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Osteoid osteoma with a multicentric nidus: a case report and review of the literature

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Abstract To date, 23 cases with osteoid osteoma (OO) including multiple nidi in single bone have been reported in the world literature. A case report of an 18-year-old boy with an OO on his left femoral neck, which contained double nidi is presented. Plain radiography, computed tomography (CT) and magnetic resonance imaging (MRI) of the proximal femur showed OO with a multicentric nidus. Bone scintigraphy demonstrated increased activity in the left femoral neck region. The tumor was removed with curettage and shaving using lateral approach. The patient was asymptomatic for 5 years after surgery.

Keywords Osteoid osteoma · Multicentric nidus · Femur

Introduction

Osteoid osteoma (OO) is a comparatively common tumor comprising approximately 11% of all benign bone neoplasms [1]. The essential part of an OO is the nidus, consisting of highly vascularized osteoid tissue surrounded by sclerotic bone and generally less than 1 cm (up to 2 cm) in diameter [13, 24]. It occurs mainly in children and young adults.

Monofocal OOs have been described in various parts of the skeleton; however, most cases occur in the long bones, particularly the femur and tibia. Usually an OO is monostatic and unilocular, i.e. has a single nidus. In only rare instances, OO may be multicentric [12, 13]. By definition, "multicentric" OO is a lesion with 2 or more nidi [24]. To date, 23 cases with the multicentric OO in single bone have been described by several authors [3, 7, 10–13, 16, 17, 19, 23–25, 32, 33, 35, 37].

In this report, we present a case of multicentric OO with 2 nidi in the femoral neck.

Case report

An 18-year-old boy was admitted to our clinic with the complaint of pain in the left hip for 2 years. The pain was especially severe at night and was relieved by salicylate. Physical examination and laboratory studies revealed no abnormalities. Pelvis and left femur antero-posterior radiographs revealed two contiguous radiolucencies with peripheral reactive sclerosis within the left femoral neck (Fig. 1). Preoperative localization of the double nidi was precised by CT and MRI. An axial CT scans revealed two contiguous hypodense nidi with prominent peripheral sclerosis within the left femoral neck (Fig. 2). The diameter of the lesions were 0.7 and 0.9 cm in CT. Coronal T_1 weighted MRI showed two contiguous hypointense lesions with central hyperintense focuses (Fig. 3). Bone scintigraphy demonstrated increased activity in the left femoral neck region correlated with the location of the nidi radiographically (Fig. 4).

In the operation, double nidi was removed with curettage and shaving from trochanteric site of femur, using lateral approach under image intensification. Bone grafting was not performed after curettage. Postoperative radiography confirmed the complete excision of the tumor. Culture was

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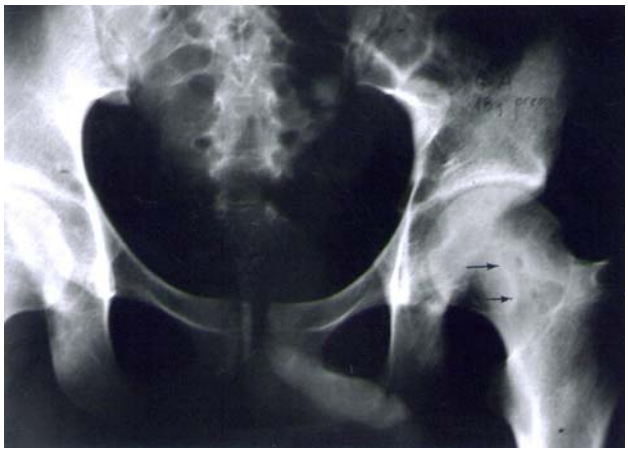

Fig. 1 The anteroposterior radiograph of pelvis (hip) showing the presence of two discrete radiolucent foci of osteoid osteoma in left femoral neck

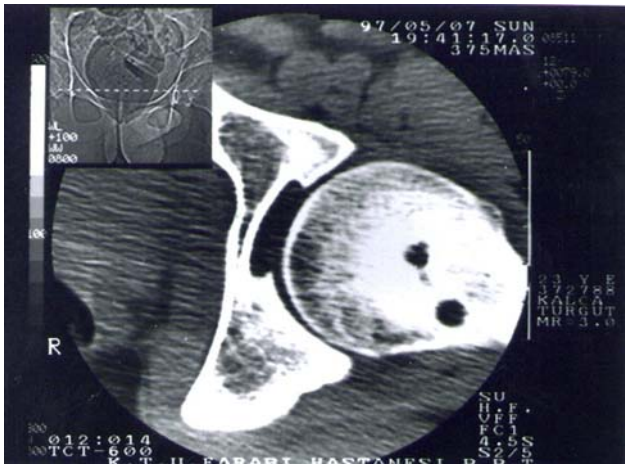

Fig. 2 Computer tomography showing the intramedullar localization of the osteoid osteoma with double nidus

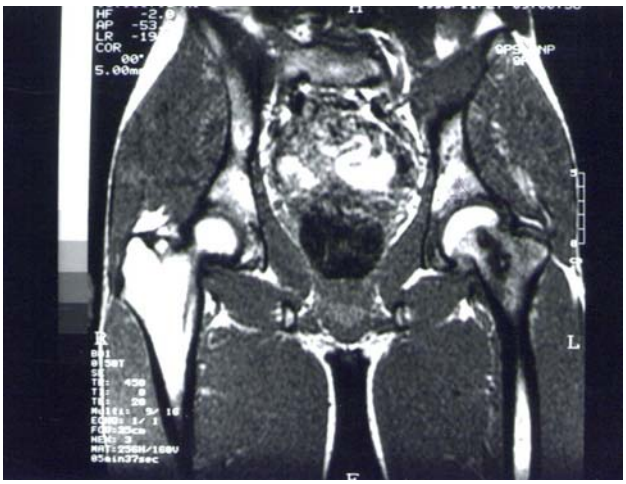

Fig. 3 The double nidus are clearly seen on magnetic rezonans imaging

Fig. 4 Bone scintigraphy showing increased activity in the left femoral neck region

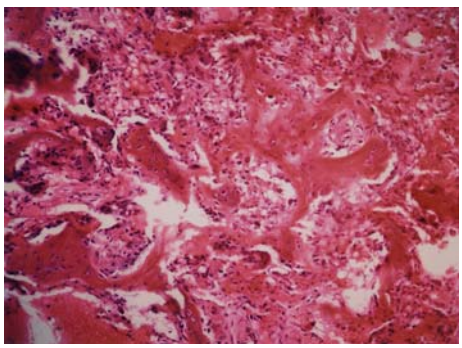

Fig. 5 This histologic section demonstrates bone trabeculae formed by cells with osteoblastic differentiation in a fibrovascular background (HE × 100) (colour and black–white)

negative. Histologic examination demonstrated the existence of osteoid sheets, strongly vascularized, and rich in osteoblasts, thus confirming the diagnosis of OO (Fig. 5).

Typical roentgenograms, the clinical picture, histologic section and resolution of symptoms after surgery supported our diagnosis. During follow-up examination for 5 years, the patient was free of pain and had a full range of hip motion.

Discussion

The pathology of OO was first described by Bergstrand [4]. A few years later Jaffe [14] established this benign neoplasm as a clinical entity and named it OO. Osteoid osteomas are benign lesions of bone that usually present in the early decades of life with characteristic pain and radiographic appearance. The pain is typically intense at night and relieves with salicylate or non-steroidal anti-inflammatory drugs [25]. Half of the patients respond well to salicylate as seen in our case [34]. Most patients eventually require surgical treatment for complete relief of pain. The

classic approach to this lesion is direct surgical excision or curettage. In recent years, several CT-guided percutaneous techniques have been used in order to achieve removal or destruction of the nidus [21]. A new treatment called CT-guided percutaneous radiofrequency ablation, first described in 1995 [31], has been introduced [6, 30, 39]. The reported results seem to be very good and free of complications as minimally invasive treatment for OO. There are few case reports treated non-operatively and resolution of pain in cases with OO [18]; so, we surgically treated our patient and in early postoperative period the pain was relieved.

The radiographic appearance is a radiolucent nidus surrounded by reactive bone [25]. CT is the most useful preoperative study in localizing the nidus. Localization of the lesion is done using plain radiography, CT, bone scans and/or MRI. In our case, plain radiographs, CT, MRI and bone scan were used for precise localization and characterization.

Multicentric OO is a very rare variant. So far, only 32 cases with OO including multiple nidi have been reported in both single and multiple bones in the world literature. Multifocal nidi were found either close to each other in the single bone in 23 cases [3, 7, 10–13, 16, 17, 19, 23–25, 32, 33, 35, 37], in two adjacent bones in 6 cases [2, 15, 20, 27, 29], or in two widely seperated bones in 3 cases [5, 28]. Multiple nidi may be found in close proximity in a single bone or neighboring bones, or as widely seperated lesions in the same bone or different bones. The sites of the 23 cases with multiple-nidi in the same bone are listed in Table 1. The femur is a common site (34.8%) for the multicentric forms. The number of the nidi within the lesion varied from 2 to more. The present case was localized to the femur and had 2 radiolucent nidi.

Osteoid osteomas are traditionally classified into cortical, medullary or cancellous, and subperiosteal or periosteal. OO is most commonly located within the cortex [16]. In cortical localizations, the sclerosis is very dense, occasionally masking the central nidus. In medullary locations, the osteoma should appear as an opaque or a radiolucent zone without perifocal sclerosis [3]. An intramedullary nidus is less easily identified on plain radiography because very little or no reactive sclerosis is present. In our patient, the nidi of OO were located in spongious bone and had sclerotic reaction.

Proper pre- and intraoperative localizations of the tumor are critical to ensure an adequate resection and minimize the chance of recurrence. There is general agreement in the literature that complete excision is the treatment of choice and that incomplete removal of the nidus leads to recurrence of symptoms. Once the nidus is removed, the patient is relieved of pain immediately and may notice this in postoperative period. It has been suggested that local recurrence

Table 1 Cases of multicentric OO in the same bone from the literature according to years

Case No	Age	Sex	Bone affected	Foci	Reference
1	3	M	Tibia	3	[35]
2	16.5	M	Femur	2	[19]
3	5	F	Femur	2	[11]
4	26	M	Femur	Several	[23]
5	12	F	Lumbar Vertebra (L_2)	Several	[23]
6	–	–	Ethmoid bone	Several	[33]
7	20	F	Pubic bone	Several	[33]
8	–	–	Sacrum	Several	[33]
9	–	–	Humerus	3	[33]
10	12	M	Femur	2	[10]
11	17	M	Humerus	3	[10]
12	12	M	Tibia	3	[13]
13	27	F	Tibia	Several	[37]
14	22	F	Dorsal Vertebra (D_7)	2	[7]
15	18	M	Lumbar Vertebra (L_5)	2	[17]
16	10	M	Finger	2	[3]
17	28	M	Scafoid	2	[24]
18	2.5	M	Femur	2	[25]
19	5	M	Femur	3	[16]
20	16	M	Tibia	2	[12]
21	24	M	Humerus	2	[32]
22	10	F	Femur	3	[8]
23	19	M	Femur	2	[40]
24	18	M	Femur	2	The present case

may sometimes be due to the development of a second lesion, however, it is generally assumed to be the result of incomplete removal of the original lesion [12]. Recurrences are so rare, however, that they may be the result of partial excision of multicentric osteoid osteomas [10, 12]. In our patient, the tumor was precisely localized preoperatively, completely excised, and then checked under scopy and by direct X-ray for possible tumor remnants. For that reasons, we did not observe any recurrence during follow-up period.

Osteoid osteoma is generally diagnosed by means of clinical findings and radiological appearances [21]. The practice depends on the fact that certain diagnoses are so radiographically typical, such as OO, that excision is indicated without a biopsy [9, 22]. Several osteoid osteomas, not verified histologically, have been reported in the literature [5, 9, 18, 19, 32, 38]. In recent years, many patients were diagnosed as OO treated with CT-guided percutaneous radiofrequency ablation technique without histological verification [21, 30, 31, 39]. Curettage of an OO may pose some problems regarding the histologic diagnosis [34]. Bloc resection is therefore preferable to curettage when technically feasible [12]. In our case, lesion was located intramedullary in femoral neck, so we could not perform en

bloc resection. In histological examination of the bones removed with curettage and shaving, we found that the nidus and histologic sections supported our radiologic preceding diagnosis. Our reason for not using CT-guided radiofrequency ablation was the size of lesion. This technique is not applicable for large tumors [39].

In summary, an extremely rare case of multicentric intramedullar OO and review of the literature were presented. Our case indicates that more than one OO can rarely occur in a single bone. Accurate evaluation of the radiographic findings is very important. The definitive diagnosis of a double-nidus OO was suggested after careful analysis of the initial radiographs demonstrated the existence of two distinct intramedullar nidi. Careful preoperative CT studies should help in the detection of possible companion lesions. We think that satisfactory result could be achieved with curettage in the treatment of intramedullary located OO. In recurrent lesions, it must always be kept in mind that the lesion could not be removed completely or there might be multiple nidi.

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