

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

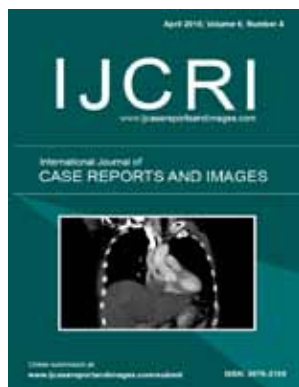
Jorge H. Costa, Tiago P. Marques, Miguel Pádua

ABSTRACT

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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 operative 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.

Jorge H. Costa¹, Tiago P. Marques², Miguel Pádua³

Affiliations: ¹MD, Orthopedics and Traumatology Resident at Hospital Curry Cabral, Lisboa, Portugal; ²MD, Orthopedics and Traumatology Resident at Hospital Santarém, Santarém, Portugal; ³MD, Orthopedics and Traumatology Resident at Hospital Fernando da Fonseca, Amadora, Portugal.

Corresponding Author: Jorge Homero Costa, Orthopedics and Traumatology Resident at Hospital Curry Cabral, Lisboa 1100-291, Portugal; Ph: +351 960333405; Email: jorgehomerocosta@hotmail.com

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Figure 2: Computed tomography scan morphological changes of the median half of the clavicle (increased thickness) and cortical destruction.



Figure 3: A coronal T1-weighted magnetic resonance imaging (MRI) scan showing an increased thickness of left clavicle and changes in the surrounding soft and muscular tissue and bulky with numerous locoregional ganglion formations.



Figure 4: En bloc resection of 7 cm of clavicle.



Figure 5: Postoperative X-ray showing a radiopaque area corresponding to the excision of the medial portion of the clavicle



Figure 6: Image compatible with a neo clavicle, three months after surgery.

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

Table 1: Proposed major and minor diagnostic criteria of CRMO*

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

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

lesions with pain and swelling recurring over months to years, accounting of 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 palmoplantaris 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.

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Author Contributions

Jorge H. Costa – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Tiago P. Marques – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Miguel Pádua – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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