Neurochirurgie 65 (2019) 430–431

Disponible en ligne sur
ScienceDirect
www.sciencedirect.com

Elsevier Masson France
EM|consulte
www.em-consulte.com

Letter to the Editor

# Sacral Hibernoma: Presentation of a rare case and a treatment algorithm

*Hibernome sacré, présentation d'un cas rare et d'un algorithme de traitement*

## 1. Introduction

Mammalian adipose tissue includes two types of fat: white and brown. The latter facilitates the thermoregulation of hibernating animals [1,2]. In humans, the maximum proportion of brow fat is found in newborns [2,3], while it comprises only 0.1% of total body weight by the age of 70. Residual brown fat is embedded in the axilla, chest wall, mediastinum, retroperitoneum, perineum, paravertebral and adrenal regions [3,4] while it can also be found in 2% of soft tissue lipomas [1]. Hibernoma is a lesion that contains brown fat in significant proportion.

## 2. Case report

A 54-year-old Caucasian female presented in our outpatient department due to a 2-year history of low-back pain, extending to her left buttock and accompanied by causalgia. Her medical history included a surgically treated breast carcinoma 12 years previously, with no signs of recurrence. No sciatica, bladder or bowel dysfunction or other neurological deficit was found on neurological examination. The patient had already undergone a bone scan and SPECT with no pathological findings. MRI revealed a well-defined 1.4 × 1.3 cm ovoid intraosseous lesion at the 4th sacral vertebra, on the left side. The lesion was high intensity on T2 and STIR sequences, with intermediate low intensity on T1. It was also significantly enhanced by contrast injection (Fig. 1). Subsequently, CT was performed for better visualization of surrounding bone elements. The lesion had an osteolytic ground-glass appearance (Fig. 2). The patient underwent total lesion removal under X-ray guidance through a left paramedian sacral incision. She was mobilized 6 hours after the operation, with no neurological deficit. The histological specimen findings were characteristic of hibernoma (Fig. 3). The patient was discharged after 1 day's hospitalization. Postoperative course up to the time of writing was uneventful, without causalgia or other pain.

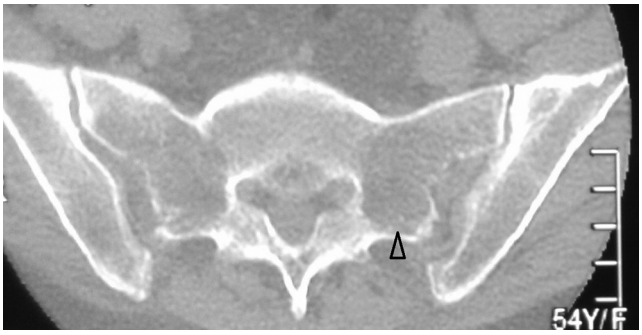


**Fig. 2.** CT scan demonstrating a round lesion with sclerotic margin (arrowhead) and hypodense core.

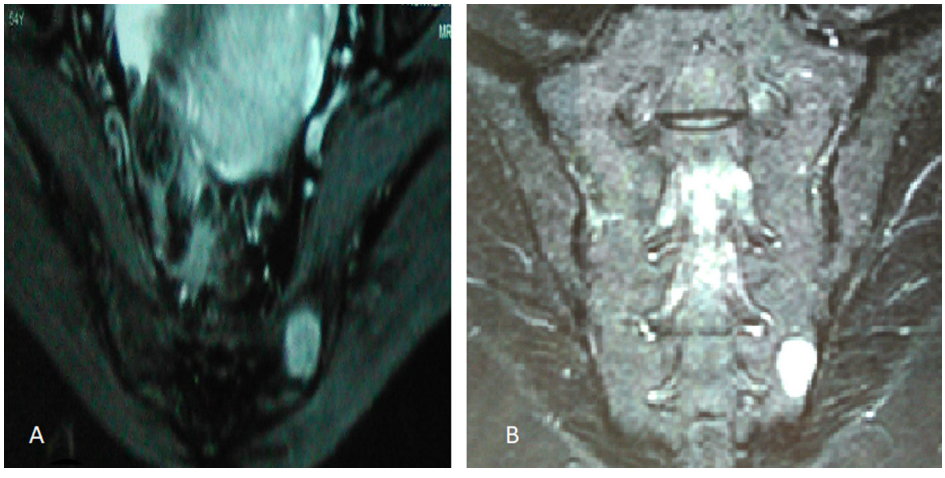


**Fig. 1.** MRI scan. A. Axial T2 sequence demonstrating a round high-intensity lesion within the sacrum. B. Coronal T1 sequence with gadolinium enhancement. Note the significant enhancement caused by the contrast agent.

https://doi.org/10.1016/j.neuchi.2019.06.002

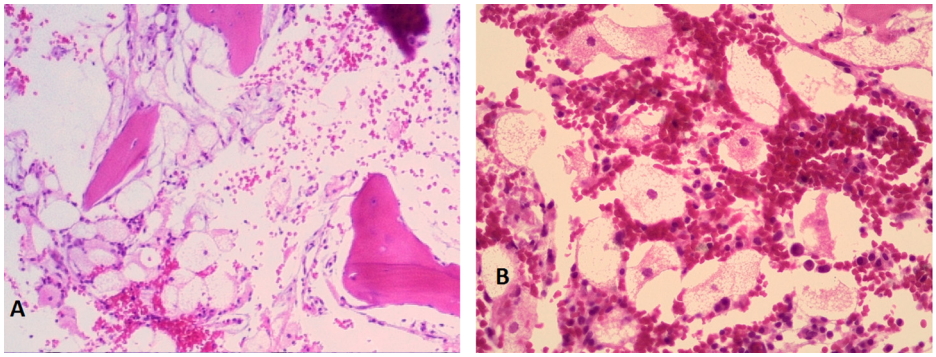


**Fig. 3.** Microscopic images of the intraosseous hibernoma. A. Hematoxylin and eosin image shows adipose cells within the lesion. B. Note the characteristic large cells with foamy cytoplasm of brown fat in contrast with the surrounding tissue.

## 3. Discussion

Hibernoma is a rare subtype (1.5%) of lipomatous tumor; intraosseous hibernomas are even rarer [5–7], the present case being only the 18th reported in the international literature.

Macroscopically, an intraosseous hibernoma resembles a brown soft elastic mass [4] due to its significant vascularity and high number of cytoplasmic mitochondria [4], and is surrounded by sclerotic bone.

MRI is considered the gold standard examination, although aspects are variable due to various histological subtypes. On plain radiographs or CT, the aspect is of a ground-glass lesion with sclerotic margins [5,7]. The radiological characteristics of the present case were typical: mildly hypointense on T1 sequences, with high signal on T2 and no suppression on STIR. Contrast medium significantly enhanced the lesion, which is uncommon but not unknown in the literature [1,2,7].

Differential diagnoses cover both benign and malignant osseous and adipose tissue lesions, such as bone island, hemangioma, metastatic carcinoma, lipoma, angiolipoma, elastofibroma, myxoid liposarcoma and liposarcoma [1,5].

Intraosseous hibernoma is detected incidentally. To our knowledge, our case is only the 9th symptomatic [1,2,4,6–10] and the 5th sacral [4,6,7,9] in the English-language literature. The main symptom in the other 8 cases was also persisting pain [1,2,4,6–10]. Additionally, in 1 case an intraosseous hibernoma was related to myeloproliferative disease [3].

Management of hibernoma is controversial. In contrast with their soft tissue counterparts, intraosseous hibernomas are considered a physiological deviation [5]. There are no documented cases of recurrence or metastatic spread after total removal [2]. Biopsy is advocated by some authors to diagnose hibernoma [2,7]. Others propose radiological follow-up, as extension is slow [5], and there is increased risk of hemorrhage during biopsy due to prominent vascularization.

Five out of the 8 symptomatic intraosseous hibernomas were treated conservatively; the lesion was removed, either by open surgery [8,10] or percutaneously [6], in only 3 cases, due to failure of conservative treatment or the risk of further bone erosion [10].

In the present case, the persistence of symptoms for 2 years despite analgesic treatment dictated resection. The postoperative pain relief justified this, even though most hibernomas are treated conservatively. It is up to the doctor to decide whether the radiological image of the suspect lesion is sufficient for diagnosis or to perform biopsy in an asymptomatic patient.

## Declarations of interest

The authors declare that they have no competing interest.

## Acknowledgment

Pr. Dimitris Konstantinou was the senior surgeon during the procedure described above. He was the chairman of the Patras University Neurosurgery Clinic and my tutor. He passed away on Mount Aconcagua in the Andes last year.

## References

[1] Bai S, Mies C, Stephenson J, Zhang PJ. Intraosseous hibernoma: a potential mimic of metastatic carcinoma. Ann Diagn Pathol 2013;17:204–6.
[2] Botchu R, Puls F, Hock YL, Davies AM, Wafa H, Grimer RJ, et al. Intraosseous hibernoma: a case report and review of the literature. Skeletal Radiol 2013;24(7):1003–5.
[3] Thorns C, Schardt C, Katenkamp D, Kahler C, Merz H, Feller AC. Hibernoma-like brown fat in the bone marrow: report of a unique case. Virchows Arch 2008;452(3):343–5.
[4] Westacott L, Collins A, Dickenson I. Intraosseous Hibernoma in the sacrum of an adult. Int J Surg Pathol 2016;24(8):749–52.
[5] Bonar SF, Watson G, Gragnaniello C, Seex K, Magnussen J, Earwaker J. Intraosseous hibernoma: characterization of five cases and literature review. Skeletal Radiol 2014;43(7):939–46.
[6] Ringe KI, Rosenthal H, Länger F, Callies T, Wacker F, Raatschen HJ. Radiofrequency ablation of a rare case of an intraosseous hibernoma causing therapy-refractory pain. J Vasc Interv Radiol 2013;24(11):1754–6.
[7] Kumar R, Deaver MT, Czerniak BA, Madewell JE. Intraosseous hibernoma. Skeletal Radiol 2011;40(5):641–5.
[8] Yahia M, Laabidi B, M'sakni I, Bougrine F, Bouziani A. Intraosseous hibernoma: a case report and review of the literature. J Afr Cancer 2015;7:259–62.
[9] Jerman A, Snoj Z, Kuzmanov BG, Limpel Novak AK. Intraosseous hibernoma: case report and tumour characterization. BJR Case Rep 2015;1:20150204.
[10] Vlychou M, Teh J, Whitwell D, Athanasou NA. Intraosseous hibernoma: a rare adipocytic bone tumour. Skeletal Radiol 2016;45:1565–9.

K. Faropoulos [a,*]
K. Apostolopoulou [a]
M. Gkermpesi [b]
[a] *Neurosurgical Clinic, University Hospital of Patras, Patras, Greece*
[b] *Department of Pathology, University Hospital of Patras, Patras, Greece*

* Corresponding author at: Iouliou Typaldou 15, 1077 Nicosia, Cyprus.
*E-mail address:* konfaropoulos@gmail.com (K. Faropoulos)

Received 28 March 2019

Received in revised form 1st June 2019

Accepted 6 June 2019
Available online 25 June 2019