Clavicle chronic recurrent multifocal osteomyelitis: Surgical excision and neo-clavicle

Jorge H. Costa, Tiago P. Marques, Miguel Pádua

ABSTRACT
Abstract is not required for Clinical Images
CASE REPORT

This is a case of an 11-year-old boy with left clavicle pain with two months of evolution presented to emergency department. No history of trauma, fever or systemic signs, recent illness and no relevant personal or familiar medical history. No neurological or vascular abnormalities were found in the upper arms. Left shoulder X-ray revealed a hyperdense lesion and increased thickness of left clavicle (Figure 1). In this context a computed tomography (CT) scan and after a magnetic resonance imaging (MRI) scan were ordered. Both scans demonstrated morphological changes of the median half of the clavicle, cortical destruction and periosteal reaction. Also changes in the surrounding soft and muscular tissue and bulky and numerous locoregional ganglion formations (Figures 2 and 3). Blood analysis, blood cultures and bone biopsy were negative for infection and neoplastic disorder. In this context, by exclusion, and in the presence of two Jansson major diagnostic criteria (radiologically proven osteolytic/osteosclerotic bone lesions and sterile bone biopsy with signs of inflammation) [1], we reached the diagnosis of chronic recurrent multifocal osteomyelitis (CMRO) (Table 1). Patient was treated with NSAIDs for six months and a short course of corticoids without any response. By keeping complaints of marked and recalcitrant pain in clavicle region, we opted for the surgical treatment: resection of the lesion en bloc, about 7 cm of clavicle (Figure 4), maintaining the integrity of periosteum to allow neo-osteogenesis of the clavicle. No bone grafting or another material was interposed in the dead space left by the resection of the clavicle. Post operatory X-ray, showing a radiopaque area corresponding to the excision of the medial portion of the clavicle (Figure 5).

At third month after surgery, there is an image compatible with a neo-clavicle and patient is asymptomatic (Figure 6).

Figure 1: X-ray revealed a hyperdense lesion and increased thickness of left clavicle.
DISCUSSION

Chronic recurrent multifocal osteomyelitis is a non-bacterial bone inflammation (osteitis), that has symptoms similar to the conventional osteomyelitis, but without infection. It is characterized by multifocal bone...
lesions with pain and swelling recurring over months to years, accounting for 2 to 5% of all cases of osteomyelitis [2–3] mostly children and adolescents. The differential diagnosis includes bacterial osteomyelitis, Ewing sarcoma, leukemia, lymphoma, rhabdomyosarcoma, neuroblastoma metastasis, eosinophilic granuloma or Langerhans cell histiocytosis. The tubular long bones, pelvis, hip, sternum and mandible are the most affected bones [4–5]. The CRMO, in pediatric patients, may be associated with various skin disorders: Pustulosis palmoplantar is syndrome [6–8], diffuse pustulosis, psoriasis vulgaris, acne [9–11], Sweet’s syndrome and pyoderma gangrenosum [9]. Multiple sites of apparent osteomyelitis with negative pathology and cultures and no response to antibiotherapy characterize it [12]. NSAIDs as naproxen, indomethacin or aspirin are the initial therapy. A short course of corticosteroids can be used in refractory cases. Other treatments such as sulfasalazine or methotrexate [6], bisphosphonates [13] and hyperbaric chamber [14] are also indicated in refractory cases. Antibiotics do not seem to be helpful. Although surgical treatment has been used, its role is not yet clearly defined. We opted for a surgical excision of the clavicle, because of the constant and intense pain with no response to six months of conservative treatment. We think that the good response to surgery makes this case exceptionally unusual. The prognosis for these patients is however good. In one study, 17 of 23 patients had complete resolution of the clinical findings, at an average of 5.6 years after diagnosis. Six patients continue to have active disease, and the other six had intermittent relapses or chronic pain. 78% had no physical impairment [2], but further prospective studies are needed to determine the optimum outcome measures and treatment strategy [15].

**CONCLUSION**

The absence of positive microbiologic results, the clinical course and the presence of reactivating and remitting lesions over time suggest the diagnosis of chronic recurrent multifocal osteomyelitis. As the disease is self-limiting, knowledge of its characteristics appearance can lead to the most appropriated treatment, and can prevent overly aggressive medical and surgical evaluation and treatment.

---

<table>
<thead>
<tr>
<th>Major diagnostic criteria</th>
<th>Minor diagnostic criteria</th>
</tr>
</thead>
<tbody>
<tr>
<td>Radiologically proven osteolytic/sclerotic</td>
<td>Normal blood count and good</td>
</tr>
<tr>
<td>bone lesion</td>
<td>general state of health</td>
</tr>
<tr>
<td>Multifocal bone lesions</td>
<td>CRP and ESR mildly-to-moderately elevated</td>
</tr>
<tr>
<td>Pustulosis palmoplantar or psoriasis</td>
<td>Observation time longer than 6 months</td>
</tr>
<tr>
<td>Sterile bone biopsy with signs of</td>
<td>Hyperostosis</td>
</tr>
<tr>
<td>inflammation and/or fibrosis, sclerosis</td>
<td>Associated with other autoimmune</td>
</tr>
<tr>
<td></td>
<td>diseases apart from pustulosispalmoplantar or psoriasis</td>
</tr>
<tr>
<td></td>
<td>Grade I or II relatives with autoimmune or autoinflammatory</td>
</tr>
<tr>
<td></td>
<td>disease or non-bacterial osteitis</td>
</tr>
</tbody>
</table>

*CRMO is confirmed by two major criteria or one major and three minor criteria*
REFERENCES


Edorium Journals: An introduction

Edorium Journals Team

About Edorium Journals
Edorium Journals is a publisher of high-quality, open-access, international scholarly journals covering subjects in basic sciences and clinical specialties and subspecialties.

Invitation for article submission
We sincerely invite you to submit your valuable research for publication to Edorium Journals.

But why should you publish with Edorium Journals?
In less than 10 words - we give you what no one does.

Vision of being the best
We have the vision of making our journals the best and the most authoritative journals in their respective specialties. We are working towards this goal every day of every week of every month of every year.

Exceptional services
We care for you, your work and your time. Our efficient, personalized and courteous services are a testimony to this.

Editorial Review
All manuscripts submitted to Edorium Journals undergo pre-processing review, first editorial review, peer review, second editorial review and finally third editorial review.

Peer Review
All manuscripts submitted to Edorium Journals undergo anonymous, double-blind, external peer review.

Early View version
Early View version of your manuscript will be published in the journal within 72 hours of final acceptance.

Manuscript status
From submission to publication of your article you will get regular updates (minimum six times) about status of your manuscripts directly in your email.

Our Commitment

Six weeks
You will get first decision on your manuscript within six weeks (42 days) of submission. If we fail to honor this by even one day, we will publish your manuscript free of charge.

Four weeks
After we receive page proofs, your manuscript will be published in the journal within four weeks (31 days). If we fail to honor this by even one day, we will publish your manuscript free of charge and refund you the full article publication charges you paid for your manuscript.

Mentored Review Articles (MRA)
Our academic program “Mentored Review Article” (MRA) gives you a unique opportunity to publish papers under mentorship of international faculty. These articles are published free of charges.

Favored Author program
One email is all it takes to become our favored author. You will not only get fee waivers but also get information and insights about scholarly publishing.

Institutional Membership program
Join our Institutional Memberships program and help scholars from your institute make their research accessible to all and save thousands of dollars in fees make their research accessible to all.

Our presence
We have some of the best designed publication formats. Our websites are very user friendly and enable you to do your work very easily with no hassle.

Something more...
We request you to have a look at our website to know more about us and our services.

We welcome you to interact with us, share with us, join us and of course publish with us.

CONNECT WITH US

Edorium Journals: On Web
Browse Journals

This page is not a part of the published article. This page is an introduction to Edorium Journals and the publication services.