CASE REPORT

Garre's sclerosing osteomyelitis—with perimandibular soft tissue inflammation and fistula

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Abstract

Garre's sclerosing osteomyelitis is a form of chronic osteomyelitis which commonly affects children and young adults. Here, we report one such case of Garre's sclerosing osteomyelitis in a 20-year-old female who presented with facial asymmetry and inability to open mouth. On clinical examination, it was bony hard swelling with trismus. History of infected second molar tooth extraction was present. Computed tomography (CT) scan showed thickening and sclerosis of the ramus and condylar process of mandible, on right side, with proliferative periostitis. Magnetic resonance imaging (MRI) showed soft tissue edema and inflammation, in the form of enlargement of right masseter and pterygoid muscles with intramuscular fluid collection. On the basis of history, clinical signs and imaging features, diagnosis of Garre's osteomyelitis with fascial space infection was made. To our knowledge, very few cases of Garre's osteomyelitis present with superimposed fascial space infection, as it is otherwise a non-suppurative condition. Fistula formation is a very rare incidence as it is seen in our case.

Key words: CT proliferative periostitis; MRI Garre's osteomyelitis; suppurative osteomyelitis

Introduction

Chronic osteomyelitis with proliferative periostitis (also known as periostitis ossificans or Garré's sclerosing osteomyelitis) is a distinctive form of chronic osteomyelitis.[1] It is commonly associated with odontogenic infections in children and young adults.[1,2] The first case was reported in tibia[2] and Berger described this condition to affect mandible for the first time.[1] The common causative pathogens of this condition are staphylococci, klebsiella and streptococci.[2] The underlying pathology is the periosteal reaction to inflammation and hence, precisely known as chronic osteomyelitis with proliferative periostitis.[1]

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This condition is usually non-suppurative and asymptomatic without signs of inflammation.[2-4] The SAPHO syndrome is characterised by the presence of osteomyelitis in other bones, arthritis and skin diseases.[2] However, rare instances of this condition, resulting in abscess and fistula formation are described in literature.[3,4] We report one such rare case of Garré's sclerosing osteomyelitis associated with unusual surrounding soft tissue inflammation and intra-muscular abscess and fistula to skin.

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Case History

A 20-year-old girl presented with swelling in the right inferior border of the mandible. On clinical examination, diffuse and tender swelling was noted which was hard in consistency. The patient also had severe trismus and unamenable for oral examination. She gave history of toothache before the onset of symptoms. Intra-oral examination showed caries right second molar tooth. Contrast enhanced computed tomography (CECT) was advised. The imaging findings in CECT include swollen and oedematous right masseter and pterygoid muscles [Figure 1]. There was intramuscular fluid collection within the masseter muscle and fistulous communication to skin [Figure 2]. Three dimensional (3-D) CT of mandible showed cortical thinning with periosteal thickening involving ramus and condylar process of mandible, on right side, characteristic of Garré's sclerosing osteomyelitis [Figure 3]. MRI also showed hyper-intensities in right masseter and pterygoid muscles, which implicates inflammation [Figure 4]. Incision and drainage of the collection, followed by biopsy from expanded cortical regions was done and the patient was put on intravenous antibiotics.

Discussion

Garre 's sclerosing osteomyelitis was first described by Carl Garre ̃in 1893 in tibia, which resulted from radiation exposure.[1] However, it was Berger who first described this condition affecting the jaw bones.[1] This chronic form of osteomyelitis is usually asymptomatic without any signs of local inflammation.[2] The common organisms encountered in the disease process include staphylococcus, Klebsiella and streptococcus, resulting in phases of remission and exacerbation.[2] The commonest cause is odontogenic infection but it can also occur in gunshot wounds, fractures, pyoderma, postoperative bone interventions etc.[2] The severity and duration of disease depends of many factors like, the virulence of the causative organisms, the presence of underlying diseases and the immunity of host.[2] It is often unilateral and non-suppurative.[1] However, rare instances of this condition, resulting in abscess and fistula formation are described in literature.[3,4]

Facial asymmetry is often the presenting complaint and pain is not a characteristic finding.[3,5] The other markers of

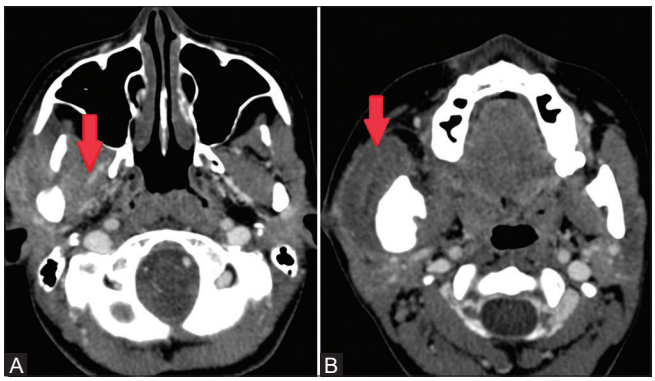

Figure 1 (A and B): (A) CECT (axial section) shows edematous pterygoid muscles on right side (red arrow). (B) CECT (axial sections) show edematous right masseter muscle (red arrow)

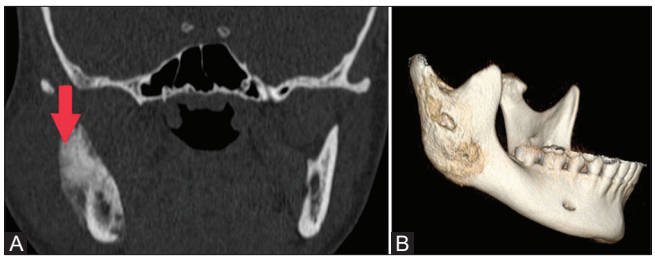

Figure 3: (A and B) (A) Coronal CT reconstruction of mandible show proliferative periostitis (red arrow). (B) 3-D reconstruction of mandible show proliferative periostitis

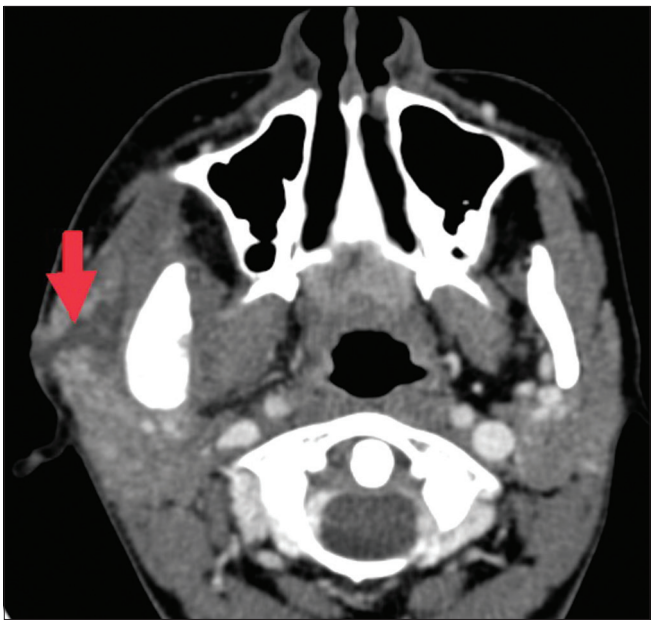

Figure 2: CECT (axial section) shows fluid collection in masseter muscle with fistulous communication to skin (red arrow)

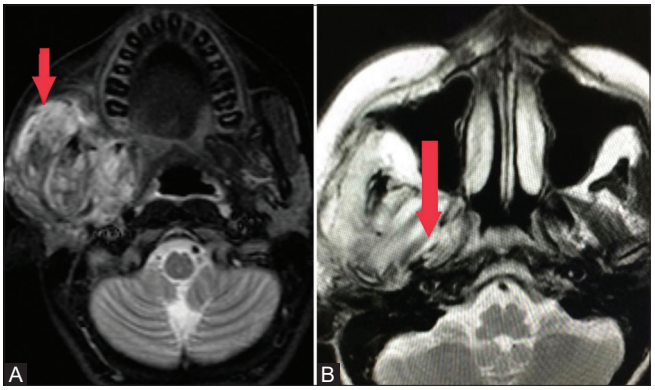

Figure 4 (A and B): (A) MRI (STIR axial section) shows oedematous right masseter muscle (red arrow). (B) MRI (T2 axial section) shows oedematous right pterygoid muscle (red arrow)

acute inflammation like fever, white blood cell count and C-reactive protein may also not elevate characteristically.[5] The disease process starts in the spongiosa and extends into periosteum, resulting in osteoblastic reaction.[3] Unfortunately in some patients the disease process can further extend to the peri-mandibular soft tissue with resultant abscess and fistula formation.[3,4] In such a scenario, there can be severe trismus resulting from the masticator space infection.[4]

Imaging plays an very important role in the diagnosis of this condition.[3] The CT imaging features of periostitis ossificans include cortical thinning and periosteal thickening with lamellar appearance (onion skin), commonly affecting the ramus of mandible.[2,3,6] The laminated appearance is due to modulation of fibroblasts in the adjacent soft tissue, which develop osteoblastic capacity and give rise to sheets of new bone in multiple layers.[7] The differential diagnosis of this type of periostitis includes Caffey disease, Ewing's sarcoma, osteosarcoma, Fibrous dysplasia, osteoma, exostosis and ossifying subperiosteal hematoma.[3,8]

Caffey disease or infantile cortical hyperostosis is a rare self-limiting condition of infancy which is characterised by cortical hyperostosis, particularly affecting the mandible and facial bones.[7] This condition is bilateral and multiple bones are involved unlike Garrè's osteomyelitis.[3] Ewing's sarcoma and osteosarcoma are the two malignant conditions with similar periosteal reaction, although they are very rare in mandible and characterised by 'sun ray' appearance.[3,8] While, the latter is characterised by codman triangle, the former, in addition, shows osteolytic areas and neurological symptoms like facial neuralgia and lip paresthesia.[3]

Fibrous dysplasia is typically characterised by the 'ground glass appearance' and the enlargement is seen in the bone matrix, whereas in Garre's osteomyelitis it is seen on the outer surface of the cortex.[3] The appearance of ossifying subperiosteal hematoma or fracture with callus formation may mimic Garre's osteomyelitis clinically. However, the former does not exhibit uniform radiopacity and display mottled appearance or trabecular structure while the absence of trauma history can exclude both the conditions.[3]

Once the diagnosis of Garré's osteomyelitis is made, the most commonly accepted treatment is the administration of antibiotics and the extraction of the infected tooth.[3] However, when complicated with abscess formation as demonstrated in CECT as peripherally enhancing fluid collection, it requires incision and drainage.[4] MRI is helpful in presuppurative stages, when muscle edema is detected as increased signals within the muscles in the T2 and STIR sequences. It is important to identify the peri-mandibular soft tissue infection, as otherwise conservative therapy and removing the causative factor are usually sufficient for Garre's osteomyelitis.[1]

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their 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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Conflicts of interest

There are no conflicts of interest.

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