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Case Report Open Access

The Case Report of Sclerosing Osteomyelitis Garre of the Mandible

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Abstract

A 21 years old female patient reported with the complaint of face asymmetry, pain, acne and rash on the skin in the mental area of the lower jaw (right side) and became sick at the age 1.5 years old after face trauma. She has experienced 4 low-efficient operations for diagnosis correction. Preoperative examination by 3D CT showed sclerosing focies in mental area of the mandible. On the base of anamnesis, clinical signs (pain, enlarged mandibular bone tissue, skin diseases, and typical radiological picture) were strongly suspected Garre osteomyelitis associated with SAPHO syndrome. Subsequent computer simulation (stereolithographic model, 3D-printed) allowed planning surgery. In January 2018 was held operation with extraoral approach–partial resection and leveling of the sclerosed bone in the area of lesion (according to printed CT scan model), dissection of soft tissue scars after previously performed operations. In 5 days, the patient left hospital in good condition with recommendations for further examination, sanation of odontogenic infection, radiological control. This disease refers to atypical primary chronic forms of osteomyelitis and is rare in clinical practice of maxillofacial surgeons, especially in adults. Treatment consists of surgical and conservative phases. The prognosis is favorable, but syndrome is often persistent.

Keywords: Sclerosing osteomyelitis; Mandible; Diagnosis; Computer tomography; Treatment

Introduction

Garre Syndrome (GS) is a specific type of chronic atypical primary sclerosing osteomyelitis (also known as chronic non-suppurative sclerosing osteomyelitis, chronic osteomyelitis with proliferative periostitis and periostitis ossificans). It was first described by German surgeon Carl Garre in 1893 as irritation, induced focal thickening of periosteum and cortical bone of tibia. Area of jaws affects quite rare, but observing prevalence of mandible [1]. Primarily affects children and adolescents.

Garre Syndrome is one of the most confusing and misinterpreted lesion regarding a typical forms of osteomyelitis [2].

The disease is caused by a conditionally pathogenic microorganisms (most frequent pathogens are *staphylococci*, *klebsiella* and *streptococci*). Microbes penetrate into bone tissue through blood or damaged skin. Commonly associated with an odontogenic infection, but also could developed under risk factors as are: gunshot wounds; the presence of pyoderma; fractures; phlegmons; tuberculosis; chronic inflammation of the tonsils; operative interventions on bones and joints; decreased immunity. The risk group includes long-term starving and weakened people. Penetration of infection contributes stress, poor nutrition, respiratory diseases and hypothermia. In some cases, osteomyelitis develops in newborns. The signs of this pathology are nonspecific. Sclerosing osteomyelitis leads to a deformation of the bones. The disease proceeds with the phases of remission and exacerbation.

Due to Chamot et al. [2], the Garre Syndrome considers as osteomyelitis, associated with the synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO syndrome). When it affects the jaw, this generally originates as an infection of low virulence, such as dental decay, mild periodontitis, dental eruption, previous dental extraction or experienced trauma in the lesion area with proliferative periostitis and periostitis ossificans [1,3] .

Clinicists [3-5] are recognized mandibular osteomyelitis as nonsuppurative inflammatory process, usually asymptomatic with no accompanying general and local signs of inflammation, although the clinical picture may have variety of other symptoms [4,6,7]. In some cases originating as an odontogenic infection, caused reactive process accounts for the hard swelling of the jaw and surrounding tissues [1]. The severity of the symptoms and duration of disease depends of such factors as: the virulence of the causative microorganisms, the presence of underlying diseases and the immune status of the host [1]. Characterized with peripheral subperiosteal bone deposition caused by mild irritation and infection, mixed pattern on radiography, with solid type periosteal reaction, external bone resorption, and bone enlargement. In bone exists areas of mildness with some suppuration and necrosis signs, but, nevertheless, no sequesters and fistulas forms [2,5]. The presence of osteomyelitis in other bones, arthritis, or skin diseases (palmoplantar pustulosis, pustular psoriasis, and acne) points SAPHO syndrome. Antibiotic therapy, as opposed to bacterial form of osteomieleties, is usually ineffective and the symptoms of SAPHO syndrome are often persistent. Surgical intervention is complicated by the fact that in a sclerotized bone it is difficult to detect and eliminate numerous of small osteomyelitis foci, which could lead to a relapse of the disease [3,4].

An erroneous oncological diagnosis is most typical for this disease and is noted in more than 60% of cases. Clinically and Radiographically quite often signs of sarcoma or fibrosis dysplasia simulated, so differential diagnosis of this pathology has to be carried out with osteosarcoma, fibrous dysplasia, bone damage in congenital etc. [1]. Typical Radiographic feature of Garre osteomyelitis (well

known as "onion skin") showing new periosteal proliferation located in successive layers to the condensed cortical bone [1,3,6].

Clinical observation

At the Department of Oral and Maxillofacial Surgery of the Bogomolets National Medical University, patient A, female, 21 years old, complained of periodic pains in the lower jaw area and asymmetry of the lower face area. According to the patient`s story, she is ill from the early childhood (since 1.5 years) after falling and getting facial trauma. In 3-4 monthes after event was arise infiltration and swelling in mental area of lower jaw the right side. In approximately 10-14 days on this location was formed tight infiltrate, which start slowly enlarging and condensiting. The same time arised dermatological symptoms over the area of leision. It was token about lymphadenitis, but after surgical opening of submandibular area was found no enlarge nodes (operation was performed in regional hospital). The patient did not receive a histological confirmation of the diagnosis. In age of 3 years old was performed operation (in kids clinic in Kiev) biopsy and removing the bone tissue in area of lesion. Morphologically was confirmed ossificative periostitis. In 4-6 months "tumor" started growth again and next removing was performed in 2006 (biopsy of enlarged lymph node in submandibular space), than next in 2011(biopsy of bone tissue–due to suspicion osteoma in area of lesion, not confirmed). Since 2014 patient complains on pain in mental area right side of mandibula after stresses and cooling, radiating in other areas of face (left part).

Objectively face asymmetry due to deformation of the bone and soft tissues in mental area and chin of the lower mostly right side; there were atrophic scars in this area from approaches of previous surgical interventions, palpation-compaction of soft-elastic consistency, the skin was slightly hyperemic and blue due to surface rashes and acnes in this area, opening mouth free, the mucous membrane of the oral cavity without visible pathological changes.

In the series of 3D computer tomograms (CT), a spindle-shaped deformation and a thickening of the bone in mental area and the chin of the lower jaw were determined on the right. Cortical plate thickened. Excessive sclerosis was observed (Figure 1). Interpretation of X-ray and CT caused discussion between radiologists and surgeons about such diagnoses as sarcoma, Fibrous dysplasia, and osteomyelitis. Its result and conclusion could significant influence on operating procedure.

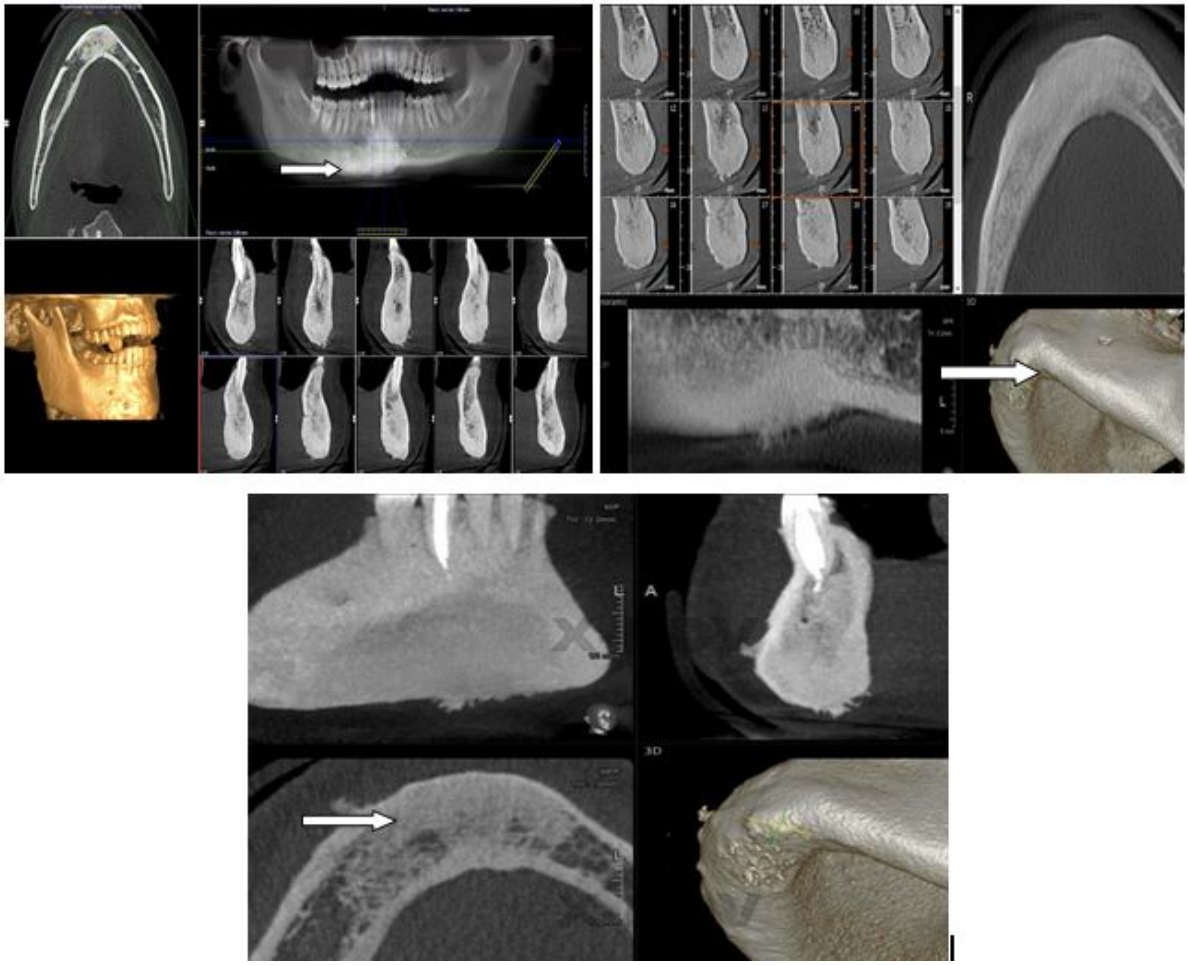

Figure 1: Series of 3D CT scans and computer modeling for surgery.

Presumptive clinical diagnosis of sclerosing osteomyelitis of the mandible

For planning the surgery, the stereolithographic model of the lower jaw was printed by necessary measurements and defined surgical access (Figure 2). On the base of anamnesis, clinical signs (pain, enlarged mandibular bone tissue, skin diseases, and typical radiological picture) were strongly suspected Garre osteomyelitis associated with SAPHO syndrome. The preoperative biopsy was not performed because of such reasons: slow developing of disease (since 1.5 years old) decrease probability of sarcoma, absence of typical clinical symptoms ("cacao spots" on the skin, wrong hormones production etc.) was against Fibrous dysplasia. On January 11/2018, the patient was performed a partial resection and leveling of the sclerosed bone in the chin of the lower jaw with extraoral approach through the old scars, simultaneously dissected (Figures 3 and 4). The bone samples were sent for a pathohistological investigation (Figure 5).

Figure 2: Stereolithographic model (3D-printing) of the lower jaw.

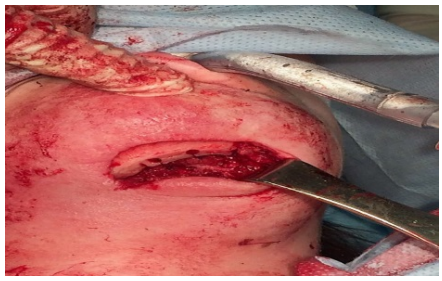

Figure 3: Submandibular approach provided access to the deformating area.

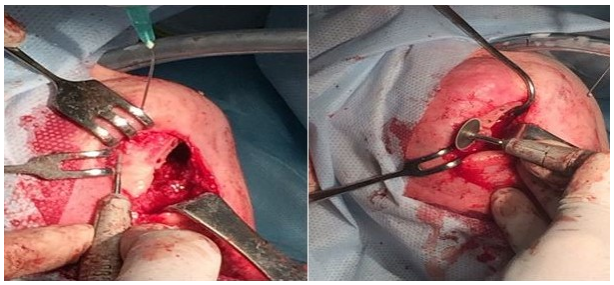

Figure 4: Partially resection and leveling of the sclerosed bone.

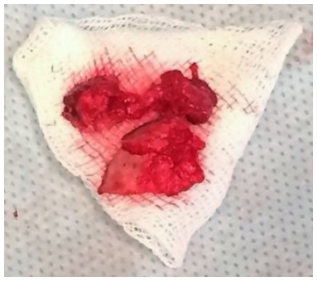

Figure 5: Sclerotized bone fragments (macropreparations).

Postoperative swelling of the submandibular and mental areas lasted for 7 days. The control X-ray showed essential diminishing of lesions` area, quite correct shape of mandibular (Figure 6).

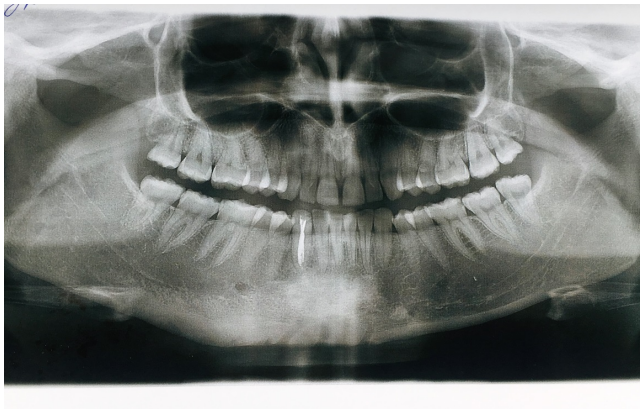

Figure 6: The control X-ray picture after operation.

Conclusion

Fragments of compact bone tissue and parasomal fibrous tissue contain many intestinal foci of osteonecrosis, in some places there are vascular channels with infiltrates, an admixture of neutrophils. This confirmed our diagnosis (Figure 7).

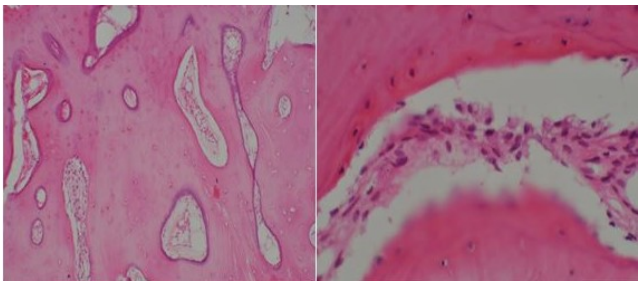

Figure 7: Microphotos of histological preparation, hematoxylin-eosin staining.

On January 17 the patient was discharged in satisfactory condition, further examination and radiological control being recommended.

Discussion

With this form of inflammation of the bone, it is dominated by osteosclerotic processes with gradual desolation of the bone marrow and channel. The disease also runs torpidly, lastingly, with an unusual clinical picture: slight night-time pain in the jaw and during exercise, thickening of the bone, local pain during palpation. Body temperature moderately raised. The diagnosis is based on X-ray data: spindle-shaped thickening of the bone, against which small cavities with small sequesters can be seen. Rarely, the disease begins acutely, with body temperature and intoxication. These phenomena quickly disappear, and the process becomes chronic.

Interpretation of X-ray and CT is complicate due to necessity to exclude sarcoma and Fibrous dysplasia. The treatment is combined and prognosis is usually favorable. Follow-up examinations of patients should be performed after three months, followed by an X-ray examination.

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