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CLINICAL REPORT

Osteoid osteoma distal to a hip arthroplasty

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Summary The authors report a case of osteoid osteoma distal to a hip prosthesis in a 56-year-old patient. This rare association was difficult to diagnose; at first, the pain seemed to be of mechanical origin, suggesting a delayed painful reaction to the prosthesis. The results of bone scan as well as the CT scan ones helped orient the diagnosis. Excision biopsy, using bone trephining, completed by an iliac bone auto-graft resulted in a cure with no residual instability of the prosthesis above the tumor. Bone scan with radio-isotopes to localize the lesion was particularly helpful in this instance to secure the final diagnosis.

Observation

A 56-year-old patient consulted with left femoral pain extending down the leg that appeared to be mechanical, with no associated trauma, and increasing at night, that had been present for a year. Pain was relieved by taking aspirin.

Twelve years before, the patient had received a left hip prosthesis due to coxarthrosis and had been totally asymptomatic until the year before.

The clinical examination did not show any pain from movement or during palpation, no curving or any visible deformity. Hip mobility was normal. Plain films did not show any sign of loosening of the prosthesis and were considered normal. It was only after the diagnosis was established by other imaging techniques that a dense fusiform diaphyseal reactional sclerosis was seen on the edge of the antero-posterior view of the plain film (Fig. 1) and a radiolucent image surrounded by ringlike mineralization was identified in a lateral view (Fig. 2).

Bone scintigraphy was performed because of persistent pain showing increased uptake in an image three centimeters below the tail of the prosthesis. A targeted CT scan showed a characteristic lucent area surrounded by reactive bone formation characteristic of osteoid osteoma in the middle third of the left femur (Figs. 3 and 4).

After localization of the lesion by CT scan, the patient underwent surgery for excision by bone trephining and an iliac bone graft. The anatomopathological analysis of the tissue sample did not provide further information, because the sample was fragmented due to the use of the trephine, but the patient stated that pain completely disappeared immediately after surgery and it has not recurred 3 years later. All plain film images have also remained normal.

Discussion

Osteoid osteoma is a frequent tumor and the third most frequent of the benign bone tumors. The present case illustrates the clinical symptoms that are reported in this disease, in particular pain, which is found in most cases (95 to 99% of cases according to Kransdorf et al. [1]), which is

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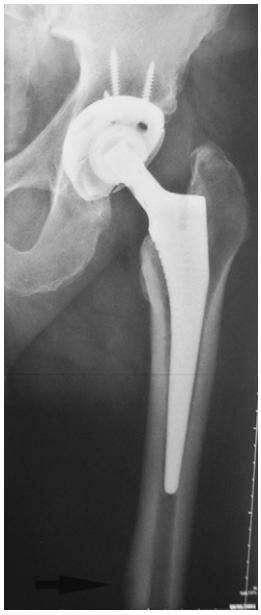

Figure 1 Anteroposterior plain film of the left femur. Dense fusiform diaphyseal reactive sclerosis (arrow).

focalised (usually in the long bones, especially the femur [2]), continuous, but predominantly nocturnal [1], chronic [3] and worse during effort at first [1,3]. It is relieved by NSAID and aspirin because these drugs inhibit the synthesis of prostaglandins by the nidus which results in an inflammatory and painful reaction [3—6]. This was also found in our patient.

It is usually found in men (a sex ration between 1.8/1 and 4/1) but unlike our case, usually affects young patients in half the cases between 10 and 20 years old, with a median age of 19 and a range of between 7 months and 72 years old. [1]. The etiology is unknown and usually develops for

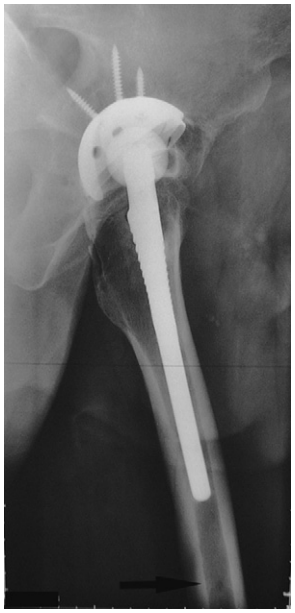

Figure 2 Plain film of the left femur. Lateral view. Radiolucent area with ringlike mineralization (arrow).

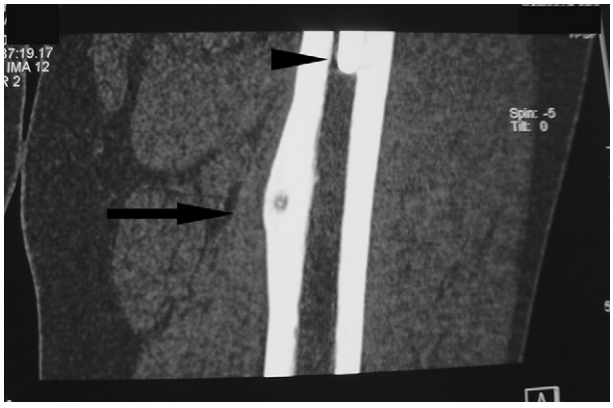

Figure 3 Coronal slices on CT of the left femur. Lucent area surrounded by reactive bone formation suggesting osteoid osteoma (arrow) below a hip prosthesis (arrow head).

no identifiable cause. Although numerous cases have been described of osteoid osteoma after a fracture, we did not find any other cases in patients with hip prostheses in our review of the literature.

Thus, although our patient presented with predominantly nocturnal pain that was relieved by aspirin, the diagnosis was delayed and misled by the presence of a hip prosthesis directly over the lesion which was thought to be the cause of the pain.

With this clinical picture, in the presence of diaphyseal lesions, traditional X-ray can be used to identify a radiolucent image with a ring of mineralization, the characteristic lucent area surrounded by reactive bone formation when the center of the nidus is calcified. Our clinical case once again confirms this, as these signs were identified retrospectively on the plain films, but were not identified initially because of the supposed responsibility of the hip prosthesis in the patient's described symptoms.

Targeted CT scan provided thin millimetric slices showing the same signs but which are perfectly identifiable in

Figure 4 Axial slices on CT of the left femur. Lucent area surrounded by reactive bone formation. Image suggesting osteoid osteoma.

reactional osteosclerosis, so that the lesion could be localised and measured [1,5]. This is therefore the best technique for detecting and characterizing osteoid osteomas, as long as the radiologist is aware of the topography.

Because of the hypervascularisation of the nidus, there is increased uptake of radioactive tracers with bone scintigraphy and a double radiolucent ring due to the peripheral sclerotic bone [1,4]. This is the ''double-density sign'' [5] and is a very important examination to help diagnose atypical cases such as ours, although false negatives may occur.

Although certain authors suggest using MRI, this should be limited because it is less reliable than a scan [5—7]. The images also risk showing extensive lesions because of the edema thus suggesting malignant tumors [3].

The diagnosis is therefore made in the presence of typical clinical signs, increased uptake in scintigraphy and CT scan with the rosette image. Besides these obvious signs, a differential diagnosis must be made.

In addition to having to make the classic differential diagnoses for osteoid osteoma and exclude in particular non-ossifying fibroma, chronic osteomyelitis caused by Brodie's abscess, osteoblastoma, fatigue fracture [4] and bone infarct [3], in our patient, chronic pain from the hip prosthesis was the most important question.

Thus, delayed bone infection could be excluded because of the absence of clinical, biological and radiological signs suggesting infection. In the same way, aseptic loosening was excluded by X-ray and by the lack of correlation between pain and physical effort. There was no sign of the ''stress shielding'' phenomenon corresponding to bone resorption in contact with the prosthesis because this phenomenon is associated with a plain film image of diffuse bone lysis, most often proximal, which was not found in our patient. The diagnosis of spinal or algodystrophic pain was excluded by the delayed appearance and depth of the pain located in the middle third of the thigh. The diagnosis of pain in reaction to the hip prosthesis, even if it was delayed, was harder to exclude, which was why bone scintigraphy was prescribed, and showed localised uptake under the tail of the prosthesis, thus helping orient the diagnosis.

These lesions may sometimes naturally regress over several years, which is why certain practitioners recommend a medical approach with long term NSAID treatment or aspirin. [1,3,5]. However, in addition to the risk when taking these drugs long term, the possible progression of the disease, especially in young patients and the risk of limb deformation from the osteogenic reaction which may result in professional disabilities and an incapacity to practice sports, as well as the unpredictability of the mechanism of regression, do not support this approach in patients who can receive other treatment.

Thus the rule is surgical treatment with complete excision of the nidus, resulting in complete, immediate disappearance of symptoms.

There are two steps to treatment:

- Localisation of the lesion which is necessary because of its small size, is usually made by CT scan which, as mentioned above, is the most accessible and reliable technique, with the placement of a pin in the nidus, although other techniques have been described. Tetracycline labelling which makes the nidus fluorescent with special glasses, may also be used or bone scintigraphy with a radioactive tracer, although this requires special equipment [2].
- Excision can be performed by various methods. The so-called traditional method includes complete resection with a chisel or oscillating saw and a graft or support osteosynthesis depending on the amount of loss of bone substance. Although this method is used less and less in favor of less invasive methods, it still has a place if there is doubt about the diagnosis, making anatomopathological confirmation necessary [3,5]; percutaneous resection of the nidus [8]; interstitial laser [5,9] or radiofrequency [6,10] photocoagulation are all techniques which are being actively investigated but which have the disadvantage of not providing a definite anatomopathological diagnosis, unlike the percutaneous technique which provides it in 50% of cases [3,5].

Nevertheless, the most effective sign of successful treatment of this lesion is the complete disappearance of pain in the immediate post-operative period. Thus, if pain re-occurs this is a sign of incomplete resection.

The more specific problem in the present case is the presence of a hip prosthesis above the lesion. Indeed, treatment must include total resection of the osteoid osteoma, avoiding any damage to the prosthesis, in particular any infection, which might cause sepsis, but also instability of the prosthesis and favour loosening. Moreover, the restraining pins in the distal end of the prosthesis, because of the differences in elasticity between the bone and the metal, associated with the weakening of the bony structure from the resection, were factors increasing the risk of secondary fracture of the distal end of the femur. This is why we decided upon an approach which spared as much bony structure as possible, by percutaneous resection of the nidus, which we associated with an iliac graft because of the risk of instability of the prosthesis.

Conclusion

In conclusion, we report the case of a 56-year-old patient presenting with osteoid osteoma just below a total hip prosthesis treated by percutaneous approach. The originality of this case is the presence of a hip prosthesis above the lesion making diagnosis and treatment difficult, a case which we did not find in our study of the literature.

Conflict of interests

None.

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