Case Report

A Rare Case of an Osteoid Osteoma of the Cervical Spine

Francisco Silva Gomes^{a, b} Cristina Alves^{a, b} Pedro Sa Cardoso^{a, b}
Tah Pu Ling^{a, b}

^aHospital Distrital da Figueira da Foz, Figueira da Foz, Portugal; ^bServiço de Ortopedia Pediátrica do Hospital Pediátrico, Centro Hospitalar e Universitário de Coimbra, Coimbra, Portugal

Keywords

Osteoid osteoma · Cervical spine · Posterior laminectomy · Cervical arthrodesis

Abstract

Osteoid osteoma (OO) is a benign bone tumor rarely affecting the cervical spine. OO, currently diagnosed by X-ray, computed tomography (CT), bone scan, and magnetic resonance imaging, is difficult to identify when located in the cervical spine based on spine radiographs due to their usually small size and the complex anatomy of the cervical spine. CT scans successfully diagnose 20–30% of the small osteolytic lesions with dense sclerotic rings and central calcifications and the anatomic location of the nidus. CT-guided radiofrequency ablation is a noninvasive treatment option widely used. However, in specific cases, surgical resection of the nidus is recommended. We present a rare clinical case of cervical spine OO of a 16-year-old female patient. The lesion was located on the left lamina of C7, and once the diagnosis was established by physical examination and imaging, laminectomy and cervical arthrodesis (C6-T1) were performed. Twelve months after the surgical intervention, the patient showed complete remission of the symptoms and no disabilities.

© 2020 The Author(s)
Published by S. Karger AG, Basel

Introduction

Osteoid osteoma (OO), a rare bone-producing tumor, can occur anywhere in the skeleton, [1, 2] and in the spine it is frequently located in the posterior elements involving the lamina, pedicles, or the transverse and spinous processes [3, 4]. Very rarely, OO develops in the cervical spine [1], being more common in children and young patients, with 90% of the patients being aged between 5 and 25 years [5].

The early diagnosis of OO is difficult, given the deceptive symptoms and the young age of most patients, postponing complementary studies [6–8]. The first symptom is generally pain, with patients' reporting a moderate intermittent pain near the tumor that is more severe at night. In the early stages of the disease, patients feel a temporary pain relief with nonsteroidal anti-inflammatory drugs (NSAIDs). However, cervical pain worsens becoming no longer responsive to analgesics.

Plain X-ray is usually the first imaging choice [5]. However, it is not very informative when the OO is located in the cervical spine because of the tumor's small size and the complex anatomy of its setting. Computed tomography (CT) successfully diagnoses 20–30% of the small osteolytic lesions with dense sclerotic rings and central calcifications. CT is a precise technique as it reveals the anatomical location of the nidus with an accuracy superior to that of magnetic resonance imaging (MRI) [5, 9]. Bone scan is also used in OO diagnosis, especially when the radiographic results are ambiguous or negative [10, 11]. However, the false-negative bone scans and reduced sensitivity for bone marrow disease confines bone scan use [12, 13].

CT-guided radiofrequency ablation (RFA) is considered the firstline therapy since the early 1990s [14]. In spite of its advantages over surgery and the facility to treat high-risk localizations (e.g., spine) [15–17] with a success rate ranging from 94 to 100%, RFA is only indicated for spinal OO lesions with no neurological deficits, intact cortical bone, and abundant cerebrospinal fluid around the lesion to prevent neurological damage by heat. Otherwise, surgery is the treatment of choice [1, 18, 19], provided the vertebral canal is not within the ablation zone, and the approach (anterior or posterior) to be used is selected according to the location of the tumors [20, 21].

If surgery is the treatment option, to avoid OO recurrence, complete surgical excision of the nidus is necessary. Quite often, this is difficult due to the risk of iatrogenic instability or neurological injury [22–24]. When complete resection of the tumor is not possible, many authors propose selective embolization and, for aggressive tumors, radiotherapy as adjuvant therapies [19, 22].

Herein we present a rare case of cervical OO, on the left lamina of C7, of a 16-year-old female patient, who underwent surgical treatment with complete resection by C7 laminectomy and cervical arthrodesis (C6-T1).

Methods

The OO diagnosis was obtained by physical examination of the patient in conjunction with X-ray followed by MRI, CT scan, and single-photon emission computed tomography (SPECT)-CT. The surgical excision of the lesion was performed using the posterior approach. The histological examination of the resection specimen was used to confirm the diagnosis.

Results

A 16-year-old female patient presented to the outpatient clinic with cervical pain and torticollis, more intense on the left side. The symptoms started 1 year before, and worsened progressively. X-ray revealed a segmental inversion of the cervical lordosis at C6-T1; however, it was not possible to identify a corresponding bone lesion (Fig. 1). MRI showed a focal area with hyperintense signal in T2-weighted images and short tau inversion recovery, hypointense signal in T1-weighted images, and enhancement after contrast in the left vertebral body of C7 (Fig. 2). Subsequent bone scan revealed a focus of intense osteoblastic hyperactivity in the left lamina of the C7, compatible with the diagnosis of OO (Fig. 3). This lesion was confirmed and better defined on the CT scan and SPECT-CT (Fig. 4, 5). The patient's age and the central location of the edema on the left lamina of C7, where it was possible to observe a sclerotic lesion with a peripheral ring and sclerotic lamina, also suggested an OO, as this type of tumor is usually round or ovoid, with a diameter <2 cm, a nidus and well-defined margins, and flexible adjacent osteosclerosis [25]. As symptoms were persistent, surgical treatment was offered. The surgical excision of the lesion was performed through a posterior approach. This technique allowed the execution of a laminectomy, followed by cervical arthrodesis (C6-T1), in order to avoid instability and iatrogenic kyphosis (Fig. 6). 12 months after surgical resection, the patient had no pain and scored 1% on the Neck Disability Index.

Discussion

OO tumors are more frequent in children and young adults. Rarely, this type of tumor appears in the cervical spine and may evolve towards an osteoblastoma [26]. It is very difficult to diagnose an OO in the cervical spine because of its small size, the complex anatomy of the cervical spine, and the obfuscation by the overlying soft tissue [27]. Due to the small size of these tumors and their location, the diagnosis may be inaccurate, particularly if thin-slice CT scan is not performed. It may take several months to get an accurate diagnosis after the first clinical symptoms [28, 29]. Tumors of a small size are very difficult to detect by MRI as the nidus may be covered by the fibrovascular zone, reactive sclerosis, and the presence of edema in the bone [30–32]. As bone scan has a limited diagnostic specificity and reduced sensitivity for OO, false-negative cases have been reported [12, 13].

This study reports the case of a 16-year-old female patient with intense cervical spine pain, worst at night, which was partially relieved with ibuprofen. Even though X-ray revealed only a segmental inversion of the cervical lordosis, the hypothesis of a spine OO emerged as causative of the patient's symptoms due to the painful and stiff spine deformity in a young patient and the fact that the patient's pain was relieved by NSAIDs [33]. CT scan and MRI images identified a sclerotic lesion at C7, with surrounding edema of the adjacent tissues, compatible with an OO. Yet, although OO has particular characteristics on the CT, only the tumor's histology provided a definitive diagnosis.

Although OO may have spontaneous resolution between 2 and 8 years after its initial symptoms, the frequent and severe pain and the risk of evolution to osteoblastoma [26, 34] prompted us to offer surgical resection as the treatment option for this patient. Although RFA is the first-line therapy with a success of 94%, its use is usually recommended when there are no neurological deficits, complete bone cortex around the lesion on CT, and abundant cerebrospinal fluid between the lesion and nerve root or spinal cord (>1.0 mm) to avoid neurologic injury by heat [35]. Following the patient's careful examination, the option for surgical

resection was taken considering the risk of surgical sequelae and the risk associated with RFA ablation due to the tumor's localization and recurrence due to incomplete ablation.

En bloc tumor resection was performed by laminectomy. It was necessary to violate the facet joint with significant resection of the superior articular facet. As a significant portion of the articular facet was resected, a cervical arthrodesis (C6-T1) was performed to avoid instability and secondary neurologic injury [14]. This procedure allowed the maintenance of stability of the posterior spine, which was potentially compromised by the lesion of the facet joint. Moreover, this resection was performed in a transitional level with a high risk of post-laminectomy kyphosis.

The diagnosis was confirmed by the histological examination of the resection specimen (Fig. 7), and the patient's follow-up revealed complete resolution of her long-standing cervical pain and disabilities 12 months after surgical excision of the OO.

Conclusion

Benign cervical spine bone-producing tumors are uncommon, but their presence should be considered in children, teenagers, and young adults with stiff spine deformity and persistent cervical pain worsening during the night and partially being relieved with NSAIDs. The diagnosis of OO requires a careful physical examination and appropriate imaging to ascertain the tumor characteristics and to consider different treatment options.

Although OO may regress spontaneously by unknown mechanisms, the persistence of pain and its long-term responsiveness to NSAIDs is an indication that the tumor should be removed. In the case reported, surgery was the treatment option to remove the cervical spine OO of a young female to avoid neurologic damage and incomplete tumor ablation. Complete resection of the lesion and resolution of the symptoms were achieved, and 12 months after tumor excision, the patient showed a good functional result.

Acknowledgements

The authors thank the parents of our patient and the patient herself for their support and permission to publish this report.

Statement of Ethics

This manuscript complies with the guidelines for human studies and research and was conducted ethically in accordance with the World Medical Association Declaration of Helsinki. The patient's parents and, very recently, the patient gave their written informed consent to publish this case including the images.

Disclosure Statement

All authors declare that there are no financial and personal relationships with people or organizations that could inappropriately influence their work. Therefore, all authors declare there are no conflicts of interest to disclose.

Funding Sources

Authors worked for Serviço de Ortopedia Pediátrica do Hospital Pediátrico, Centro Hospitalar e Universitário de Coimbra, Coimbra, Portugal, which provided their corresponding salaries. No additional funding was given nor required since all expenses were supported by the Serviço de Ortopedia Pediátrica do Hospital Pediátrico.

Author Contributions

F.S.G. was involved in the surgical intervention, collected the data, and wrote the first draft of the manuscript. C.A. reviewed the manuscript and the patient data, and critically reviewed and edited the drafts. P.C. and T.P.L. are members of the spine unit, and T.P.L. is the coordinator of the unit. Both were involved in the clinical evaluation of the patient, surgical planning, intervention, and follow-up.

References

- 1 Kan P, Schmidt MH. Osteoid osteoma and osteoblastoma of the spine. *Neurosurg Clin N Am*. 2008 Jan;19(1):65–70.
- 2 Brook S, Piccora R. Osteoid osteoma: diagnosis and treatment. Available from: <https://www.scribd.com/document/37039097/Osteoid-Osteoma>.
- 3 Thakur NA, Daniels AH, Schiller J, Valdes MA, Czerwein JK, Schiller A, et al. Benign tumors of the spine. *J Am Acad Orthop Surg*. 2012 Nov;20(11):715–24.
- 4 Zileli M, Cagli S, Basdemir G, Ersahin Y. Osteoid osteomas and osteoblastomas of the spine. *Neurosurg Focus*. 2003 Nov;15(5):E5.
- 5 Khan NA, Chandramohan M. (2005). Osteoid Osteoma. eMedicine [serial online]. Available from: www.emedicine.com/radio/topic498.htm.
- 6 Dezen EL, Avanzi O, Salomão JC, Pinto Filho WC. Tumores benignos da coluna cervical. *Rev Bras Ortop*. 1994;29(3):94–8.
- 7 Rothman RH, Simeone FA, editors. *The spine*. Volume 2. 4th ed. Philadelphia: W.B. Saunders; 1992. pp. 1171–3.
- 8 Cecin HÁ. [Proposition of an anatomic and functional reserve into the spinal canal, as an interference factor on the medical degenerative low back pain and discopathy]. *Rev Assoc Med Bras* (1992). 1997 Oct-Dec;43(4):295–310.
- 9 Arriaza-Loureda R, Canto-Lelián B, Sampedrocurbera C. Presentación atípica de un osteoma osteoide. *Acta Orthop Gallega*. 2005;1(1):24–6.
- 10 Etchbehere M. (2005). Localização Intra-operatória do osteoma osteoide com auxílio de uma sonda de raios gama. PhD Thesis.
- 11 Wu GM. (1994). Osteoid Osteoma. Department of Radiology at Brigham and Women's Hospital. Available from: <http://brighamrad.harvard.edu/Cases/bwh/hcache/91/full.html>.
- 12 Fehring TK, Green NE. Negative radionuclide scan in osteoid osteoma. A case report. *Clin Orthop Relat Res*. 1984 May;(185):245–9.
- 13 Leroy V, Couturaud M, Lathelize H, Valette C, Beck C, Dunoyer J, et al. [Bone scintigraphy is unreliable in studying osteoid osteoma]. *Rev Rhum Mal Osteoartic*. 1980 Jan;47(1):53–6.
- 14 Rosenthal DI, Hornicek FJ, Wolfe MW, Jennings LC, Gebhardt MC, Mankin HJ. Percutaneous radiofrequency coagulation of osteoid osteoma compared with operative treatment. *J Bone Joint Surg Am*. 1998 Jun;80(6):815–21.
- 15 Lanza E, Thouvenin Y, Viala P, Sconfienza LM, Poretti D, Cornalba G, et al. Osteoid osteoma treated by percutaneous thermal ablation: when do we fail? A systematic review and guidelines for future reporting. *Cardiovasc Intervent Radiol*. 2014 Dec;37(6):1530–9.
- 16 Karagöz E, Özel D, Özkan F, Özel BD, Özer Ö, Coşkun ZÜ. Effectiveness of computed tomography guided percutaneous radiofrequency ablation therapy for osteoid osteoma: initial results and review of the literature. *Pol J Radiol*. 2016 Jun;81:295–300.
- 17 Kuyumcu G, Mason EG, Ilaslan H. Air-blocking ablation of osteoid osteoma; a technical note. *Skeletal Radiol*. 2017 Jul;46(7):957–60.

- 18 Saccomanni B. Osteoid osteoma and osteoblastoma of the spine: a review of the literature. *Curr Rev Musculoskelet Med*. 2009 Mar;2(1):65–7.
- 19 Avanzi O, Joilda FG, Donato JP, Dezen EL, Pinto WC. Tumoures benignos e lesões pseudotumorais da coluna vertebral. Análise de 60 pacientes. *Rev Bras Ortop*. 1996;31(2):131–42.
- 20 Ozaki T, Liljenqvist U, Hillmann A, Halm H, Lindner N, Gosheger G, et al. Osteoid osteoma and osteoblastoma of the spine: experiences with 22 patients. *Clin Orthop Relat Res*. 2002 Apr;397:394–402.
- 21 Chew FS, Pena CS, Keel SB. Cervical spine osteoblastoma. *AJR Am J Roentgenol*. 1998 Nov;171(5):1244.
- 22 McLeod RA, Dahlin DC, Beabout JW. The spectrum of osteoblastoma. *AJR Am J Roentgenol*. 1976 Feb;126(2):321–5.
- 23 De Souza Dias L, Frost HM. Osteoblastoma of the spine. A review and report of eight new cases. *Clin Orthop Relat Res*. 1973 Mar-Apr;(91):141–51.
- 24 Jaffe HL, Mayer L. An osteoblastic osteoid tissue-forming tumour of a metacarpal bone. *Arch Surg*. 1932;24(4):550–64.
- 25 Lassalle L, Campagna R, Corcos G, Babinet A, Larousserie F, Stephanazzi J, et al. Therapeutic outcome of CT-guided radiofrequency ablation in patients with osteoid osteoma. *Skeletal Radiol*. 2017 Jul;46(7):949–56.
- 26 Cappuccio M, De Iure F, Amendola L, Corghi A, Gasbarrini A. Cervical osteoid osteoma progression to osteoblastoma. *Spine J*. 2014 Jun;14(6):1070–1.
- 27 Houang B, Grenier N, Gréselle JF, Vital JM, Douws C, Broussin J, et al. Osteoid osteoma of the cervical spine. Misleading MR features about a case involving the uncinate process. *Neuroradiology*. 1990;31(6):549–51.
- 28 Jackson RP, Reckling FW, Mants FA. Osteoid osteoma and osteoblastoma. Similar histologic lesions with different natural histories. *Clin Orthop Relat Res*. 1977 Oct;(128):303–13.
- 29 Kaweblum M, Lehman WB, Bash J, Grant AD, Strongwater A. Diagnosis of osteoid osteoma in the child. *Orthop Rev*. 1993 Dec;22(12):1305–13.
- 30 Kong J, Xiao H, Liu T, Yan W, Qian M, Song DW, et al. The valuation of using FDG PET-CT in detecting osteoid osteoma of the cervical spine. *J Spinal Disord Tech*. 2015 Mar;28(2):E67–73.
- 31 Keim HA, Reina EG. Osteoid-osteoma as a cause of scoliosis. *J Bone Joint Surg Am*. 1975 Mar;57(2):159–63.
- 32 Weinstein JN, McLain RF. Primary tumors of the spine. *Spine*. 1987 Nov;12(9):843–51.
- 33 Dubousset J. Scoliose raide et douloureuse chez l'enfant : attention! *Rev Pédiatr*. 1970;6:597–606.
- 34 Bruneau M, Polivka M, Cornelius JF, George B. Progression of an osteoid osteoma to an osteoblastoma. Case report. *J Neurosurg Spine*. 2005 Sep;3(3):238–41.
- 35 Wang B, Han SB, Jiang L, Yuan HS, Liu C, Zhu B, et al. Percutaneous radiofrequency ablation for spinal osteoid osteoma and osteoblastoma. *Eur Spine J*. 2017 Jul;26(7):1884–92.

Fig. 1. Cervical X-ray with segment inversion of the cervical lordosis.

Fig. 2. Preoperative T2-weighted MRI showing a focal hypersignal in the left vertebral body of C7 and minimal soft tissue involvement.

Fig. 3. Bone scan with a focus of intense osteoblastic hyperactivity in the left lamina of C7.

Fig. 4. CT scan showing a dense lesion of the left lamina of C7, surrounded by a lytic halo.

Fig. 5. SPECT-CT of the lesion.

Fig. 6. Postoperative C6-T1 arthrodesis.

Fig. 7. Central nidus composed of bony trabeculae, surrounded by a zone of sclerotic bone. The nidus and surrounding bone are sharply demarcated from each other, confirming the diagnosis.