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Sclerosing Osteomyelitis of Garré. Case Report and Discussion

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Abstract

Adolescent JRM, male, mixed-race, 18 years old, presents with pain in his arm for months, treated in another service, who is referred to our consultation at the orthopedics and traumatology service, after a first diagnostic impression he is performed studies and surgical treatment with biopsy. Garré's chronic sclerosing osteomyelitis was diagnosed. The clinical, imaging, and pathological findings are illustrated and discussed.

Keywords: Osteomyelitis, Adolescent, Humerus

Introduction

When faced with a bone lesion, the attending physician should try to determine whether it is a pseudo-tumor lesion, a benign bone tumor or a malignant bone tumor, to attempt better therapeutic management.

Pseudo-tumor lesions make up a group of non-neoplastic bone lesions that bring together very diverse conditions. The humerus is the third most common site of bone tumors; in Garré sclerosing osteomyelitis the bone is hypertrophic, very sclerotic, dense, without a medullary cavity. Often clinically inactive, without inflammatory symptoms, but it can be painful.

Garré sclerosing osteomyelitis is a type of chronic osteomyelitis that mainly affects children and young people with a history of chronic pain in the metaphyseal area of the long bones or the jaw [1].

Sclerosing osteomyelitis of Garré is a rare and specific type of chronic osteomyelitis that mainly affects children and young adults. It is a well-described entity in the dental literature and the most commonly involved bone is the mandible. Involvement of long bones such as femur and tibia has been reported. A few terms have been used to describe this subtype, such as ossifying periostitis, chronic osteomyelitis with proliferative periostitis, and non-suppurative sclerosing osteomyelitis. Laboratory results usually show mild elevation of inflammatory markers such as C-reactive protein (CRP) and erythrocyte sedimentation rate (ESR). Cultures from blood and tissue are usually negative [2, 3].

There are few clinical cases of Garre's Chronic Osteomyelitis (COG) cited in the literature. Apart from the fact that it is a very rare entity, this could be due to the fact that there were controversies and confusions in the use of terminology. The terms Osteitis and Osteomyelitis were used interchangeably by many authors, which has generated confusion. For this reason, the authors decide to present this striking case and review the literature on the subject.

Patient Presentation

We present adolescent JRM, male, mixed-race, 18 years old, who comes to consultation with pain in his right arm for several months. He is a student. He has a personal history of suffering from thrombosis of the right upper limb. Several months ago, he began to have pain, after which several weeks ago he presented an increase in volume in the middle and distal portion of the arm throughout its circumference, he had difficulty performing physical effort, but it did not disable him from carrying out daily life and his studies. He was evaluated by angiology in a first consultation, from which he was referred to our service where he was evaluated collectively.

We perform a general and special interrogation and physical examination of the upper limb.

During the interrogation, he stated that his psychomotor development was adequate, he was not sickly, he did suffer a thrombotic injury in his right upper limb several years ago without knowing what the cause of said event was. Now it started with low-intensity dull pain in the afternoons, which has not increased at any time. He did not report any general condition or fever. Several weeks ago, an increase in volume began in the middle and distal third of the right arm.

During the general physical examination, we found right axillary lymphadenopathy of more or less one to one and a half centimeters, we did not find other general physical alterations. Upon special examination of the right upper limb, there is a visible and palpable tumor throughout the entire circumference of the right arm that involves from the middle third to the elbow. The mobility of the right upper limb and the right hand maintain good mobility. Figure 1.

From the appearance of the first symptoms to the definitive diagnosis, around 3 years passed.

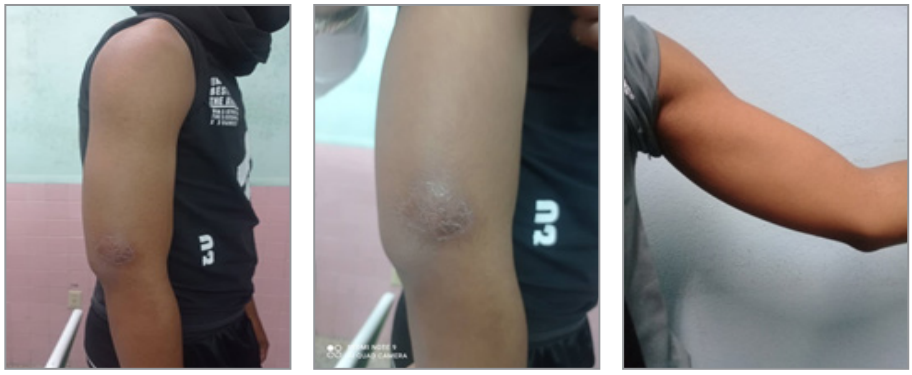

Figure 1: Note how there is an increase in volume in the mid-lower region of the right arm.

To confirm the suspected diagnosis, we perform complementary hematological, radiological and pathological examinations.

Table 1: Complementary hematological tests performed and their results.

Complementary	Results	Reference values
Hemoglobin	14,3 G/dl	13.0 – 17.0 G/dl
Leukogram	9,2x109 /L	5 – 10 x 109 /L
Erythro- sedimentation	104 mm/hr	0 – 10 mm/hr
Rheumatoid factor	Negativo	< 8 UI/ml
C- reactive protein	12,73 miligramos por litro (mg/l)	10 miligramos por litro (mg/l)
Serum calcium	1.9 mmol/L	2.0 – 2.6 mmol/L
Match	1.3 mmol/L	0.8 – 1.6 mmol/L
Alkaline phosphatase	219 U/L	98 – 279 U/L
Serum creatinine	87 µmol/L	46 – 106 µmol/L

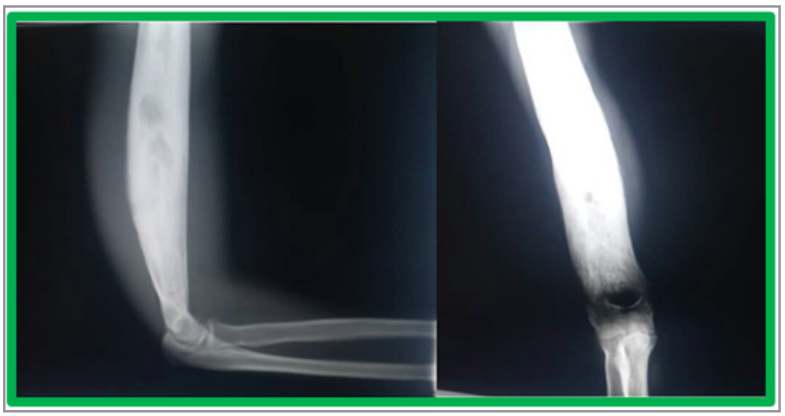

Figure 2: X-ray of the right arm showing a bone lesion that affects the humerus in its entirety, osteolytic lesions and loss of the spinal cavity appear.

In simple radiology at the outpatient clinic level, a significant lesion was found in the entire distal half of the humerus bone with loss of bone structure, sclerosis and signs of periostitis.

Tomographic studies on a multi-slice tomography (No: 0247-24) are indicated on the thorax, abdomen and right upper limb.

Tomographic Report

Chest. Pulmonary fibro emphysema with small emphysematous bullae towards the right upper lobe, with associated fibrous tract. No pleural effusion or mediastinal lymphadenopathy.

Abdomen. Liver of homogeneous density, without focal lesion. No hepatomegaly, normal gallbladder, kidney, pancreas and kidneys. Prostate measuring 31x27 mm. We did not find free fluid in the cavity, nor did we find retroperitoneal lymphadenopathy.

Right upper limb. We found lymphadenopathy, the largest measuring 23mm. At the level of the right humerus we find an increase in bone density throughout the diaphysis with areas of osteolysis that in some places causes rupture of the cortex, an increase in the soft tissues, with changes in density, the possibility of infiltration in the soft tissues. be taken into account. Figure 3.

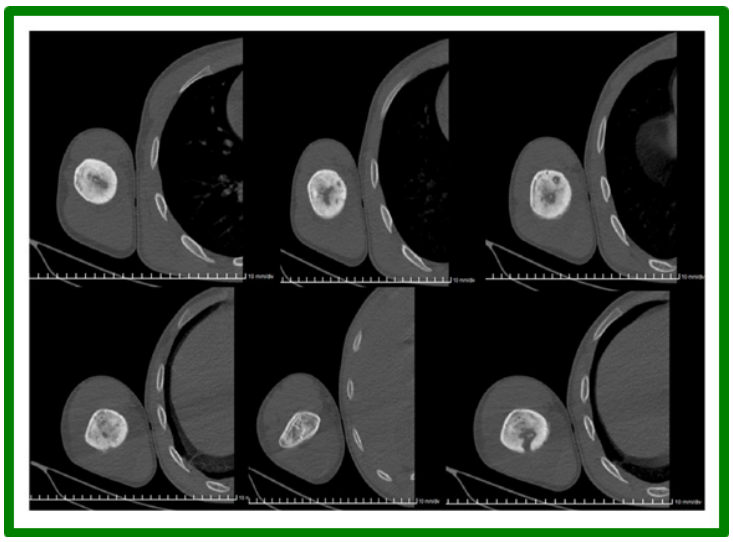

Figure 3: Images of bone condensation and loss of the spinal canal with sclerosis and periostitis are observed. The last image shows the surgery to decompress the lesion and take a sample for biopsy.

Biopsy 0244-24-1-2024. Observe osteoblastic conglomerates, without signs of mitotic activity or malignant cells.

Discussion

In 1893 Garré described a chronic form of osteomyelitis, which caused distention and thickening of the bone and was not accompanied by suppuration, sequestration or fistula formation. Later Hardmeier introduced the term primary chronic sclerosing osteomyelitis, to differentiate it from chronic osteomyelitis. These two bone infectious entities have great differences [4].

According to López Álvarez et al., Garré chronic sclerosing osteomyelitis is an extremely rare variety, which mainly affects children and young adults, presents great difficulties for its diagnosis and treatment, which is why a close relationship between the orthopedic, radiologist and and pathologist [5]. Most reports agree that Garré's sclerosing osteomyelitis primarily affects children and young adults. Alvares López 5 cites Segev and Morrissy, who suggest that this type of osteomyelitis is almost exclusive to these ages. The age of our patient corresponded to what was described. The most common location is the metaphyseal regions of the long bones, but the mandible is also highly susceptible.

Garré sclerosing osteomyelitis has been reported to be associated with several chronic autoimmune diseases, including inflammatory bowel conditions, Wegener's granulomatosis, psoriasis, and Takayasu vasculitis. It may represent a juvenile form of seronegative spondyloarthropathy or a pediatric variant of SAPHO syndrome (synovitis, acne, pustulosis, hyperostosis and osteitis). This disease usually appears in people under 25 years of age, when the activity of the osteoblasts of the periosteum is at its peak. The most common location is the jaw, but it can occur in any bone. Most patients have an elevated erythrocyte sedimentation rate or C-reactive protein during exacerbations, with a normal white blood cell count. Result very similar to that presented in our case [6].

In Garré sclerosing osteomyelitis the bone is hypertrophic, very osteosclerotic, dense, without a medullary cavity. Often clinically inactive, without inflammatory symptoms, but it can be painful. The clinical course is characterized by an insidious onset with manifestations of pain and local distention of the affected bone, light or intense swelling in the area, with a moderate increase in erythrocyte sedimentation rate. It is a pathology that tends to recur, even over the years. Due to the clinical and radiological characteristics of this lesion, it is almost impossible to distinguish it from a bone tumor before making a pathological

diagnosis. Therefore, it must be differentiated from osteogenic sarcoma, Ewing sarcoma, osteoblastoma, fibrous dysplasia and Paget's disease, which have a lot of clinical and radiological similarity [7].

The radiographic changes described by Garré in the initial stage are characterized by pronounced sclerosis with isolated cystic areas, while in the more advanced form sclerosis is the dominant factor. See Figure 2, here very similar characteristics are shown. to those described by Garré [8-10].

For clinical diagnosis, it is proposed that criteria be met such as absence of germs, absence of pus and absence of clinical and laboratory data for infection [4, 11].

No effective form of treatment is currently described in the literature. The use of fenestration has only shown temporary relief. The administration of antibiotics, although their effectiveness has not been proven, is indicated in these patients. The use of non-steroidal anti-inflammatory drugs, corticosteroids and colchicine, the latter inhibiting the chemotaxis of polymorphonuclear cells, are planned as new forms of treatment; some authors even propose that the use of non-steroidal anti-inflammatory drugs may constitute the only form of treatment [5, 12, 13].

It is concluded that Garré sclerosing osteomyelitis is a difficult diagnosis, due to its presentation and its management is uncertain and debatable since its etiology is still unclear.

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