ORIGINAL PAPER
PŮVODNÍ PRÁCE

Management of Osteoid Osteoma in Unusual Locations

Léčení osteoid osteomu v neobvyklých lokalizacích

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ABSTRACT

PURPOSE OF THE STUDY

Osteoid osteoma is a benign tumor that forms in bone, which accounts for 3% of all primary bone tumors. The classical clinical finding is substantial nocturnal pain and imaging findings. The management of osteoid osteomas include open surgical excision or minimally invasive percutaneous interventions. Why and which treatment modality should be considered between CT-guided radiofrequency ablation and open surgical excision for osteoid osteomas in unusual locations?

MATERIAL AND METHODS

We retrospectively reviewed 17 patients with osteoid osteomas in unusual locations included cuboid, triquetrum, coronoid process, and proximal phalanx. We evaluated the duration from symptoms to diagnosis, activity related pain, clinical findings, and possible recurrence or complications. The minimum clinical follow-up was 51 ± 34.8 months.

RESULTS

CT-guided radiofrequency ablation was applied to 3 patients and open surgical excision procedures to 14. All the complaints of patients gone after treatment. No major complications were observed following CT-guided radiofrequency ablation or surgical excision. Transient weakness/paresthesia was determined in 1 patient in the treated shoulder after CT-guided radiofrequency ablation, which resolved spontaneously in the 6th week. There was only recurrence seen in 1 patient, who had 2nd proximal phalangeal osteoid osteoma. Proximal interphalangeal joint arthrodesis was performed after recurred lesion.

DISCUSSION

The main challenge in management of the osteoid osteomas of the unusual locations are the diagnosis. When we examined the literature, the interval from the beginning of the symptoms to accurate diagnosis did not change over the past decades. Techniques for management of these lesions should be chosen with consideration of the location of the lesion.

CONCLUSIONS

If there is long-term complaint of undiagnosed limb pain, the physician should suspect osteoid osteoma. However, the selection of treatment modality should be considered according to the location of the lesion. Which management modality is superior may change depending on the location of the lesion between CT-guided radiofrequency ablation and surgical excision.

Key words: osteoid osteoma, unusual locations, CT guided, radiofrequency ablation, benign bone tumor.

INTRODUCTION

Osteoid osteoma (OO) is a benign tumor that forms in bone, which accounts for 3% of all primary bone tumors and mostly affects male patients in the first three decades of life (25). OO has a central "nidus" surrounded by dense sclerotic bone tissue, usually < 20 mm in size (15). The classical clinical finding is substantial nocturnal pain, which generally persists until the lesion diagnosed (20). Typical radiographic findings of OO include a nidus, which may display a centrally calcified geographic lesion, accompanied by cortical thickening and reactive sclerotic rim in a long bone diaphyseal cortex (Fig. 1). Most OOs develop in long bones of the limbs (especially metaphyseal and diaphyseal regions of the femur and tibia), and these comprise approximately 50% of cases, but lesions can be found in any bone (2, 7). Among other localizations, OOs occur in upper limbs in 13% to 31% of cases, in vertebrae in approximately 10% of cases and in intra-articular locations in 12% of cases (7, 10, 17). OOs in other locations have a low prevalence.

The management of OOs include open surgical excision or percutaneous interventions (3, 17, 23). In addition to having various advantages over each other, there are also studies in which both are referred to as the gold standard treatment (4, 9). The location of OOs may affect the choice of treatment. The hypothesis of this study was that the selection of treatment modality should be considered according to the location of the lesion. The

Fig. 1. Axial and coronal CT images of a 17-year-old female (patient-3) showing an ovoid medullary-based radiolucent lesion (white arrow) of the proximal fibula measuring 14×9 mm in diameter (A-B). Axial and coronal MRI images showed that the lesion has central calcifications and is associated with moderate peripheral osteosclerosis and marked periosteal reaction (black arrow) (C-D). Pre-operative (E) and post-operative (F) anteroposterior radiographs of proximal fibular OO (the lesion inside the circle).

aim of this study was to present the results of CT-guided radiofrequency ablation (RFA) and open surgical excision for OOs in unusual locations.

MATERIAL AND METHODS

Following institutional review board approval, a search was made of an orthopaedic department database. A total of 102 patients referred for management of OO between 2009 and 2017 were retrospectively reviewed and 17 (16.7%) were identified as OO with unusual location (Table 1). Medical records were reviewed to confirm study inclusion on the basis of the following criteria: a diagnosis of OO involving other than usual localizations. The exclusion criteria were OO in usual locations defined as proximal femur and tibia, and distal femur, and incomplete data in the radiographic or clinical/pathology records. Demographic and clinical characteristics of patients were given in table 2.

Radiographs and CT scans of the lesions were obtained for all patients, and MRI was obtained for 9 patients. Radiographs were examined for coexistence of cortical thickening, central sclerosis, and a radiolucent nidus with the OO. MRI was examined for the coexistence of perilesional bone marrow edema, diffuse synovitis within the adjacent joint, a joint effusion, and a nidus with the OO. CT examinations were examined for the coexistence of periostitis and a radiolucent nidus with the OO, and for the exact location of the lesion (cortical, medullary, subperiosteal; intra-/extra-articular).

The patient clinical files were examined for coexistence of nocturnal pain, pain with activity and relief

Table 1. Clinical and radiological characteristics of OOs for each patient

Patient Number	Age (years old)	Side	Location	Site	Nidus diameter (mm)	Intraarticular nidus / Joint effusion	Central sclerosis	Follow-up (months)	Treatment	Night pain	Relief with NSAIDs
Lower extremity											
1	22	R	acetabulum	cortical	7	- / +	-	26	RFA	+	+
2	19	L	acetabulum	medullary	9	- / +	-	27	RFA	+	+
3 (Fig.1)	17	L	proximal fibula	medullary	9	- / -	+	43	excision	+	+
4	26	L	diaphysis of fibula	cortical	4	- / -	-	36	excision	+	NA
5	24	L	talar neck	cortical	6	+ / +	-	112	excision	+	-
6	17	R	cuboid	cortical	5	- / -	+	120	excision	-	+
7	16	R	3rd distal phalanx	cortical	4	- / -	+	26	excision	+	NA
Upper extremity											
8	24	R	proximal humerus	cortical	6	- / -	+	112	excision	+	+
9 (Fig.2)	15	R	proximal humerus	cortical	10	- / -	+	40	RFA	+	+
10 (Fig.3)	26	L	coronoid process	cortical	5	+ / +	+	28	excision	+	-
11	13	R	diaphysis of radius	cortical	7	- / -	+	31	excision	+	+
12	26	L	distal radius	subperiosteal	6	- / +	-	31	excision	-	-
13	27	L	triquetrum	cortical	4	+ / +	-	32	excision	+	+
14	22	R	2nd proximal phalanx	cortical	4	+ / +	-	38	excision +PIP arthrodesis	+	+
Spine											
15	10		C5	cortical	5	- / -	-	99	excision	+	+
16	23		C6	cortical	9	- / -	-	43	excision	+	+
17	9		L1	cortical	10	- / -	+	24	excision + posterior instrumentation	+	+

Table 2. Demographic and clinical characteristics of patients (n = 17).

Age at diagnosis (years old)*	19.8 ± 5.8
Male / Female ratio	3.25:1 (13 / 4)
Lesion location	
Lower extremity	7 (41 %)
Upper extremity	7 (41 %)
Spine	3 (18 %)
Lesion site	
Cortical	14 (82 %)
Medullary	2 (12 %)
Subperiosteal	1 (6 %)
Nocturnal pain	11 (64.7 %)
Activity related pain	9 (53 %)
Relief with NSAIDs †	7 (41.2 %)
Duration from symptoms to diagnosis (months) ‡	7 (3 - 24)
Delayed diagnosis (>6 months)	10 (59 %)
Average follow-up*	51 ± 34.8

*(*Data are presented as the mean and the standard deviation. †Data were only available for 15 patients. ‡Data are presented as the median, with the range in parentheses)*

with NSAIDs, duration from symptoms to diagnosis, recurrence, primary presenting complaint, and the reason for delayed diagnosis.

As mentioned in previous studies (2, 17), biopsy was not applied to the patients in this study and diagnosis was made clinically and radiologically. However, some authors have recommended pre-intervention biopsy (21, 29).

All RFA procedures were performed with the patient under general anesthesia in the CT room under aseptic conditions. An experienced interventional radiologist and orthopaedic surgeon (SAG) performed all CT-guided RFAs with an anesthesiologist present during the entire operation (Fig. 2). The surgeries were performed by two orthopaedic surgeons (HBC, SAG).

Patients were informed about the treatment options which were open surgery and RFA. The follow-up was only clinical, with a minimum length of 24 months (mean, 51 months; SD ± 34.8). All patients were invited for final clinical evaluation. No patients were lost to follow-up. Informed consent was obtained from all patients.

RESULTS

All patients returned to normal activity within one week after RFA and 2 weeks after surgery, and had complete pain relief. There was only one recurrence seen in

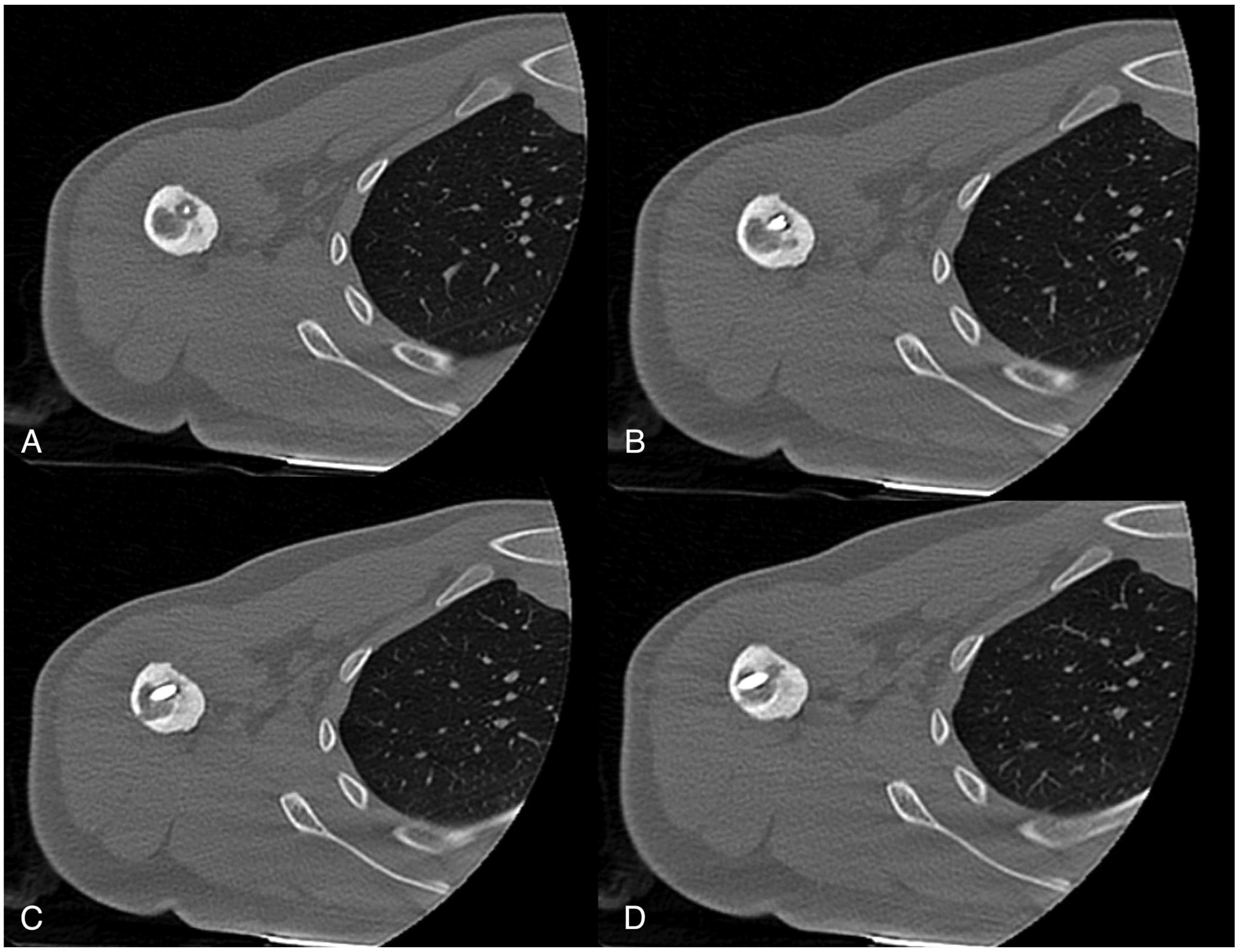

Fig. 2. RFA for a 10 mm diameter OO in the proximal humerus region in a 15-year-old male is shown (patient-9). The direct lateral approach was used. CT scans show (A) the cortical nidus with surrounding sclerosis, (B, C, D) the radiofrequency needle placed in the nidus can be seen.

patient No. 14, who had 2nd proximal phalangeal OO. The localization of the OO was intra-articular and the PIP joint was deformed. Therefore, PIP arthrodesis was recommended and this operation was performed with the patient's informed consent. No fractures, neurovascular injury, or other late complications were observed in any patient.

In all patients, the primary presenting complaint was pain. Approximately 48% of the patients experienced radiating pain and they were not able to localize the painful spot. Nocturnal pain (64.7%) and pain relief with NSAIDs (41.2%) were common.

On radiographic examination, the radiolucent nidus was able to be seen in only 29%, and central sclerosis and/or cortical thickening could be seen on the radiographs of 41% of the patients. The precise diagnostic tool was CT, which showed a radiolucent nidus in all patients. Of 9 patients applied with MRI, synovitis was observed in 3 (33%) and effusion in 5 (56%). All the MRI scans showed focal perilesional osseous edema.

Initial mis-diagnoses were identified in 59% (10/17) of cases, as minor trauma (40%, 3/10), synovitis (20%, 2/10), growing pains (20%, 2/10), stress fracture (20%, 2/10), and infection (10%, 1/10). None of the patients underwent any operative procedures for these mis-diagnoses, but 70% (7/10) had medical treatment and/or bracing. In each patient, the pain persisted until the OO was treated. Delayed diagnosis (>6 months) was seen in 59% (10/17) of patients. The main reasons for delayed diagnosis were insufficient imaging examinations and exaggerated MRI findings. Scintigraphy had been applied to 3 patients before referral to our hospital.

RFA was applied to 3 patients (18%) and open surgical excision procedures to 14 (82%). The average clinical follow-up was 51 months (range, 24–120 months). All patients had available follow-up information.

No major complications were observed following RFA or surgery. Transient weakness/paresthesia was determined in patient No. 9 in the treated shoulder after RFA, which resolved spontaneously in the 6th week.

DISCUSSION

When an osteoid osteoma is located in the limbs, it is predominantly seen in the proximal femur, but is also common in the distal femur, and proximal tibia, where

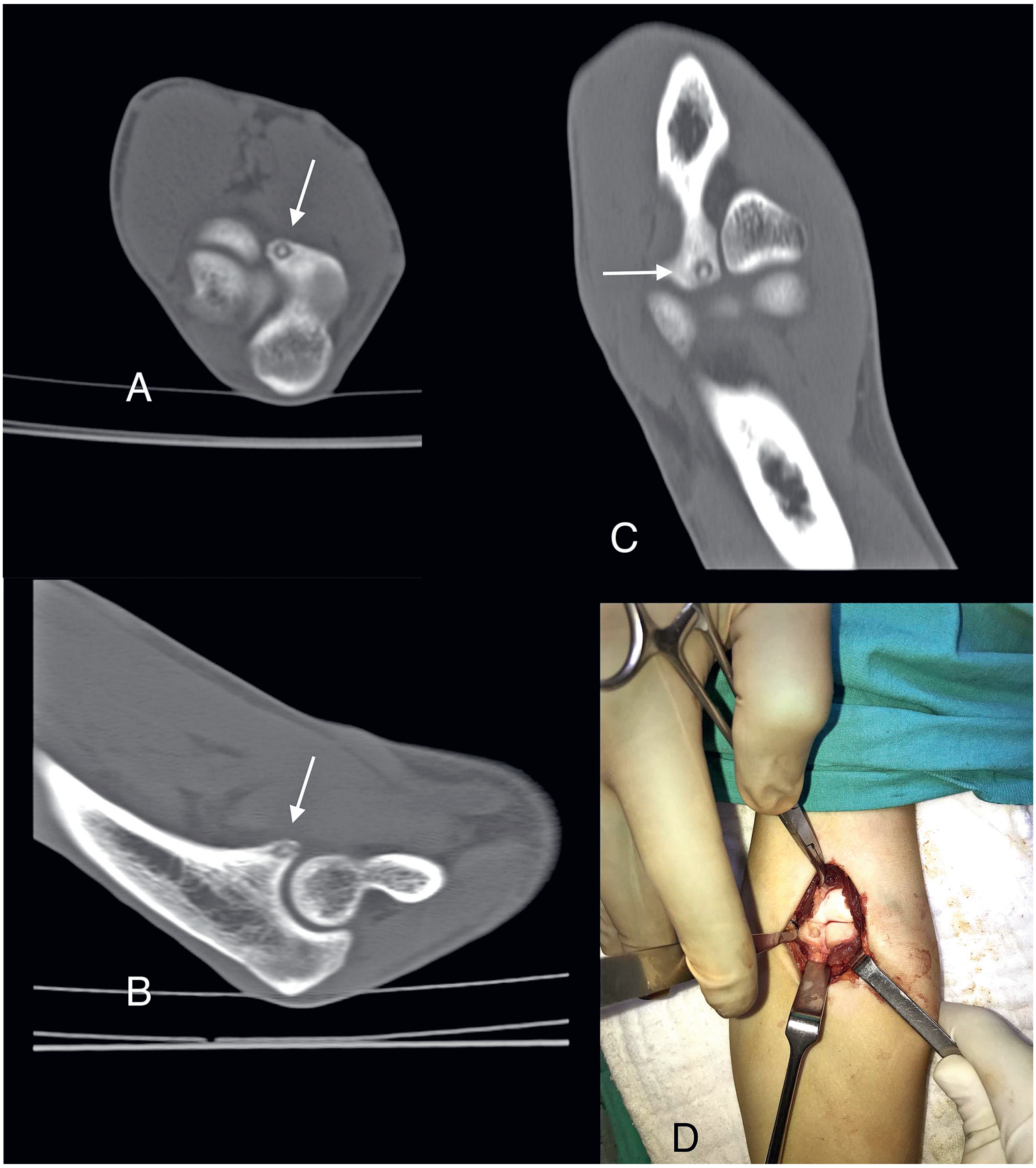

Fig. 3. Surgical excision for a 5 mm diameter OO of the coronoid process in a 26-year-old male is shown (patient-10). Axial (A), sagittal(B), and coronal (C) CT scans show the medullary nidus with surrounding sclerosis. Intra-operative (D) image shows the nidus of OO in the coronoid process at the time of surgery.

it is more often diaphyseal or meta-diaphyseal. The almost constant and often only symptom is pain, with a typical nocturnal pattern, relieved by NSAIDs. The main radiographic sign is a nidus surrounded by dense sclerotic bone tissue. The diagnosis is based on clinical and radiological findings. The diagnosis of OOs may be delayed in patients where there is coexistence of an unusual location with an atypical clinical presentation, because the latter can mimic other clinical entities, such as synovitis, stress fracture, or infection (1, 26). In some patients, the pain may not be relieved at all by NSAIDs because of associated synovitis and/or bone marrow edema (13). Therefore, OOs can be mis-diagnosed. In this study, unusual location of OOs was seen in 17 patients and the mean delay in diagnosis was 8.4 months (range, 3–24 months).

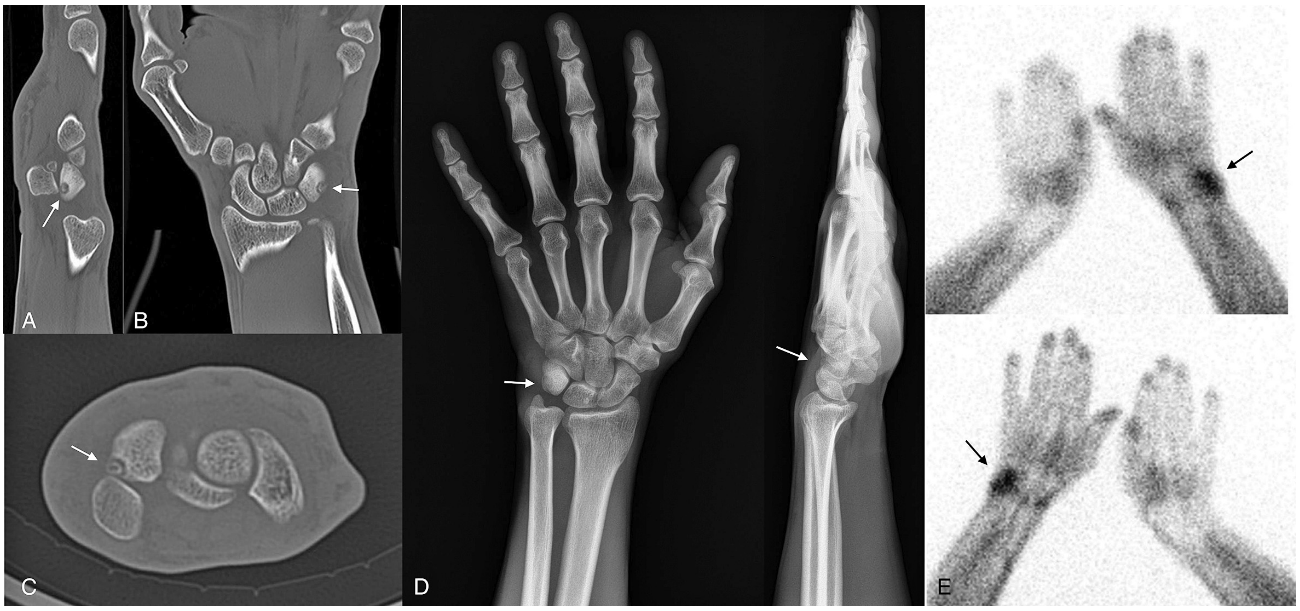

Fig. 4. Sagittal, coronal and axial CT images of a 27-year-old male (patient-13) showing an ovoid cortical-based radiolucent lesion (white arrow) of the triquetrum measuring 4×4 mm in diameter (A-B-C). Pre-operative anteroposterior and lateral radiographs of the wrist showing triquetrum OO (D). Radionuclide bone scan of cortical osteoid osteoma shows focal intense uptake of radioisotope corresponding to the site of radiographic abnormality, which is consistent with osteoid osteoma (E).

Nocturnal pain (64.7%) and pain relief with NSAIDs (41.2%) were common in the current study patients, although these findings were not comprehensive. In literature, these findings have been reported to range from 61% to 90%, and from 67% to 88% respectively (14, 24). A possible reason why these classic symptoms were less common in the current study could be that the OOs were in unusual locations.

Radiography is an initial imaging modality for bone lesions such as OO. Dense sclerotic bone tissue is seen in extra-articular OOs, but may not be observed in intra-articular OOs (Fig. 3). As seen in previous studies, this was also the case in the current study (24). In this study, radiolucent nidus was seen in 7 (41%) patients and osseous, and dense sclerotic bone tissue was seen in 9 (53%) patients. Due to the complex anatomy, radiography may not always visualize the nidus of OO (28), therefore further imaging investigations are needed. However, CT showed all the lesions in this study, in accordance with the findings of previous studies (5, 6). In comparison with CT, MR imaging has been stated to be of limited value in visualizing the nidus (18). Sometimes, MRI shows exaggerated images which occur with the presence of non-specific perilesional synovitis, effusion, and marrow edema, thereby leading to mis-diagnoses. In this study, these findings may have caused the misdiagnosis in some of the 9 patients applied with MRI. It has been reported that OOs may be missed in patients where MRI alone is performed (11). May et al. recommended scintigraphy for unusual localized lesions or atypical clinical presentation (24).

Some authors have also used scintigraphy in addition to radiography and CT to confirm the exact location of the OO (2, 22). Pratali et al. stated that the best method for OO localization is bone scintigraphy (27). In the current study, as there was no doubt about the diagnosis and exact location, there was no need for scintigraphy. In 3 of the current study patients, scintigraphy had been applied at another center before referral to our institution (Figure 4). In these 3 patients, no definitive diagnosis could be made after radiography and MRI examination, so scintigraphy was applied with the consideration of OO as a pre-diagnostic option among other pre-diagnosis. It can be recommended that OO should be considered as a pre-diagnosis in cases of localized long-lasting limb pain.

Unfortunately, OOs may misdiagnosed for various reasons (8, 14, 16, 24) and therefore a delay in diagnosis may often be encountered. In this study, there were similar rates of initial misdiagnoses (59%), with synovitis, growing pains, and stress fracture being the most common.

Although the pain caused by OOs can be relieved with NSAIDs in some patients, mis-diagnosis will lead to prolonged unnecessary pain. OOs in skeletally immature patients may lead to skeletal deformity and joint degeneration (especially in cases with intra-articular lesions) (12, 24). In patient No. 14, the OO was located in the 2nd proximal phalanx at the PIP joint, and PIP arthrodesis was performed because of severe intra-articular degeneration. There were no fractures, no neurovascular injury, or any other late complications seen throughout the post-operative 38-month follow-up period.

According to some studies, the gold standard treatment for the OO is RFA, while for others it is excision (4, 9, 19, 30), and the various advantages of both are known (19). In this study, RFA was applied to 3 patients and excision was applied to 14 patients. All the patients were informed about these treatment options. However, in cases where the lesion was located close to the joint or joint capsule, to the neurovascular structures, or to the skin, it was considered preferable to perform surgical excision rather than RFA for treatment.

The limitations of this study include the small patient population, but in contrast to studies of larger populations, these were patients with OOs in unusual locations. Other limitations could be said to be the relatively short follow-up and the retrospective/observational nature of the study.

CONCLUSIONS

In conclusion, orthopaedic surgeons should suspect OO in especially young patients presenting with long-term complaints of undiagnosed limb pain. Which treatment is superior may vary depending on the location of the lesion between RFA and surgical excision. Although the patient's choice is important in the selection of treatment in OOs, the final decision for treatment should be made considering the site of the unusually located lesion.

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