Case Report

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Chronic Recurrent Multifocal Osteomyelitis with Non-Contiguous Involvement of the Spine: A Case Report and Review of Literature

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Abstract

Background: Chronic Recurrent Multifocal Osteomyelitis (CRMO) is a chronic non-bacterial osteomyelitis that affects children and adolescents.

Case Presentation: A 9-year-old previously healthy boy initially presented with vague abdominal pain, for which he was investigated but no active intervention was undertaken. There was increasing pain and deformity over the back noted a week later.

CT thorax was performed to identify the lesion in the rib and an open biopsy was done (resection of diseased rib and transverse process). Biopsy report was suggestive of chronic non-infective osteomyelitis (CRMO). A whole-body magnetic resonance imaging (MRI) showed an additional lesion in the right acetabulum. There was significant relief of symptoms with oral naproxen and intravenous Zoledronate.

At 15 years of age, there was no pain or deformity in the spine and vertebral lesion had reconstituted. The patient was asymptomatic.

Conclusion: CRMO is an under-diagnosed condition. A delay in the diagnosis can result in morbidity in the form of prolonged antibiotic therapy and multiple invasive investigations (biopsy). In most cases, it is a diagnosis of exclusion. This case of a boy with CRMO with main involvement of the spine is presented for its rarity.

Keywords: CRMO, Spine, Zolendronate, Osteomyelitis.

Background

Chronic recurrent multifocal osteomyelitis (CRMO) is a rare disorder of autoimmune etiology affecting children and adolescents [1]. It is a non-infective osteomyelitis which commonly affects the metaphysis of the long bones, pelvis, and shoulder girdle, rarely involving the mandible and spine [2]. Lack of specific signs and symptoms on examination and imaging makes it a diagnosis of exclusion. The diagnosis of CRMO is often delayed, sometimes as late as 18 months [3]. As it is more often undiagnosed, it is well understood that true prevalence of the condition is underestimated [4]. There have been very few case reports of isolated, contiguous, and non-contiguous involvement of the spine in the setting of CRMO. We present our experience of a 9-year-old boy who was diagnosed and treated for CRMO with non-contiguous involvement of the spine.

Case Report

A 9-year-old previously healthy boy initially presented to an outside institution with vague abdominal pain, for which he was investigated, and no active intervention was undertaken. There was increasing pain and deformity over the back noted 1 week later. Radiographs of the spine showed sclerotic lesions in D8 and D12 (Fig. 1) with collapse of D8 and a lesion in head of 4th rib. ESR was elevated, while Serum Electrophoresis showed diffuse band in gamma region (Fig. 2). A bone marrow biopsy and a needle biopsy of the D8 lesion revealed no obvious pathology.

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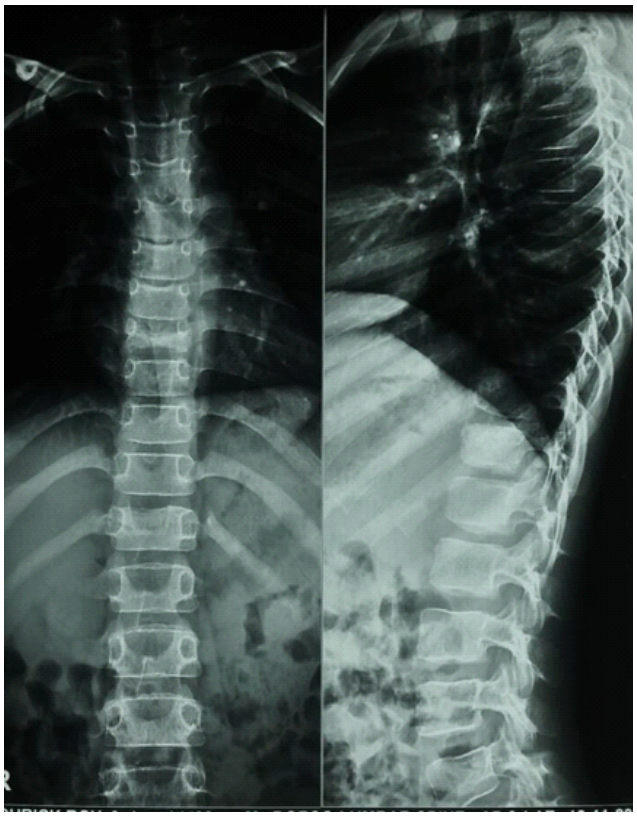


Figure 1: Spine X-rays

The child presented to our institute a few days later with severe pain and deformity over the back. There were no constitutional symptoms. He was ambulating with his hands on the thigh for support. Examination revealed severe para-spinal muscle spasm. Neurological examination of the lower limbs was normal.

Most of the blood tests were within normal limits except for ESR, which was 48 mm/hr. CT guided biopsy of D8 was inconclusive (no cellular atypia/granuloma). Child was started on oral Naproxen and there was symptomatic relief. CT thorax was performed to identify an additional lesion in the rib (Fig. 3) and an open biopsy was undertaken (resection of diseased rib and adjoining transverse process). Biopsy report (Fig. 4) was suggestive of chronic non-infective osteomyelitis (CRMO).

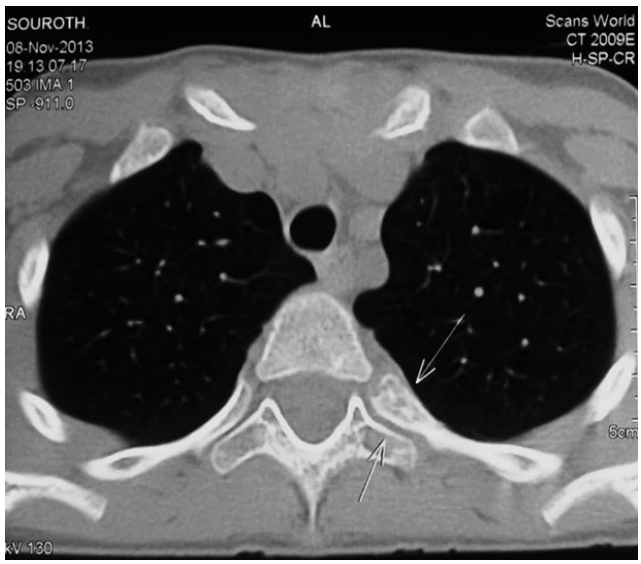


Figure 3: CT Thorax

In view of this diagnosis, a whole-body MRI was done which showed an additional lesion in the right acetabulum (Fig 5). With anti-inflammatory medications, the ESR reduced to 18 m/hr

There was significant relief of symptoms with oral naproxen followed by 5 doses of intravenous zoledronate at yearly intervals. At 15 years of age, there was no pain or deformity in

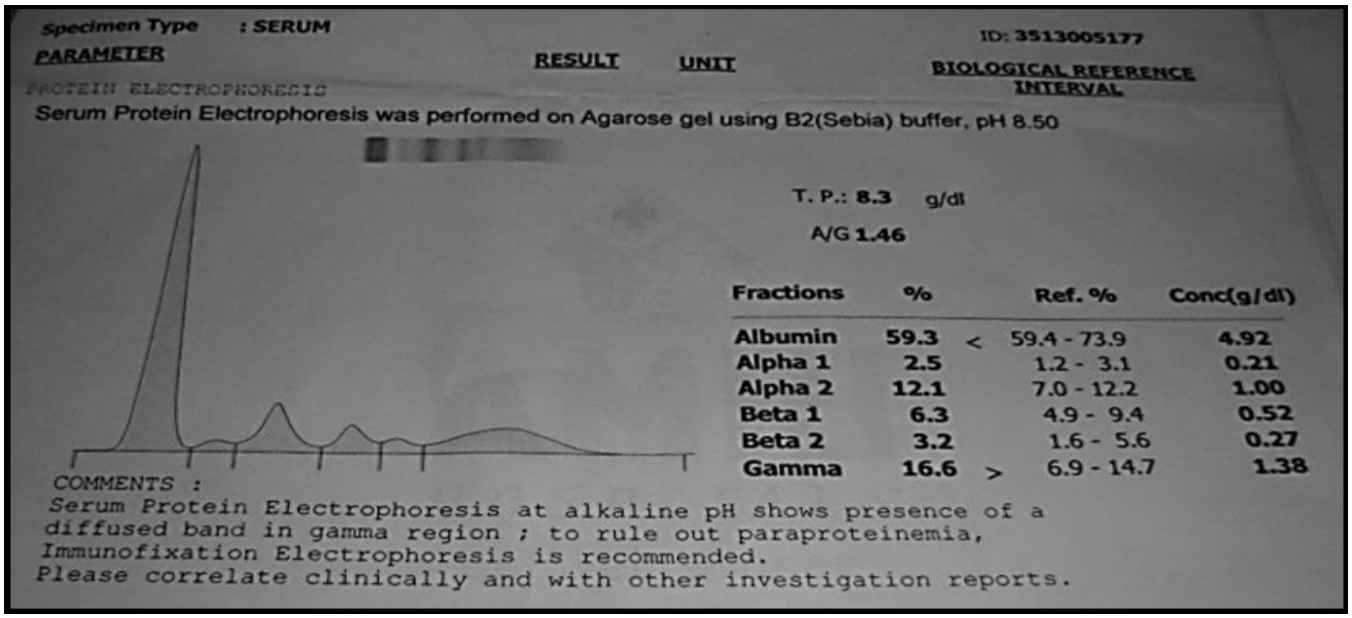
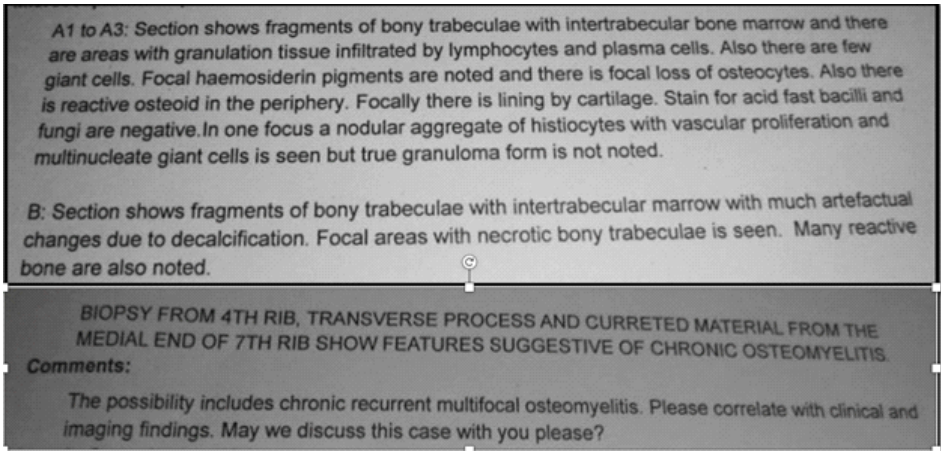


Figure 2: Electrophoresis


A1 to A3: Section shows fragments of bony trabeculae with intertrabecular bone marrow and there are areas with granulation tissue infiltrated by lymphocytes and plasma cells. Also there are few giant cells. Focal haemosiderin pigments are noted and there is focal loss of osteocytes. Also there is reactive osteoid in the periphery. Focally there is lining by cartilage. Stain for acid fast bacilli and fungi are negative.In one focus a nodular aggregate of histiocytes with vascular proliferation and multinucleate giant cells is seen but true granuloma form is not noted.

B: Section shows fragments of bony trabeculae with intertrabecular marrow with much artefactual changes due to decalcification. Focal areas with necrotic bony trabeculae is seen. Many reactive bone are also noted.

BIOPSY FROM 4TH RIB, TRANSVERSE PROCESS AND CURRETED MATERIAL FROM THE MEDIAL END OF 7TH RIB SHOW FEATURES SUGGESTIVE OF CHRONIC OSTEOMYELITIS.

Comments:

The possibility includes chronic recurrent multifocal osteomyelitis. Please correlate with clinical and imaging findings. May we discuss this case with you please?

Figure 4: Biopsy Report

the spine and the vertebral lesion (Fig. 6) had reconstituted. The boy was asymptomatic.

Discussion

CRMO is a rare autoimmune disease of childhood and adolescence and was first described in 1972 by Giedion et al. [5] (Chronic Symmetrical Osteomyelitis). The term CRMO (Chronic Recurrent Multifocal Osteomyelitis) was proposed by Probst et al. [6]. Being very common in the first decade (age group 4-14 years), there is a female predilection [7]. Our case pertains to a 9-year-old boy who presented initially with vague symptoms in the abdomen followed by involvement of axial skeleton.

CRMO can be considered as a paediatric form of synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO) syndrome [8]. Although the exact cause is unknown, it is found to be associated with psoriasis, inflammatory arthritis, inflammatory bowel disease, Majeed syndrome, and Sweet syndrome [9]. It is common to encounter a delay in diagnosis of several months from appearance of first presenting symptoms. In our case, there was a delay of 3 months from the initial presentation to the final diagnosis.

The most common manifestation of CRMO is painful bony lesions mainly affecting the metaphysis of the long bones; tibia being the commonest followed by the distal femur and pelvic bones [10]. Other locations include the humerus, clavicle, and spine. Hospach et al. [11] proposed that spine involvement is up to 37% but rarely diagnosed. In our case, even with repeated attempts of needle biopsies and image guided percutaneous biopsies, the results were inconclusive. An open biopsy with adequate tissue for sampling was necessary to reach the diagnosis

In the available literature, we found a few case reports of isolated and non-contiguous fractures of the spine in setting of CRMO and one with contiguous three-level involvement [12]. A summary of these has been listed in Table 1.

Treatment is usually initiated with non-steroidal anti-inflammatory drugs (NSAIDS), commonly Naproxen with good response [4]. Our experience was similar, but the side effects of long-term NSAIDs preclude their continued use.

The use of bisphosphonates (pamidronate/ zoledronic acid) has been described recently in the literature with promising results [13, 14] although there are no randomized studies. In the present case, zoledronic acid was administered intravenously at regular intervals (yearly), and showed complete resolution of symptoms and reconstitution of the spinal lesion over a period of 5 years.

The Childhood Arthritis and Rheumatology Research Alliance in 2019 had proposed three consensus treatment plans for patients with NSAID-refractory CRMO, including

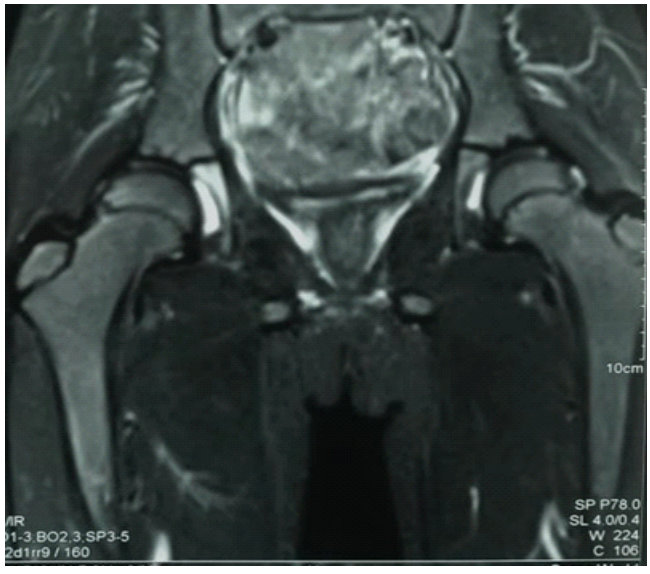


Figure 5: MRI

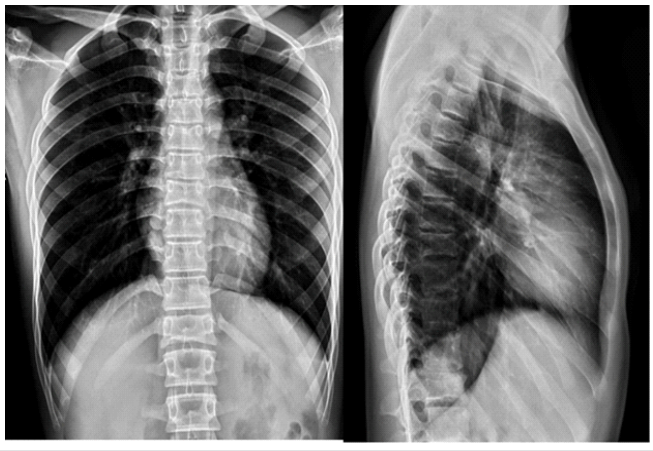


Figure 6: X-ray latest

bisphosphonates and tumour necrosis factor inhibitors (TNFi) [15].

Conclusion

CRMO is a condition of childhood and adolescence. Due to the lack of widely accepted diagnostic criteria or disease biomarkers, CRMO remains a diagnosis of exclusion. Treating clinicians should be familiar with this entity, especially in cases presenting with atypical osteomyelitis, either unifocal or multifocal. Prompt diagnosis of CRMO will allow patients to avoid the risks associated with lengthy courses of antibiotic therapy and repeated bone biopsies.

Table 1: ???

Authors	Year	Number of cases	Site of vertebra
Baulot et al.	1998	1	T2 fracture dislocation
Walls et al.	2006	1	T6 and T7 fracture
Hospach et al.	2010	27 patients had spine involvement, 14 scoliosis, 6 kyphosis, 2 patients had fractures	Site not mentioned
Habibi et al.	2013	1	Cervical and thoracic
Yamashita et al.	2018	1	T4 vertebra plana with fractures of T7 and T11

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Declaration of patient consent : The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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