RESEARCH ARTICLE

SURGICAL MANAGEMENT OF OSTEOID OSTEOMA IN CHILDREN: ABOUT 13 CASES

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Abstract

Osteoid osteoma is a rare benign bone tumor that accounts for 10% of all benign bone tumors and 2 to 3% of all bone tumors in children. It is preferentially located in the long bones, specially the tibia and the femur (Campanacci 1999).

It is characterized by a specific structure (the nidus); made of fabricosteoid, surrounded by a condensation reaction (Greenspan 1993).

The difficulty of the surgery of this lesion comes from its localization, sometimes deep, its anatomical relationships and its small size.

Our report is a retrospective study about a series of 13 cases of osteomaosteoid in children, collected in the pediatric orthopedic surgery department of the CHU Mohammed VI of Marrakech over a period of 7 years.

The aim of our study is to analyze our clinical, paraclinical and therapeutic data, to compare them to the literature, and to focus on the various current therapeutic methods.

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Patients and Methods:--
This is a retrospective study of 13 cases of osteoid osteoma in children, collected in the Pediatric Orthopedic Trauma Department of Mohamed the VI University Hospital Center of Marrakech over a period of 7 years from January 2011 to December 2017.

The data were collected from patient records and consultation follow-up data. We collected: epidemiological data, clinical information, radiological, pathological and therapeutic data.

Patient data were analyzed by SPSS 22.0 software. Qualitative variables were expressed as a percentage and quantitative variables were expressed as means and extremes.

Results:--
Osteomas osteoid represented 5.5% of all bone tumors and 12.6% of benign bone tumors in our study. The ages of our patients ranged from 4 to 15 years with an average age of 9.6 years. We noted a clear female predominance with a sex ratio of 0.3.

Pain was present in all of our patients and was typical with night rhythm and aspirin response in 69.2% of cases. The other clinical manifestations were represented by swelling and lameness, both found in 7 patients.

X-ray were performed in all our patients (figure 1). It highlighted a small lacunar image associated with osteocondensation in 9 patients (69.2%). In 4 patients (30.7%), an image of isolated cortical condensation was noted. A regular homogeneous periosteal reaction was found in 2 cases.

The CT, carried out in 2nd intention after the standard radiography in all the patients, objectified an image of nidus appearing as a small hypodensity with clear contours in the whole of our patients (figure 2). This hypodensity presented a central calcification with regular contours in 10 patients (76.9% of the cases). The location was cortical in 12 patients (92.3%), and medullar in one patient.

Bone scintigraphy was performed in 11 patients. It objectified an intense hyperfixation of the nidus which appeared as a bright and rounded spot surrounded by a lesser crown of fixation.

Magnetic resonance imaging, performed in only two patients, showed a typical aspect of osteoid osteoma with T1 and T2 hyposignal images associated with a perilesional edema, centered by a hypersignal in T2 weighted images corresponding to the nidus.

In our study, the location of the lesion was distributed in the skeleton as follow: Five cases in the femur including 2 cases at the level of the lower femoral metaphysis, 1 case at the level of the neck, 1 case at the inter-trochanteric level and one case at the level of the diaphysis; six cases at the tibial diaphysis; one case at the upper third the humeral shaft and one case at the 1st phalanx of the 3rd finger.

All our patients received Non-Steroid Anti-Inflammatory Drugs before surgery. A good response was noted in 8 patients (61.5%).
Figure 1: Front x-ray of the upper end of the femur showing thickening cortex surrounding an uncalcified lacunar area.

Figure 2: CT cross section showing a hypo dense lesion centered of a calcification with peripheral hyperdensity of the right humeral shaft.

The surgical treatment consisted of a resection en bloc in 11 patients and a curettage of the nidus in 2 patients. Postoperative immobilization was systematic, lasting 45 days for the lower limbs and 30 days for the upper limbs.

There was no operative incident and the post-operative courses were simple in all of our patients. The length of hospital stay varied between 2 and 5 days with an average length of stay of 4 days.

We observed in all our patients a complete relief of tumor pain within few days after the intervention. All the pathological results confirmed the diagnosis of osteoid osteoma.

No early or late complications were noted. However, a recurrence occurred in 2 patients after 14 and 16 months. These two patients were reoperated by enblock resection and did not develop a new recurrence.
Discussion:
This lesion was first described by Berstrang in 1930 and was named osteoid osteoma by Jaffe in 1935 who admitted its neoplastic origin. (Jaffe 1935).
Osteoid osteoma is characterized by the presence of two entities: the nidus, which is the tumor proper, and a variable peripheral osteogenesis reaction.

The nidus is composed of hyper vascularized connective tissue, giant cells, osteoblasts and osteoid substance. Reactive osteogenesis is of the classic lamellar type. (Jaffe 1935, Aynaci et al. 2007).

The ethiopathogenesis of this lesion has not yet been fully elucidated. Several pathogenic hypotheses have been advanced, notably neoplastic, congenital, embryonic, infectious, inflammatory and traumatic (Jaffe 1935, Goldman et al. 1993, Vancamp 2015).

Osteoid osteoma is observed mainly in young subjects: 50% of cases occur before the age of 20, the peak age being between 10 and 20 years (Aschero et al. 2009). Contrary to our results, a male predominance is found in most of the series published in the literature.

The pain associated to osteoid osteoma has been described by several authors as mechanical pain occurring during movement and progressing gradually to a typical inflammatory pain predominantly nocturnal, usually responding to aspirin (Wu et al. 2017). Lameness is the second sign in order of frequency. The swelling is more common in case of long bones involvement. (Lee et al. 2006)

The long bones constitute the elective location of osteoid osteomas, the femur being the most affected site, followed by the tibia, the phalanges and then the vertebrae (Atesok et al. 2011).

The osteoid osteoma can be explored by standard radiography, computed tomography scan (CT), scintigraphy or MRI (Chai et al. 2010). Plan radiograph May be normal or may show a solid periosteal reaction with cortical thickening. The nidus is sometimes visible as a well-circumscribed lucent region, occasionally with a central sclerotic dot (Greenspan 1993).

CT is excellent at characterizing the lesion and is the modality of choice. It typically shows a focally lucent nidus within surrounding sclerotic reactive bone. A central sclerotic dot may also be seen. It also allows a size measurement and a precise endo-osseous topographical location, essential for any operative procedure (Assounet al. 1994).

Skeletal scintigraphy will show typical focal uptake and at times will show a double intensity sign which, if present is highly specific (Chai et al. 2010). Although MRI is sensitive, it is non-specific and is often unable to identify the nidus (Davies et al. 2002).

The treatment of osteoid osteoma has a symptomatic component and a curative component. Acetylsalicylic acid is quite effective against pain by inhibiting prostaglandins. The response rate varies among series between 70% to 100% (Falappa et al. 2011, Whitmore et al. 2016, Erol et al. 2017, Wu et al. 2017). It was 61.5% in our series.

Surgical excision constitutes the standard curative treatment. The conventional method is a block resection of the nidus with a few millimeters of reactive osteosclerosis. However, this technique can be difficult to perform in locations where the approach is narrow (Bisbinaset al. 2004). The alternative is then curettage of the lesion. Percutaneous bone resection drilling can also be proposed (Rauxet al. 2014). Other less invasive techniques have proven successful, including laser photocoagulation, radiofrequency ablation, cryoablation, high intensity focused MRI guided ultrasound (FUS-MG) or alcohol ablation (Falappa et al. 2011, Whitmore et al. 2016, Erolet al. 2017, Wu et al. 2017).

The results of these different techniques in terms of pain remission, complications and recurrence are more or less comparable (Table 1).
There is growing evidence, that osteoid osteoma naturally resolves spontaneously with time and can be treated conservatively with NSAIDs in certain groups of patients (Feletar and Hall 2002). The average time to resolution is 33 months.

Table 1: Comparison between the different operating techniques.

| Study                        | Radiofréquence (Feletar and Hall 2002) | Cryoablation (Whitmore et al. 2016) | Laser ablation (Wu et al. 2017) | Alcoolisation (El-Mowafiet al. 2003) | FUS–MG (Masciocchi et al. 2016) | Conventionalsurgery | Our study |
|------------------------------|----------------------------------------|-------------------------------------|---------------------------------|-------------------------------------|---------------------------------|---------------------|-----------|
| number (N)                   | 51                                     | 29                                  | 36                              | 15                                  | 15                              | 13                  |           |
| Hospitalisation time (jours) | 4                                      | 1                                   | -                               | 3 à 4                               | 2                               | 4                   |           |
| Pain release                 | 100%                                   | 96%                                 | 100%                            | 100%                                | 96%                             | 100%                |           |
| Major complications (a)      | 100%                                   | 0%                                  | 0%                              | 0%                                  | 0%                              | 0%                  |           |
| Minor complications (b)      | 21%                                    | 40%                                 | 6,6%                            | 20%                                 | 0%                              |                     |           |
| Relapses                     | 2%                                     | 0%                                  | 0%                              | 0%                                  | 6,6%                            | 15,3%               |           |

(a) Postoperative fracture, neurological or vascular deficit.
(b) Skin burns, inflammatory reactions, skin necrosis, infections, weak numbness and Persistent intermittent pain in the extremities with intense physical exercise [43]

Conclusion:

The osteoid osteoma is a rare benign tumor of which the treatment is based on complete surgical excision of the nidus; it is the necessary and sufficient condition for healing.

The Up-to-dateness of its management is represented by the progress of minimally invasive resection techniques.

The new techniques: laser, radiofrequency and cryoablation are increasingly used in children because of their minimally invasive nature.

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