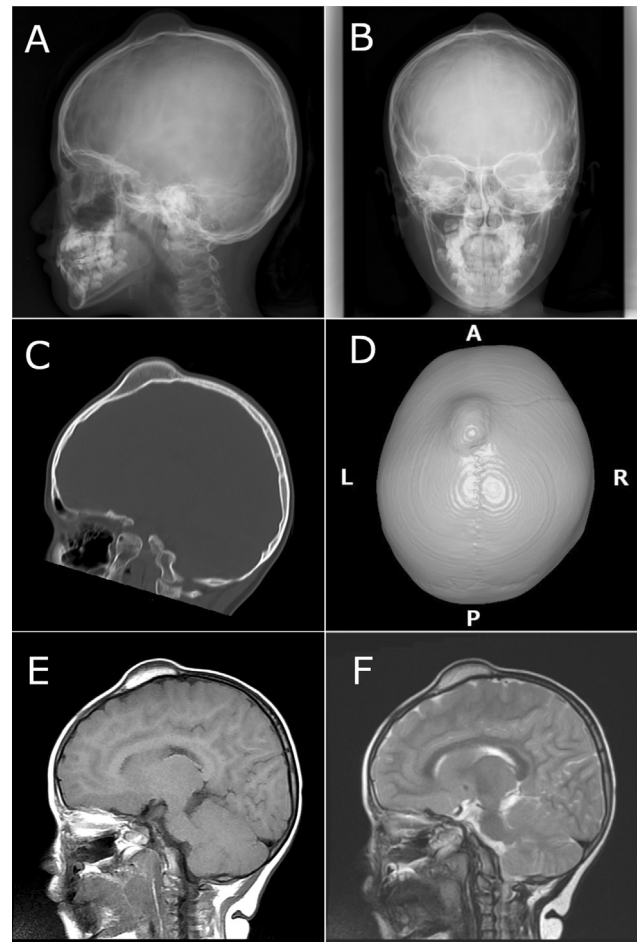


Fig. 2 Plain lateral (A) and frontal (B) skull radiographs display a radiolucent mass at the vertex of the skull. Sagittal computed tomography (CT) scan (C) illustrates a spindle-shaped low-density mass expanding the diploe, and three-dimensional CT reconstruction (D) renders the mass located at the intersection of the coronal and the sagittal suture. Magnetic resonance imaging shows high signal intensity on sagittal T1- (E) and T2-weighted (F) images.

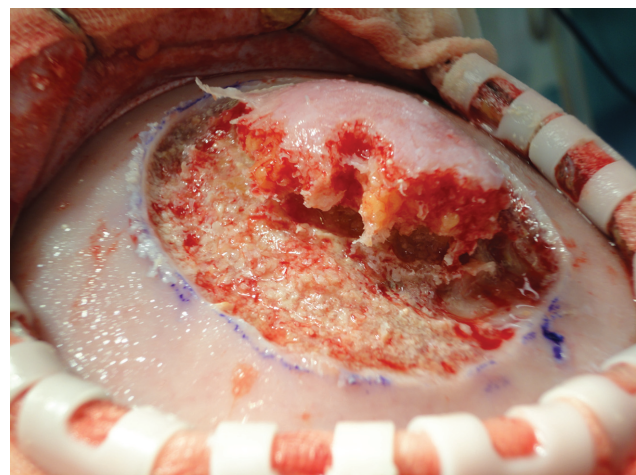


Fig. 3 Intraoperative photograph reveals the expanded diploe resembling a normal cancellous bone.

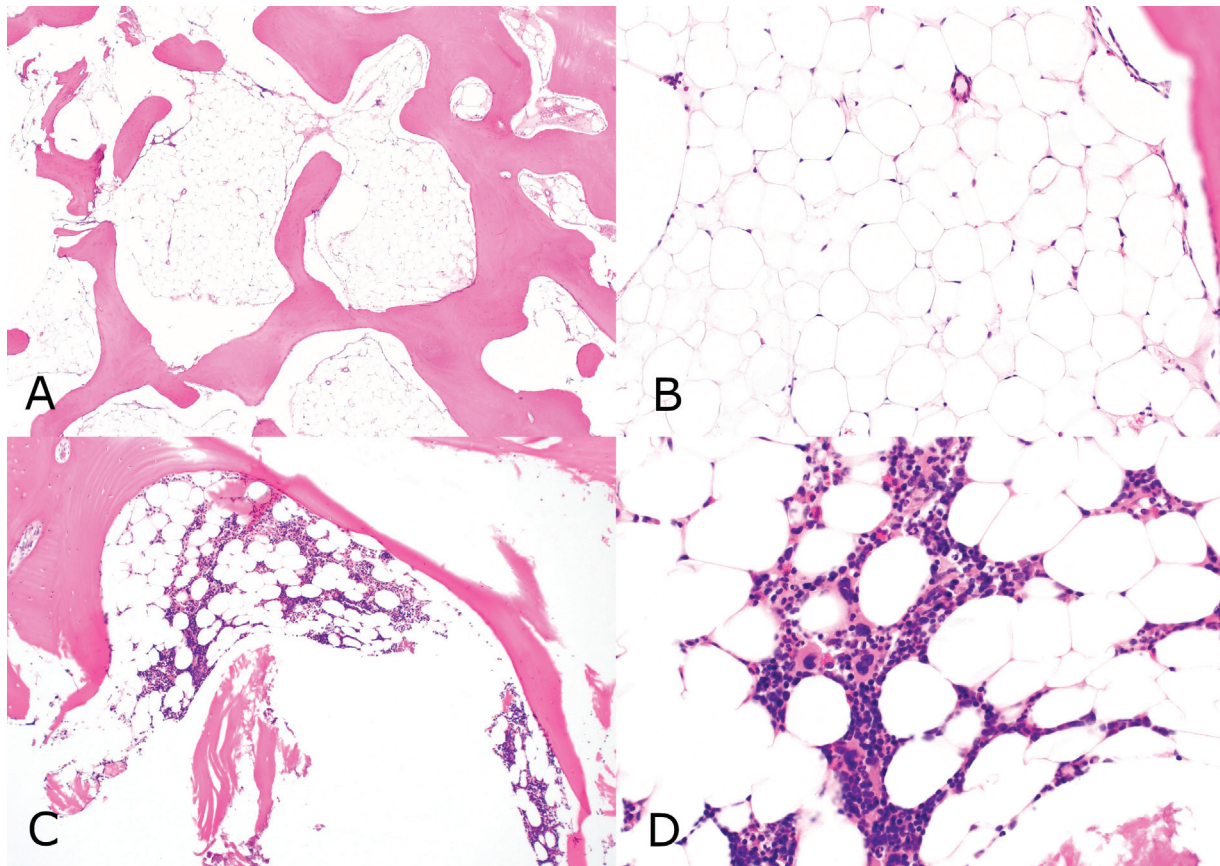


Fig. 4 Microscopic image of the lesion, which consists of mature adipose tissue and trabeculae of lamellar bone resembling a normal fatty marrow (A). Intertrabecular adipose tissue seems monotonous and lacks hematopoietic elements (B). The area that is regarded as a preexisting marrow (C) contains hematopoietic elements (D).

The postoperative course was uneventful, and the patient was discharged 3 days after the operation. No sign of recurrence was observed in the follow-up after 1 year.

Discussion

To date, 18 cases of solitary IOLs involving the calvaria and cranial base have been reported in the literature.^{5–21} Twelve cases, including our case, evolved from the calvaria, whereas seven cases evolved from the cranial base. The median ages of onset and diagnosis were 3 and 26.5 in calvarial IOLs and 37.5 each in IOLs of the cranial base; the calvarial cases were observed at a younger age. The most remarkable difference was that only four cases of calvaria showed calcification, while all cases of the cranial base, except for one indeterminate case, showed calcification (33 vs. 100%, Fisher's exact test, $p = 0.013$; ► **Table 1**). Also, among the calvarial

Table 1 Number of intraosseous lipoma of the calvaria and the cranial base

	Calcified	Noncalcified
Calvaria	4	8
Cranial base	7	0

Note: Fisher's exact test, $p = 0.013$.

Table 2 Details of the calvarial intraosseous lipoma

	Calcified	Noncalcified
Number of cases (male:female)	4 (2:2)	8 (3:5)
Median age of onset (range)	17 (16–18)	1 (0–36)
Median age of diagnosis (range) ^a	63 (50–67)	16 (3–36)

^aMann–Whitney U test, $p = 0.008$.

IOLs, the median age of diagnosis in noncalcified cases was significantly lower than that of the calcified cases (16 vs. 63, Mann–Whitney U test, $p = 0.008$; ► **Table 2**). In brief, IOLs of the calvaria are observed at a younger age and are less calcified than those of the cranial base; among the calvarial IOLs, younger patients are likely to present without calcification.

The natural history of IOL has been controversial. In the largest case series of IOL, Milgram proposed a three-tier staging system based on his hypothesis that the tumor undergoes involutional changes over time resulting in various radiopathological findings.¹⁴ This theory was questioned by Campbell et al, who argued that their cases of stages 2 and 3 were not dominantly observed in older patients and did not

retain the features of the stage 1 lesion.³ However, our literature review showed that calcification is more common in older patients among calvarial cases, and it does not contradict Milgram's hypothesis.

According to Chow and Lee, IOL originates from adipocyte proliferation, replacing the preexisting marrow and causing intramedullary pressure within the limited space. This pressure induces the atrophy of the bony trabeculae and other histological changes secondary to capillary occlusion.¹ Since our case presented without atrophic bony trabeculae and other secondary changes, we consider that it corresponds to the earliest stage of IOL.

Early-stage IOL poses a challenge to both radiological and pathological diagnoses. Rosenbloom and Osborne reported a purely lytic cranial IOL without calcification and suggested the limitation of radiography and CT due to a large number of differential diagnoses.¹⁶ Indeed, the differential diagnosis in our case included dermoid cyst, epidermoid cyst, fibrous dysplasia, ossifying fibroma, and cavernous hemangioma but not IOL. Regarding the histology, atrophy of bony trabeculae is reported to be a diagnostic finding in early-stage IOL.¹ However, our case did not present atrophic bony trabeculae, and the histology was almost indistinguishable from a normal fatty marrow.

The final diagnosis was made by recognizing the monotonous appearance of the intertrabecular adipose tissue, the paucity of hematopoietic elements, and its transition to a preexisting marrow. These findings were regarded altogether as *the absence of hematopoietic elements* by Eyzaguirre et al as one of the various histological findings discovered in IOL.⁷ However, our case is the first case in which the diagnosis was hinted only by the *absence of hematopoietic elements*. The case highlights the diagnostic value of this finding, especially in early-stage IOL, where a radiological diagnosis can be difficult.

Conclusion

We reported a case of calvarial IOL in the early stage presenting only an expansion of the diploe without conventional radiopathological features. The literature review illustrated that calvarial IOLs are discovered in young patients and rarely display calcification, indicating that most cases are in the early stage. Therefore, IOL should be included in the differential diagnosis in a young patient with an expanded calvarial diploe even if calcification is absent. Such cases resemble normal fatty marrow histologically, but recognizing the absence of hematopoietic elements aids the diagnosis.

Funding

None.

Conflict of Interest

None declared.

Acknowledgments

None.

References

- 1 Chow LT, Lee KC. Intraosseous lipoma. A clinicopathologic study of nine cases. *Am J Surg Pathol* 1992;16(04):401–410
- 2 Unni KK, Inwards CY, Research MFME. *Dahlin's Bone Tumors: General Aspects and Data on 10,165 Cases*. 6th edition. Wolters Kluwer Health/Lippincott Williams & Wilkins. Philadelphia, PA2009
- 3 Campbell RSD, Grainger AJ, Mangham DC, Beggs I, Teh J, Davies AM. Intraosseous lipoma: report of 35 new cases and a review of the literature. *Skeletal Radiol* 2003;32(04):209–222
- 4 Propeck T, Bullard MA, Lin J, Doi K, Martel W. Radiologic-pathologic correlation of intraosseous lipomas. *AJR Am J Roentgenol* 2000;175(03):673–678
- 5 Abdalla WM, da Motta AC, Lin SY, McCarthy EF, Zinreich SJ. Intraosseous lipoma of the left frontoethmoidal sinuses and nasal cavity. *AJNR Am J Neuroradiol* 2007;28(04):615–617
- 6 Elsurur C, Hanci D, Cakir A, Dal T. Isolated intraosseous lipoma of the ethmoid sinus. *J Craniomaxillofac Surg* 2013;41(08):718–720
- 7 Eyzaguirre E, Liqiang W, Karla GM, Rajendra K, Alberto A, Gatalica Z. Intraosseous lipoma. A clinical, radiologic, and pathologic study of 5 cases. *Ann Diagn Pathol* 2007;11(05):320–325
- 8 Han Z, Liu Y, Hao X, Li C, Tian Y. Large intraosseous lipoma of the skull: a case report and review of the literature. *World Neurosurg* 2018;120:525–529
- 9 Hazarika P, Pujary K, Kundaje HG, Rao PL. Osteolipoma of the skull base. *J Laryngol Otol* 2001;115(02):136–139
- 10 Jamrozik Z, Rosiak G, Kierdaszuk B, et al. Intraosseous lipoma of the sphenoid: a case study. *Case Rep Neurol Med* 2013;2013:519341
- 11 Kaneko Y, Hokao K, Ogasawara N, Nakano S, Masuda T. [A case of intraosseous lipoma in the frontal bone]. *No Shinkei Geka* 2011;39(10):975–980
- 12 Lanišnik B, Didanovič V. Sphenoclivar intraosseous lipoma: case report and literature review. *Skull Base* 2007;17(03):211–214
- 13 MacFarlane MR, Soule SS, Hunt PJ. Intraosseous lipoma of the body of the sphenoid bone. *J Clin Neurosci* 2005;12(01):105–108
- 14 Milgram JW. Intraosseous lipomas. A clinicopathologic study of 66 cases. *Clin Orthop Relat Res* 1988;(231):277–302
- 15 Nahles G, Schaeper F, Bier J, Klein M. An intraosseous lipoma in the frontal bone—a case report. *Int J Oral Maxillofac Surg* 2004;33(04):408–410
- 16 Rosenbloom SA, Osborne DR. Lipoma of the frontal bone. *AJNR Am J Neuroradiol* 1985;6(05):833–834
- 17 Small ML, Green WR, Johnson LC. Lipoma of the frontal bone. *Arch Ophthalmol* 1979;97(01):129–132
- 18 Solari D, Angel VF, Barcelò SR, et al. Intraosseous lipoma of the skull base, involving the sphenoclivar region: case report. *Clin Neurol Neurosurg* 2012;114(10):1368–1370
- 19 Srubiski A, Csillag A, Timperley D, Kalish L, Qiu MR, Harvey RJ. Radiological features of the intraosseous lipoma of the sphenoid. *Otolaryngol Head Neck Surg* 2011;144(04):617–622
- 20 Tomabechi M, Sako K, Daita G, Yonemasu Y. Lipoma involving the skull. Case report. *J Neurosurg* 1992;76(02):312–314
- 21 Yasuda Y, Tsukada S, Okada T, Haseda Y. Intraosseous lipoma of the skull: a report of two cases. *Ann Plast Surg* 1987;18(01):74–80