



MANAGEMENT OF GARRE'S OSTEOMYELITIS IN A CHILD WITH MARCUS GUNN JAW WINKING SYNDROME - A CASE REPORT

Dr. Ruchi Arora	Professor and Head Department of pediatric and preventive dentistry, Darshan dental college and Hospital, Udaipur
Dr. Sakshi Runwal*	Post graduate, department of pediatric and preventive dentistry, darshan dental college and hospital, Udaipur*Corresponding Author
Dr. Shruti Jain	Post graduate, department of pediatric and preventive dentistry, darshan dental college and hospital, Udaipur
Dr. Neha Kalantri	Post graduate, department of pediatric and preventive dentistry, darshan dental college and hospital, Udaipur

ABSTRACT Garre's osteomyelitis is a local thickening of the periosteum caused by a slight irritation or infection. We aimed to present the extraoral, intraoral, and radiographic findings and management of patient diagnosed with Garre's osteomyelitis. This paper describes a case of Garre's osteomyelitis in a 9-year-old girl with Marcus Gunn jaw winking syndrome with a chief complain of bony hard, tender swelling. The clinical findings indicated infection source and these findings were strongly supported by cone-beam computed tomography images and histological examination which confirmed the diagnosis of Garre's osteomyelitis. Treatment plan included elimination of the cause by extracting left primary second molar, permanent second molar along with surgical bone recontouring and long term antibiotic treatment, followed by endodontic treatment of first permanent molar.

KEYWORDS : Garre's osteomyelitis, Marcus Gunn jaw winking syndrome, recontouring, CBCT.

Introduction

The word "osteomyelitis" originates from the ancient Greek words osteon (bone) and muelinos (marrow) that means infection of medullary portion of the bone, which occurs due to bacteremia, trauma, dental eruption, surgery or surrounding soft tissue infection with a wide variety of presenting signs and symptoms.¹

Most commonly Garre's osteomyelitis presents as facial asymmetry on the lower part of face owing to a solitary swelling with normal to inflamed overlying skin. Orthopantomograph (OPG) reveals a periosteal thickening of the inferior border of the mandible due to increase in osteoblastic activity resulting in an 'onion skin' appearance.⁹ Bone biopsy is usually required to confirm the investigations. Treatment usually consists of antibiotic therapy with or without surgical debridement of the necrotic tissue.¹

Marcus Gunn Jaw winking syndrome (MGJWS) is one of the congenital cranial dysinnervation disorders (CCDD) showing variable degrees of blepharoptosis in the resting, primary position. It is also called as Trigeminal-oculomotor synkinesis. It is associated with synkinetic movements of the upper eyelid during masticating movements of the jaw. It is thought to be due to a congenital miswiring between the branches of the trigeminal nerve supplying the internal or external pterygoids, responsible for mastication, and the branches of the oculomotor nerve supplying the levator palpebrae superioris (LPS) muscle.³ The incidence of Marcus Gunn jaw-winking syndrome (MGJWS) among individuals with congenital ptosis is approximately 2% to 13%.⁴

The research conducted in all major medical databases (PubMed, Scopus, Web of Science) for this article revealed no publications regarding the management of Garre's osteomyelitis in patient with Marcus Gunn jaw winking syndrome.

Case report

A female patient aged 9 years reported to the department of paediatric and preventive dentistry with a complaint of intermittent episodes of pain and swelling in lower left back tooth region since 5-6 months. Pain was dull, moderate, intermittent and localized in nature, which aggravated on chewing and was relieved by taking medications. Patient was taking medication since last 6-7 months (augmentin 375mg, Ibuigeic 200mg). She was diagnosed with Marcus Gunn jaw winking syndrome at the age of 1 year.

Clinical examination revealed facial asymmetry and swelling on the left side of the face measuring upto 4-5 cm in diameter. On palpation

swelling was hard, tender, nonfluctuating and afebrile. Left submandibular lymphadenopathy was noted. In addition, Ptosis of left eye along with movement of the jaw was present representing the features of Marcus Gunn jaw winking syndrome.



Figure 1- extraoral findings (a & b- preoperative photographs, c -post operative photographs)

Intra-oral examination revealed proximal caries i.r.t 53, 54, 63& 64, deep occlusal caries i.r.t 55, 65, 75, occlusal caries i.r.t 74(grade II mobility) & 85, grade III mobile 73 and root piece i.r.t 84.

Pulpectomy was initiated i.r.t to 75 and extraction was done i.r.t 73, 74. In order to rule out the possibility of cross-infection, extraction was done with 65. Patient was recalled after 1 week for follow-up. Patient was inconsistent with her appointments and reported to the department 1 month later with the same swelling. Patient gave history of spontaneous occurrence, progression and regression of the swelling during this course. Informed consent was obtained from parents before and OPG, occlusal radiograph CT Scan and CBCT was carried out.

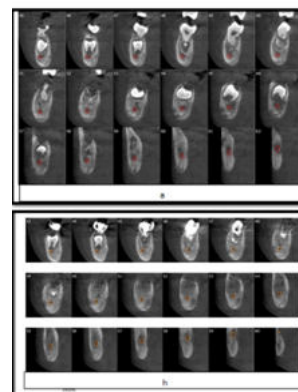


Figure 2a- preoperative CBCT, 2b – postoperative CBCT

Axial section OF CBCT revealed expansion of buccal and lingual cortical plate, reduced bone density/ hypodense along with bone resorption in the left first and second permanent molar. In the coronal section resorption of bone was seen in the periapical region of 36 and 37 including the cortical plate and onion peel appearance seen on the buccal aspect of 37. 3-D view shows moth eaten appearance on the lower border of mandible starting from 36 and extending upto ramus of mandible confirming Garre's osteomyelitis. Large osteolytic lesion was observed around developing left permanent second molar and it has been well reported in the literature that presence of unerupted tooth could be one of the reasons for developing mandibular subperiosteal thickening.

Further, the treatment was carried out under general anaesthesia, which involved extractions with 54, 75, 37, and 84, pulpectomy with 55, root canal treatment i.r.t 36, collection of a biopsy sample from the left side of mandible, and surgical bone recontouring. Histopathological examination revealed fibrovascular soft tissue lined by Odontogenic epithelium with haemorrhage and mononuclear infiltrate. Fragments of calcified and sclerosed bone were also seen with haemorrhage and mononuclear infiltrate.

Culture sensitivity test was negative .Therefore, amoxiclav 375 mg TID and metrogyl 200mg BD was prescribed for 3 months.

After a follow-up of 1 week the swelling considerably reduced (upto 80%) and patient was clinically asymptomatic hence, recalled after three months for follow-up.



Fig. 3 – surgical recontouring and elimination of the causative factor

There were episodes of sudden onset of recurrent progression and regression during the course of lesion for the first 2 months postoperative, since last one month patient is clinically asymptomatic and CBCT shows signs of healing in the cortical plate. Patient is under monthly follow-up for next 6 months.

Discussion

Garre's osteomyelitis is a localized periosteal thickening caused by mild irritation or infection. Although it is sometimes idiopathic, it is known that a moderate infection starting from the spongiosa layer of the jaw and extending into the periosteum, is the result of stimulating bone formation.⁵

It has been noted in the literature that this rare form of bone infection is becoming more and more common. Two reasons could account for this growing incidence:

1. As a result of the increase of health and living standards, people are responding in an "anabolic rather than catabolic manner."
2. The increased use of antibiotics has affected the virulence of microorganisms, turning an osteolytic process into an osteoblastic one. However, the abuse of antibiotics could be harmful to the patient⁶. This could be one of the predisposing factors in our case because the patient was consuming antibiotics since last 5-6 months.

Loveman and Gorman presented number of cases of periostitis ossificans in the mandible where the etiological factor was an erupting/ unerupted permanent second molar covered partially by a gingival flap. The site of entry of infection was thought to be pericoronal⁸.

In this case there was evidence erupting second permanent molar along with osteolytic lesion observed in the coronal section of CBCT around the unerupted tooth. Therefore, it was considered a possible source of infection which was eliminated.

Remodelling of mandible occurs after resolution of the infection, but occasionally, it remains static necessitating surgical recontouring procedures⁹. However, the clinical course usually presents as sudden onset with progression, spontaneous regression and remodeling

Medical history of the patient revealed a rare condition i.e. Marcus Gunn jaw-winking syndrome (MGJWS) which is congenital cranial dysinnervation disorders (CCDD), and these individuals have variable degrees of blepharoptosis in the resting position.⁴

Conclusion

Garre's osteomyelitis is associated with low-grade infection usually arising from an infected tooth that results in stimulation of bone formation. By confirming the diagnosis with CBCT & biopsy and formulating a correct treatment plan, Garre's osteomyelitis can be treated successfully by eliminating the source of infection to restore the impaired balance between the virulent bacteria and the oral flora.

REFERENCES

1. Barbur I, Bran S, Baciut M, Armencea G, Aghiorghiesei AI, Suciut TS, Barbur AM, Opris H, Baciut G, Dinu C. An Orthodontic Approach for Garre's Sclerosing Osteomyelitis of the Mandible. *International Journal of Environmental Research and Public Health*. 2021 Mar 18;18(6):3159.
2. Momodu II, Savaliya V. Osteomyelitis. *InStatPearls* 2021 Aug 11. StatPearls Publishing.
3. Alam MS, Nishanth S, Ramasubramanian S, Swaminathan M, Mukherjee B. The rare phenomenon of Marcus-Gunn jaw winking without ptosis: Report of 14 cases and review of the literature. *Indian Journal of Ophthalmology*. 2020 Jun;68(6):1132.
4. Senthilkumar VA, Tripathy K. Marcus Gunn Jaw Winking Syndrome. *InStatPearls* 2021 Feb 14. StatPearls Publishing.
5. Akgül HM, Çağlayan F, Günen Yılmaz S, Derindağ G. Garre's osteomyelitis of the mandible caused by infected tooth. *Case Reports in Dentistry*. 2018 Jul 8;2018.
6. Schwartz S, Pham H. Garre's osteomyelitis: a case report. *Pediatr Dent*. 1981 Sep 1;3(3):283-6.
7. Asgary S, Eghbal MJ, Ehsani S, Eslami B. An unusual finding of proliferative periostitis in a child: a case report. *International Journal of Stomatological Research*. 2012;1(1):1-5.
8. Oulis C, Berdousis E, Vadiakas G, Goumenos G. Garre's osteomyelitis of an unusual origin in a 8-year-old child. A case report. *International Journal of Paediatric Dentistry*. 2000 Sep;10(3):240-4.
9. Philip LM, Akkara F, Khwaja T, Narayan T, Kamath AG, Jose NP. Surgical management of Garre's osteomyelitis in an 8-year-old child. *Afr J Paediatr Surg* 2021;18:111-3.