Osteoid osteoma of the dens axis

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Abstract An osteoid osteoma of the dens axis was diagnosed by computed tomography and bone scintigraphy in a 14-year-old girl with a 1.5-year history of pain. No case of an osteoid osteoma of the dens axis has been published in the literature yet. Regarding its clinical and radiological appearance, this osteoid osteoma was essentially similar to osteoid osteomas in other locations, i.e., it had an osteolytic nidus surrounded by sclerosis, associated with nocturnal pain and a positive aspirin test.

Keywords Osteoid osteoma · Axis · Cervical · Spine

Introduction

The osteoid osteoma, first described by Bergstrand in 1930, was defined by Jaffe in 1935 as a primary benign osteoblastic tumor [15]. According to the WHO guidelines, it consists of an osteolytic defect with sharp margins, measuring 0.5–2 cm in size. The nidus is usually less than 1 cm in diameter and, depending on its location, is surrounded by a marginal sclerosis of varying intensity.

Histologically, the nidus consists of an irregular network of osteoid trabeculae with a varying degree of calcification and a highly vascularized stroma rich in cells.

Cortical, cancellous and sub-periosteal osteoid osteomas are classified by the location of the nidus in the bone. The cortical (classical) type is the most common, while the

cancellous and sub-periosteal types are less frequent. The intra-articular type is a special one; when located in the femoral neck or the bones of the foot, it may mimic inflammatory processes because of arthralgia or joint effusion.

The incidence of the osteoid osteoma is relatively high. It accounts for approximately 10.8–13.5% of all primary benign bone tumors [5] and approximately 3% of all primary bone tumors [5]. It occurs two to three times more frequently in men than in women [5].

Predilection rates of about 50% in the second decade and 80% between the age of 5 and 25 years have been registered [5]. The tumor is very uncommon after the age of 40 years or during infancy.

Predilection sites of the osteoid osteoma are the long bones, especially those of the lower extremity. The primary locations are in the femur and tibia, accounting for >50% of all tumors, followed by the bones of the hand and foot, the humerus and the vertebral column.

The purpose of the diagnostic investigation is to detect the nidus. It is important to locate the nidus, if necessary by employing the most modern methods, in order to avoid inappropriate therapy. In addition to scintigraphy, which usually visualizes the richly vascularized nidus as a “hot spot,” conventional tomography and angiography used to be important diagnostic methods in the past. Both procedures have receded to the background, being now replaced by modern cross-section imaging procedures such as CT and MRT.

Osteoid osteomas rarely occur in the vertebral column. The spine is only affected in about 10% of the cases, with more than 50% involving the lumbar spine, here again, mainly (90%) the dorsal parts up to the origin of the pedicle; the body of the vertebra is involved in exceptional cases [7, 14, 16, 17, 20].

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With regard to the cervical spine, an osteoid osteoma of the third cervical vertebra [21], the arc of the axis [8], the second vertebral body [18], the transverse process of the axis [2] and the transverse process of the fourth cervical vertebra [19] have been reported in literature.

We present a recently encountered, instructive case of an osteoid osteoma of the dens axis confirmed by computed tomography, MRT, bone scintigraphy and a positive aspirin test.

Case report (G. P. *1988, female, schoolgirl)

A 14-year-old girl had a 1.5-year history of neck pain, occasionally with radiating pain to the head combined with tension in the neck and occasional tenderness to touch. The patient reported constant nocturnal pain.

Computed tomography and magnetic resonance tomography studies led the radiologists to make the following differential diagnosis: persistent dens apophysis, possibly close to the base, an old dens fracture or a congenital morphological variant.

At our first investigation, we found a mildly enhanced cervical lordosis and a markedly atrophic, visibly tensed trapezoid muscle strain. Limited extension, limited lateral inclination to the right and a markedly reduced rotation to the left were recorded.

After reviewing the existing CT (Fig. 1), MRT (Fig. 2) and plain radiographs (Fig. 3) in combination with the

Fig. 2 Corresponding T2 weighted MRI scan

patient's medical history and clinical findings, we established the diagnosis of an osteoid osteoma of the dens axis. Subsequently, the patient was subjected to a two-phase bone scintigraphy, which confirmed the suspected diagnosis.

After the diagnosis had been made, the patient was given a pain diary with a visual analog scale. The routinely performed acetylsalicylic test was positive. Based on the above-mentioned gastrointestinal symptoms, and on account of the risky anatomic location of the lesion and the high morbidity associated with it, we decided to implement the following treatment based on the publication of Bottnar et al. [3]: the patient was given 200 mg celecoxib orally

Fig. 1 **a** Transversal CT images showing the nidus and perifocal sclerosis. **b** Sagittal CT images of the osteoid osteoma at the dens axis

Fig. 3 Plain X-ray depicting the nidus in an enoral technique

once daily at 10:00 p.m. This treatment led to a satisfactory reduction of pain (mean VAS score 0.5).

By applying this conservative therapeutic scheme for 2 years, the patient became free of symptoms and the CT scan performed after this period of time revealed a disappeared sclerosed nidus (Fig. 4). After this final examination, the antiphlogistic medication was canceled and the patient was free of symptoms at the time of publication.

Discussion

Osteoid osteoma is a rare tumor, accounting for approximately 10% of all benign tumors [5]. The classical cardinal symptom is pain, which reaches a maximum at night. Despite a typical medical history, the diagnosis was delayed in the present case as well. In patients with ambiguous pain, in whom the differential diagnosis includes osteoid osteoma, a computed tomography in combination with a bone scintigram has proved its value for the purpose of morphological identification. The nidus with perifocal marginal sclerosis can be optimally visualized on CT, as well as on MRT following the administration of gadolinium [22]. Bone scintigraphy, which was performed additionally, is a suitable and sensitive screening test. In conjunction with X-ray, it is sufficiently indicative of the diagnosis. In the present case, osteoid osteoma could not be clearly diagnosed on conventional radiographs (Fig. 3). A non-displaced old dens fracture and a persisting apophysis were included in the differential diagnosis. The definitive diagnosis was made after considering the clinical findings together with imaging procedures such as plain films, CT and bone scintigraphy. The location of an osteoid osteoma in the dens axis, as described here, is rare. However, other similar locations such as the corpus and the lamina of the axis have been described. In all of these cases the lesion was resected by surgery.

Minimal invasive surgery techniques like percutaneous laser or radiofrequency induced ablation [1, 4, 6, 9–13] have been well documented in literature. At our department, percutaneous CT-guided radiofrequency induced ablation is performed routinely since 1998 with successful results similar to the internationally published papers.

In the present case, surgery was not applied because of the risky anatomic location of the lesion and the high

Fig. 4 Final CT scan identifying the disappearance and complete sclerosis of the nidus

morbidity associated with it. The chosen regimen (“Wait and See” [23]) seems to be absolutely successful in this special case, as the patient is pain free and the nidus itself has disappeared at the last followup, 2 years after establishing the diagnosis.

References

1. Albissini U, Rimondi E, Malaguti MC, Ciminari R, Bianchi G, Mercuri M (2005) Radiofrequency thermal ablation of non-spinal osteoid osteoma: remarks on method. *Radiol Med (Torino)* 109(5–6):555–562
2. Biswas D, Mal RK (2006) Dysphagia secondary to osteoid osteoma of the transverse process of the second cervical vertebra. *Dysphagia*, Nov (epub ahead of print)
3. Bottnar F, Roedel R, Wortler K, Grethen C (2001) Cyclooxygenase 2 inhibitor for pain management in osteoid osteoma. *Clinical Orthop* 393:258–263
4. Cantwell CP, Obyrne J, Eustace S (2004) Current trends in treatment of osteoid osteoma with an emphasis on radiofrequency ablation. *Eur Radiol* 14(4):607–617
5. Dahlin DC (1978) In: *Bone tumours: general aspects and data on 6221 cases*, 3rd edn. Thomas Springfield, IL
6. DeFriend DE, Smith SP, Hughes PM (2003) Percutaneous laser photocoagulation of osteoid osteomas under CT guidance. *Clin Radiol* 58(3):222–226
7. Eysel B (1994) Diagnostic and therapy of the osteoid-osteoma of the lamina—review of the literature and own results. *Z Orthop* 132(5):357–362
8. Eysel P, Rompe JD (1994) Osteoid osteoma of the axis. *Eur Spine J* 3(4):231–232
9. Gangi A, Dietemann JL, Gasser B, Guth S, de Unamuno S, Fogarassi E, Fuchs C, Siffert P, Roy C (1998) Interventional radiology with laser in bone and joint. *Radiol Clin North Am* 36(3):547–557
10. Gangi A, Alizadeh H, Wong L, Buy X, Dietemann JL, Roy C (2007) Osteoid osteoma: percutaneous laser ablation and follow up in 114 patients. *Radiology* 242(1):293–301
11. Gebauer B, Tunn PU, Gaffke G, Melcher I, Felix R, Stroszczyński C (2006) Osteoid osteoma: experience with laser and radiofrequency induced ablation. *Cardiovasc Intervent Radiol* 29(2):210–215
12. Ghanem I (2006) The management of osteoid osteomas: updates and controversies. *Curr Opin Pediatr* 18(1):36–41
13. Hadjipavlou AG, Lander PH, Marchesi D, Katonis PG, Gaitanis IN (2003) Minimally invasive surgery for ablation of osteoid osteoma of the spine. *Spine* 28(22):472–477
14. Jackson RP, Reckling FW, Mants FA (1977) Osteoid osteoma and osteoblastoma. Similar histologic lesions with different natural histories. *Clin Orthop* 128:303–313
15. Jaffe H (1935) Osteoid osteoma: a benign osteoblastic tumor composed of osteoid and atypical bone. *Arch Surg*, pp 709–715
16. Mau H (1982) Osteoid osteoma of the vertebral column. *Z Orthop* 120(6):761–766
17. McLellan DI, Wilson FC Jr (1967) Osteoid Osteoma of the spine. A review of the literature and report of six new cases. *JBJS (A)*, pp 111–121
18. Molloy S, Saifuddin A, Allibone J, Taylor B (2002) Excision of an osteoid osteoma from the body of the axis through an anterior approach. *Eur Spine J* 11(6):599–601
19. Muthukumar N (2002) Osteoid osteoma—a rare cause of brachial neuralgia. *Childs Nerv Syst* 18(3–4):161–163
20. Poulsen JO (1969) Osteoid osteoma. *Arch Orthop Scand* 40(2):198–204
21. Suttner NJ (2002) Osteoid osteomas of the body of the cervical spine. Case report and review of the literature. *Br J Neurosurg* 16(1):69–71
22. Wachtel SW, Exner GU, von Hochstetter A, Sennwald G (1995) Osteoid osteoma of the hand. Case presentation with special reference to magnetic resonance tomography and literature review. *Z Orthop* 133(1):76–78
23. Winkelmann S, Hirsch W, Burdach S, Horneff G (2003) Therapy of osteoid osteomas—always surgically? *Klin Padiatr* 215(1):35–39