ODOVTOS International Journal of Dental Sciences

https://revistas.ucr.ac.cr/index.php/Odontos | ISSN: 2215-3411

CASE REPORT

DOI: 10.15517/IJDS.2022.53064

Received: 23-IX-2022 Acute Osteomyelitis in Torus of Systemically Compromised Patients: Case Series

Accepted: 27-X-2022

Published Online: 4-XI-2022

nline: Osteomielitis aguda en torus de pacientes sistémicamente comprometidos: serie de casos

Yadira V. Boza-Oreamuno DDS, MSc1; Andrea López-Soto DDS, MSc2

 Facultad de Odontología, Universidad of Costa Rica, San José, Costa Rica. https://orcid.org/0000-0002-0367-8664
 Facultad de Odontología, Universidad Latinoamericana de Ciencia y Tecnología, San José, Costa Rica. https://orcid.org/0000-0003-2707-9671

Correspondence to: Dra. Yadira V. Boza-Oreamuno - yadira.boza@ucr.ac.cr

ABSTRACT: The torus are benign bony prominences that generally do not require treatment, however, they can occasionally present osteomyelitis (OM). The objective of this article is to describe the timely management of torus osteomyelitis through a series of clinical cases. Three patients with OM are reported. Two cases of men aged 66 and 69, diabetics, as well as a 57-year-old woman with a history of basal cell carcinoma of the nose. All had torus ulcers associated with trauma and without resolution of the same. In the three cases, a biopsy was performed to confirm the diagnosis and they were treated with anti-inflammatory drugs and antibiotic therapy, achieving complete resolution. Uncontrolled diabetes mellitus and trauma can favor the development of OM in patients with bone exostoses, therefore, when there are signs of infection in these structures, it is imperative to suspect the condition and offer treatment.

KEY WORDS: Osteomyelitis; Exostoses; Mellitus diabetes; Trauma.

RESUMEN: Los torus son prominencias óseas benignas que generalmente no requieren tratamiento, sin embargo, en ocasiones pueden presentar osteomielitis (OM). El objetivo de este artículo es describir por medio de una serie de casos clínicos el manejo oportuno de osteomielitis en torus. Se informa de tres pacientes con OM. Dos casos de masculinos de 66 y 69 años, diabéticos, así como una mujer de 57 años con antecedente de carcinoma basocelular de nariz. Todos presentaban úlceras en los torus asociadas a traumas y sin resolución de las mismas. En los tres casos se

realizó biopsia para confirmar el diagnóstico y fueron tratados con antiinflamatorios y antibioticoterapia, logrando la resolución completa. La diabetes mellitus no controlada y el trauma pueden favorecer el desarrollo de OM en pacientes con exostosis óseas, por lo tanto, ante signos de infección en estas estructuras es imperativo sospechar de la condición y ofrecer tratamiento.

PALABRAS CLAVE: Osteomielitis; Exostosis; Diabetes mellitus; Trauma.

INTRODUCTION

Oral exostoses are benign, non-pathological bony prominences located on the alveolar surfaces, covered by mucosa of the same color as the surrounding tissues (1). An exact etiology has not been identified, it is believed to be multifactorial, and includes mainly genetic and environmental factors (2).

Its name varies according to its location, they are called torus palatinus (TP) when it is located along the midline of the hard palate, torus mandibularis (TM) if it is on the lingual surface of the mandible, and oral exostosis along the face buccal of the maxilla or mandible (3).

The prevalence of torus varies depending on race, ethnic group, population, and sample, reporting between 12.3% and 14.6%, with PD being more frequent than MT (4). PT has been reported to be more common in women and TM in men (1).

Radiographs reveal radiopaque images with a slightly higher density than surrounding bone (4), and histopathologic examination reveals hyperplastic bone, consisting of mature cortical and trabecular bone (5).

They are usually an asymptomatic clinical finding that normally does not require treatment (2), however, they can occasionally ulcerate and present osteomyelitis (OM). OM is an inflammation of the bone that begins as an infection of the medullary cavity with rapid involvement of the Haversian systems and extension to the periosteum (6). The main histopathological lesion of OM is sequestration, which consists of the formation of necrotic bone, which is found in the middle of the infected tissue (7).

There are many contributing factors that predispose a patient to develop OM, including age, trauma, diabetes mellitus, peripheral vascular disease, intravenous drug use, surgical implants, and immunodeficiency due to immunosuppressive diseases or medications (8). Antimicrobial therapy and surgical debridement are the main treatment modalities (9).

The objective of this work is to describe, through a series of clinical cases, the timely management of OM in TP and TM.

CASE REPORT

CASE 1

Male patient, 66 years old, married, retired. It presents with an ulcer in the mandibular torus. On direct questioning, he reported a systemic history: hypertension, diabetes mellitus, and high low-density lipoprotein (LDL) levels; current treatment with Irbesartan 150mg (2 in the morning), Metformin hydrochloride 500mg (1 a day) and lovastatin 20mg once a night. He does not smoke or drink alcoholic beverages.

When performing the clinical examination, large bilobed TMs were observed on the lingual

surface of the mandible from the incisors to the first molar region, both almost in contact in the midline, leaving a small space for the lingual frenulum to pass, covered by colored normal mucosa, except the right ulcerated towards the medial with bone sequestration, and towards the first premolar tumor lesion with pedunculated base, rounded, asymptomatic (Figure 1.A-B), the patient reports evolution of the ulcer for 3 weeks, it was injured when eating. In the neck at right level III, he presented a 2cm adenopathy, ovoid type and painful on palpation, evolution time two days. No facial contour or skin alterations were observed. The occlusal radiograph revealed a radiopaque image compatible with bilateral TM with defined borders on the left and right sides, with irregularity in the outermost contour (Figure 1.C).

Considering the findings, the clinical diagnosis of OM vs. squamous cell carcinoma is established. Ultrasound (US) of the neck with fine-needle aspiration (FNA) biopsy, laboratory tests: complete blood count, blood glucose, and coagulation times were sent. Results of the neck US presence of a level III adenopathy measuring 22mm, with absence of hilum and FNA negative for malignant cells, compatible with reactive adenopathy. Blood tests within normal limits, except fasting glucose 126mg/dL.

Surgical treatment consisted of curettage, removal of granulation tissue, sequestrectomy, bone regularization and closure of the surgical wound by secondary intention. The samples were sent to pathological anatomy and microbiology. Enantyum® 25mg (Dexketoprofen) one tablet every 8 hours for 3 days, Augmentin® 875mg/125mg (amoxicillin/clavulanic acid) one tablet every 12 hours for 7 days, PerioKin® mouth spray (chlorhexidine digluconate 0.20%) 3 sprays on the surface 3 times a day for 10 days.

The histopathological study showed squamous epithelium with regenerative changes, in the stroma fibrosis with chronic inflammation, in the center fragment of necrotic bone tissue and inflammation without data of malignancy (Figure 1.D), the microbiological study was positive for Staphylococcus aureus, diagnosis of OM. The medication was maintained and followed up, reporting feeling good and even more comfortable with the TM to move the tongue (Figure 2).



Figure 1. A-B. Bilateral mandibular torus, ulcerated in the right medial part. C. Occlusal X-ray shows a hyperdense image compatible with bilateral mandibular torus with defined borders on the left and right sides with irregularity of the outermost contour. D. Stroma with dense inflammatory infiltrate of polymorphonuclear cells and lymphocytes (H&E, 20X).

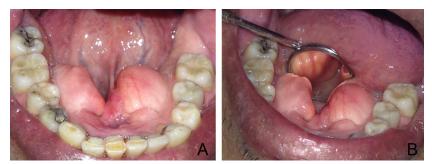


Figure 2. Control at three months, an area of the lesion with a similar appearance to the surrounding tissues is observed.

CASE 2

Female patient, 57 years old, married, housewife. She presents with the main complaint that she burned her palate 10 days ago; she has used hyaluronic acid but it does not improve and she indicated that she has bad breath. On direct questioning, he reported systemic history: basal cell carcinoma of the nose 2 years ago, current hormone replacement therapy with Angeliq® (1mg estradiol hemihydrate/2mg drospirenone), in

addition to Tecta® 40mg (pantoprazole). She does not smoke or drink alcoholic beverages.

When performing the clinical examination, lobulated TP was observed, asymmetrical with an ulcerated area on the left side of the midline (Figure 3.A), she refers that more than oral pain, what she has is discomfort when eating and swallowing. On palpation, it manifests a stinging sensation in the ulcer and pressure in the rest. Halitosis is not appreciated. At the level of the neck, no palpable lymph nodes were found. No facial contour or skin alterations were observed. She has performed oral sex with only one partner. In the occlusal radiograph, a central radiopacity compatible with TP was observed, but no bone lesion was observed (Figure 3.B).

Considering the findings, clinical diagnosis of trauma/infectious ulcer is established. Laboratory tests were conducted: complete blood count, VDRL (Venereal Disease Research Laboratory), blood glucose and coagulation times. In addition, a bland diet and application of Oddent® Periodontal gel (chlorhexidine digluconate 0.20%) on the surface 3 times a day for 5 days.

At control, the results were negative for VDRL and the rest of the blood tests were within normal limits, but clinically the lesion did not improve, it was more erythematous and enlarged (Figure 3.C). OM is suspected, so surgical treatment is performed with curettage, removal of granulation tissue, sequestrectomy, bone regularization and closure of the surgical wound by secondary intention. The samples were sent to pathological anatomy and microbiology. She was given Enantyum® 25mg (Dexketoprofen) one tablet every 8 hours for 3 days, Augmentin® 875mg/125mg (amoxicillin/ clavulanic acid) one tablet every 12 hours for 7 days and continued with Oddent® Periodontal gel.

The histopathological study showed squamous epithelium with mild regular hyperplasia, without evidence of atypia or dysplastic changes, underlying, there was necrotic bone spicule surrounded by fibroconnective tissue with chronic inflammatory infiltrate, no malignant neoplastic lesion was observed (Figure 3.D). The microbiological study was positive for Staphylococcus aureus, diagnosis of OM.

She has been followed up for two years, referring to feeling good and comfortable with the TP (Figure 4).

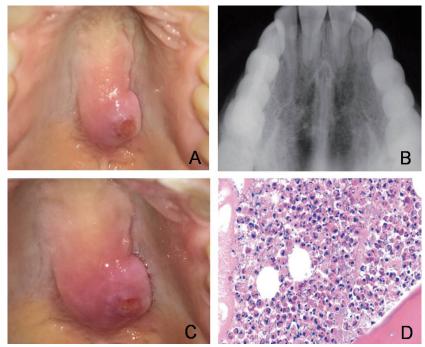


Figure 3. A. Ulcerated palatine torus 10 days after trauma. B. Occlusal radiography without bony lesions, confirming the presence of the palatine torus. C. Increase in size of the lesion at 15 days, prior to surgical removal. D. Necrotic bone spicule surrounded by fibroconnective tissue with inflammatory infiltrate (H&E, 20X).

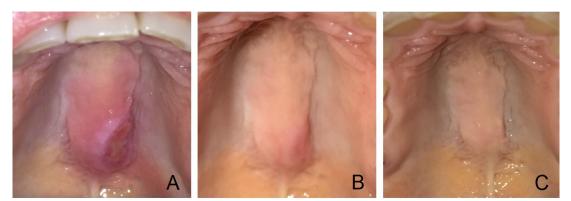


Figure 4. Controls after surgical removal A. At 15 days. B. Per month. C. At two years.

CASE 3

Male patient, 69 years old, married, retired. In the direct questioning, he reported a systemic history of diabetes mellitus in treatment with irregular adherence to it. At 22 days, when presenting for the removable partial prosthesis control, he reported slight discomfort and its non-use for 2 days.

When performing the clinical examination, an ulcer was observed in the left TM, with erythema, edema and pain on palpation, normal bone consistency (Figure 5.A-B). It is verified that he cannot use the prosthesis. At neck level, no palpable lymph nodes were found. No facial contour or skin alterations were observed. The periapical radiograph shows slight bone erosion distal to the lower left second premolar (Figure 5.C).

Considering the findings, the clinical diagnosis of OM is established. Laboratory tests were conducted: glycosylated hemoglobin, complete blood count and coagulation times; being the first of 11%, the other results within the norm. A biopsy was performed. The histopathological study showed an extensive mixed inflammatory infiltrate, with the presence of plasma cells and abundant numbers of neutrophils that form abscesses, accompanied by bacterial lumps, granulation tissue, and abundant fibrin. Inflammation is present in the stroma and extends to the epithelium, which has spongiosis and ulceration, with reactive epithelial changes. No evidence of dysplasia or neoplasia in the sample. There is no bone tissue in the sample. Diagnosis abscess and granulation tissue (Figure 5.D).

Treatment was prescribed with Mopen® Plus 875mg/125mg (amoxicillin/clavulanic acid) one tablet every 12 hours for 7 days, Zamen® 30mg (Deflazacort) once a day for 3 days, Enantyum® 25mg (Dexketoprofen) one tablet every 8 hours for 3 days and Oddent® Periodontal gel (chlorhexidine digluconate 0.20%) on the surface 3 times a day for 10 days.

He has been followed up for 2 months, currently he reports comfort, and he resumed using the lower prosthesis without problem (Figure 6).

The patients gave their free and informed consent in writing for the publication of their conditions.



Figure 5. A.B. Ulcerated mandibular torus 22 days after delivery of the lower prosthesis. C. Periapical X-ray shows slight bone erosion distal to the lower left second premolar. D. Extensive mixed inflammatory infiltrate, with the presence of plasma cells and abundant numbers of neutrophils that form abscesses (H&E, 20X).



Figure 6. Controls after surgical removal A. At 15 days. B. Per month. C. At two months, with total healing of the torus and use of the prosthesis without problem.

DISCUSSION

Patients may experience trauma or ulceration to the tori when chewing hard, sharp foods (2,3). Most cases of OM occur after bone trauma, bone surgery or secondary to vascular insufficiency (9). Therefore, it is extremely important to consider OM as a possible cause when persistent ulcerations are observed in the torus.

There is a lot of variety in terms of shapes; TP can be flat, nodular, lobular or fusiform, TM is usually nodular, unilateral or bilateral and single or multiple (4). The size of the TP is highly variable, ranging from a small pea to a huge enlargement that can cover the entire palate, the TM can also show variation in size, but is generally small in size (2). Reported cases 2 and 3 were medium, case 1 is out of the ordinary, since the TM occupied a good part of the space towards the floor of the mouth, in addition to the size of both cases of the TM they were multilobed, therefore, the possibility from trauma was older.

The diagnosis of OM frequently becomes a challenge since there are great variations in the clinical presentation (8). Acute OM involves a progression of days to a few weeks and is based on acute symptoms, such as fever, leukocytosis, lymphadenopathy, and inflammation of the affected area (9). Plain radiography has low sensitivity and specificity for the diagnosis of acute OM, as it can take up to 2 weeks for bone loss to become radiographically apparent after infection, bone marrow edema, which is the pathologic hallmark, is acquired. earlier, it is not visible on plain radiographs (10). Therefore, it is very important to rely on a good clinical history and physical examination, using laboratory studies and imaging methods as a complement. As observed in the clinical cases of this report for the diagnosis of acute OM.

The prevalence of OM in patients with diabetes varies between 10% and 68% (11). OM tends to occur more commonly in the mandible than in the maxilla, the precarious terminal vasculature, the cortico-spongy structure, and the low mucosal coverage are arguments in favor of the mandible being the preferred site of OM (12). Coinciding with case 1 and 3 where both patients were uncontrolled diabetics and presented OM in the TM. However, a serious complication in patients treated with antiresorptive and antiangiogenic agents, as well as other recently developed drugs (alpha tumor necrosis factor) that can cause similar intraoral symptoms is oral osteonecrosis (13). Therefore, it is important to include it in the differential diagnosis of this condition.

Antibiotic therapy is the treatment of first choice for OM, regardless of the site of infection (7). Most of the germs found in OM are sensitive to the combination of amoxicillin and clavulanic acid or clindamycin (12).

The removal of the torus is not always necessary, the most frequent cause of removal

continues to be the need for prosthetic treatment (4) or as a filling biomaterial, as it is a potential source of autogenous cortical bone for grafts in oral surgery (2,4.5). However, for OM at first, the infection must be eradicated with systemic antibiotic therapy, aggressive bone, and soft tissue debridement, which is consistent with the three cases described, where the patients had not had their torus removed until the OM had to be regularized as part of the treatment.

CONCLUSIONS

It is important for general dentists to be aware that the consumption of antiresorptives is not the only thing that can cause OM/osteonecrosis, uncontrolled diabetes mellitus and trauma can favor the development of this pathology. It is imperative to suspect the diagnosis early and offer antibiotic treatment. Optimal glycemic control in diabetics is mandatory to prevent such infections.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTION STATEMENT

Conceptualization and design: Y.V.B.O. and A.L.S. Literature review: Y.V.B.O. and A.L.S. Methodology and validation: Y.V.B.O. and A.L.S. Formal analysis: Y.V.B.O. and A.L.S. Research and data collection: Y.V.B.O. and A.L.S. Resources: Not applicable. Data analysis and interpretation: Y.V.B.O. and A.L.S. Writting-preparation of the original draft: Y.V.B.O. and A.L.S. Writting-reviewing and editing: Y.V.B.O. and A.L.S. Supervision: Y.V.B.O. Project management: Not applicable. Acquisition of funds: Not applicable.

REFERENCES

- Scrieciu M., Mercuţ V., Mercuţ R., Bîrjovanu C., Stan M.C., Marinescu I.R., et al. Morphological and clinical characteristics of the torus palatinus and torus mandibularis in a sample of young and adults' Romanian people. Rom J Morphol Embryol. 2016; 57 (1): 139-44.
- 2. AlZarea B.K. Prevalence and pattern of torus palatinus and torus mandibularis among edentulous patients of Saudi Arabia. Clin Interv Aging. 2016; 11: 209-13.
- 3. Isha M., Ramaswami E. Bony Exostoses : Case Series and Review of Literature. Acta Sci Dent Sci. 2018; 2 (10): 64-7.
- García-García A.S., Martínez-González J.M., Gómez-Font R., Soto-Rivadeneira Á., Oviedo-Roldán L. Current status of the torus palatinus and torus mandibularis. Med Oral Patol Oral Cir Bucal. 2010; 15 (2): e353-60.
- Smitha K., Smitha G.P. Alveolar exostosis

 revisited: A narrative review of the literature. Saudi J Dent Res [Internet]. 2015; 6
 (1): 67-72. Available from: http://dx.doi. org/10.1016/j.sjdr.2014.02.001
- Habib A., Sivaji N., Ashraf T. Maxillary Osteomyelitis: A Rare Entity. Case Rep Otolaryngol. 2016; 2016: 1-3.
- Maffulli N., Papalia R., Zampogna B., Torre G., Albo E., Denaro V. The management of osteomyelitis in the adult. Surgeon [Internet].

2016; 14 (6): 345-60. Available from: http:// dx.doi.org/10.1016/j.surge.2015.12.005

- Kavanagh N., Ryan E.J., Widaa A., Sexton G., Fennell J., O'Rourke S., et al. Staphylococcal Osteomyelitis: Disease Progression, Treatment Challenges, and Future Directions. Clin Microbiol Rev. 2018; 31 (2): e00084-17.
- Dym H., Zeidan J. Microbiology of Acute and Chronic Osteomyelitis and Antibiotic Treatment. Dent Clin North Am [Internet]. 2017; 61 (2): 271-82. Available from: http:// dx.doi.org/10.1016/j.cden.2016.12.001
- Lee Y.J., Sadigh S., Mankad K., Kapse N., Rajeswaran G. The imaging of osteomyelitis. Quant Imaging Med Surg. 2016; 6 (2): 184-98.
- Xing K., Huang G., Hua S., Xu G., Li M. Systematic review of randomized controlled trials on antibiotic treatment for osteomyelitis in diabetes. Diabet Med. 2019; 36 (5): 546-56.
- Andre C.V., Khonsari R.H., Ernenwein D., Goudot P., Ruhin B. Osteomyelitis of the jaws: A retrospective series of 40 patients. J Stomatol Oral Maxillofac Surg [Internet]. 2017; 118 (5): 261-4. Available from: http:// dx.doi.org/10.1016/j.jormas.2017.04.007
- Sacco R., Shah S., Leeson R., Moraschini V., de Almeida Barros Mourão C.F., Akintola O., et al. Osteonecrosis and osteomyelitis of the jaw associated with tumour necrosis factor-alpha (TNF-α) inhibitors: a systematic review. Br J Oral Maxillofac Surg. 2020; 58 (1): 25-33.

