KIN INTERNATIONAL AWARDS
2ND INTERNATIONAL COMPETITION OF CLINICAL CASES IN PERIODONTOLOGY AND SURGERY

UNUSUAL PRESENTATION OF AN AGGRESSIVE RESIDUAL ODONTOGENIC CYST IN THE MAXILLA AND MUCOUS PEMPHIGUS IN THE SAME PATIENT. CASE REPORT

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Abstract

A very unique case is presented of a 58-year-old female patient with a history of Chagas disease with risky habits of chewing tobacco with coca leaves and a chronic smoker, who attended a private practice of oral and maxillofacial surgery where she was diagnosed and treated for an aggressive residual odontogenic cyst in the upper jaw and pemphigoid of the oral mucosa. After 6 months of control with a cone beam tomography, there is no evidence of recurrence of the bone lesion and there is evidence of adequate healing of the surgical wound after the use of 0.20% chlorhexidine digluconate gel with 1% hyaluronic acid although there was suffering in the healing of the flap, regarding the treatment of mucosal lesions due to pemphigus, they were initially treated with topical corticosteroids and are currently being treated with systemic corticosteroids, achieving partial remission of the lesions.

Introduction

The Residual Cyst corresponds to 10% of the Odontogenic cysts of inflammatory origin. It is defined as a cyst that remains in the maxillary bone after the completion of an exodontia. Generally it is asymptomatic, when it reaches large sizes it causes pressure effects and if it becomes infected it can generate pain. Radiologically a well-defined unilocular radiolucent image with radiopaque corticalized borders is evident. Cysts may degenerate over time and may lead to radiopaque masses (dystrophic calcification) within the cystic cavity. Treatment should be surgical by enucleation and curettage.

Oral mucosal pemphigoid is a chronic autoimmune disease affecting the oral, ocular, nasopharyngeal, laryngeal mucosa and may cause genital and cutaneous lesions. They manifest as subepithelial blistering lesions, microscopically characterized by linear deposition of immunoglobulins (mainly IgG or C3) along the epithelial basement membrane. The oral cavity has been described as the only site of involvement or as the first manifestation of the disease. There is no gold standard therapy for treatment, in case of mild disease presentation it can be treated with topical or intralesional corticosteroids. For severe cases, systemic immunosuppressive drugs such as corticosteroids, cyclophosphamide, azathioprine sodium, mitomycin, interferon alfa-2b,12 and methotrexate, among others, can be administered. The response to these treatments is variable and many of them are associated with toxic effects.

Chagas disease caused by the protozoan Trypanosoma cruzi can be transmitted in different ways; by an insect vector, from mother to child, by blood transfusions or by ingestion of contaminated food. Bolivia, along with Argentina and Paraguay, is one of the countries with the highest number of cases acquired by vector transmission in the world. The presence of this disease is less frequent in Europe, the United States, Oceania and Asia. The disease has an acute and a chronic phase, with one of the main clinical manifestations being Chagas heart disease characterized by rhythm disorders, areas of fibrosis and cardiac dilatation. Diagnostic tests in the acute phase are: peripheral blood, analysis of fluids such as cerebrospinal fluid, in the chronic phase the diagnosis should be by serology test in the detection of IgG anti-T. cruzi antibodies and also by polymerase chain reaction (PCR). Treatment is by benznidazole and nifurtimox, which have variable treatment success

depending on the stage of disease.
Hyaluronic acid combined with chlorhexidine digluconate topically has recently been recognized as an adjuvant treatment for chronic inflammatory disease, in addition to its use to improve healing after minor oral surgeries.

Anamnesis

58-year-old female patient of African descent from Chulumani, province of La Paz, Bolivia, with a history of uncontrolled Chagas disease, secondary cardiomegaly, sinus bradycardia, mild anemia. Harmful habits of chewing tobacco and coca leaf, and smoking a pack of cigarettes a week for more than 20 years. The patient comes to the consultation referring an increase in volume in the anterosuperior sector of the upper jaw and requires treatment. On extraoral examination there is evidence of a tumor measuring approximately 2 x 1.5 x 0.5 cm at the level of the upper lip, causing the elevation of the left wing of the nose, with a soft consistency, slightly painful on palpation. Intraoral evaluation shows preserved oral opening, oral mucosa hydrated, a tumor of 2 x 2 x 0.5 cm at the level of the anterosuperior sector that conditions the disappearance of the bottom of the vestibule and extends towards the palate, soft consistency, with crackling areas, defined edges, normal color gum covering, at the central level there is evidence of a non-active fistula, in addition to multiple carious lesions and presence of dental calculus. On the lateral edges of the tongue there are multiple whitish macules with irregular edges that do not detach when scraped, there are also multiple blackish macules located on the floor of the mouth and bilateral cheek.

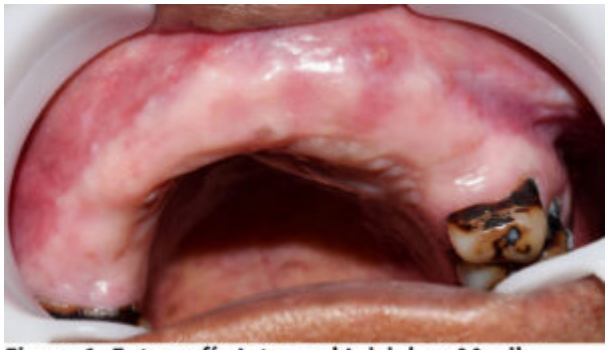

Figura 1: Fotografía Intraoral Inicial en Maxila

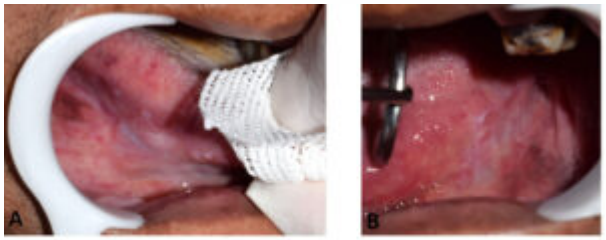

Figura 2: A)Borde lateral de lengua derecho, B) Borde lateral de lengua izquierdo

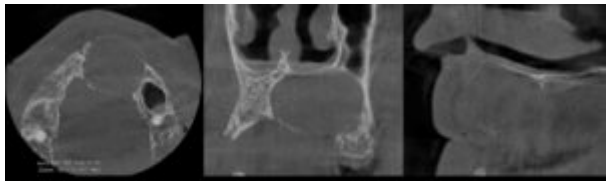

Figura 3: Corte axial , corte coronal y corte sagital de tomografía de haz cónico inicial.

Treatment plan

With the presumptive diagnosis of residual cyst, squamous cell carcinoma on the lateral edges of the tongue and oral melanosis, an interdisciplinary management was performed with cardiology and internal medicine, to subsequently perform enucleation plus curettage of the lesion in the maxilla and incisional biopsies in the lateral edges of the tongue under general anesthesia.

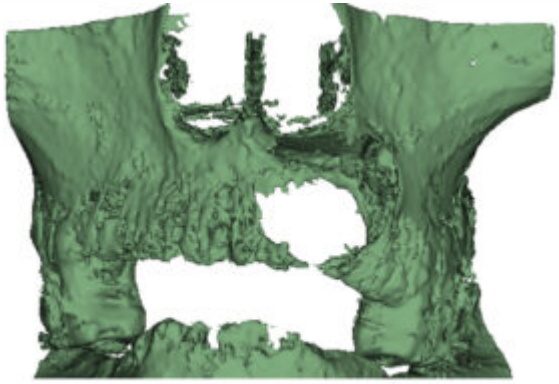

Figura 4: Reconstrucción Volumétrica 3D inicial.

Carrying out the treatment

The day before surgery, the periodontics service performed a tartrectomy and started chlorhexidine digluconate 0.12% mouthwash every 8 hours. First surgical time, under general anesthesia in the operating room was performed the infiltration of lidocaine 2% with epinephrine 1:100000, Newman type incision in the upper jaw, mucoperiosteal decolletage, the nasopalatine nerve was dissected to have a better access, cleavage point was found and the enucleation and curettage itself was performed respecting the left maxillary sinus and nasal mucosa, then hemostasis and suture with 4/0 Vicryl thread was performed. Second surgical time, losange incisions were made in both lateral edges of the tongue in the most representative areas taking affected tissue and healthy tissue, then hemostasis and suture with 3/0 Vicryl thread was performed. Both samples were sent for histopathological analysis. The patient was discharged with amoxicillin 875mg + Clavulanic acid 175 mg 1 tablet cd/12 hours for 7 days, etoricoxib 120 mg 1tab cd/24 hours for 3 days, chlorhexidine digluconate mouthwash 0.12% cd/8 hours for 14 days and application of 0.20% chlorhexidine digluconate gel with hyaluronic acid 1% in the operative wound every 8 hours for 14 days.

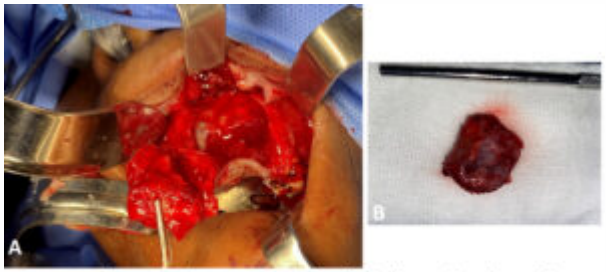

Figura 5: A)Imagen intraoperatoria de lecho quirurgico, B) Aspecto macroscópico de pieza quirúrgica

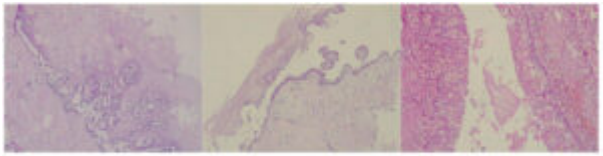

Figura 6: Cortes Histopatológicos de muestras de maxila y bordes laterales de lengua.

Follow up

At 7 days postoperative there was an area of suffering of dark purplish coloration in the central vestibular part of the flap, in its periphery there were edges with erythematous areas with signs of vascularization, later at 14 days there was a partial regeneration of the operative wound without areas of necrosis, at 21 days there was a total healing with an adequate shape and tonicity of the gum and surrounding mucosa. In the cone beam tomography control at 6 months postoperative there was no evidence of recurrence of the intraosseous lesion.
On histopathological analysis of the samples corresponding to the lateral borders of the right and left tongue : histological sections revealed fragments of mucosa lined by stratified pavement epithelium with parakeratinized and hyperparakeratinized areas exhibiting suprabasally the presence of fissures or the formation of microvesicles/blisters separated from the spinous layer, focal areas are observed totally desquamated leaving only the basal layer which has an aspect of tumulus, the presence of acantholytic cells (round shape) can be observed in these spaces or intraepithelial fissures, subepithelially discrete inflammatory of chronic nature is observed. The diagnosis of pemphigus vulgaris was reached, however, an immunofluorescence test (IgM - IgG) was required for confirmation.
As for Vial 3 corresponding to the maxilla sample: histological sections revealed fragments of cystic capsule composed of dense fibrous connective tissue permeated by an intense chronic and acute diffuse inflammatory infiltrate. This capsule is lined by a non-keratinized stratified epithelium with acanthosis and exocytosis. In other sections the presence of bacteria compatible with actinomyces spp. Fragments of viable and non-viable mature bone tissue of trabecular type and areas of hemorrhage complete the histopathological picture. The definitive diagnosis was reached by performing the clinicopathological correlation of a radicular cyst with data of bacterial infection by Actinomyces.
The lesions on the lateral border of the tongue, floor of the mouth and bilateral cheek were treated with a topical paste based on clobetasol propionate 0.05% plus retinoic acid 0.1% and orabase gel 15 gr, this was applied every 8 hours for 45 days, after the improvement of the lesions it was applied every 24 hours for 45 more days. After 6 months of control, partial

remission of the lesions was evidenced. She is currently on medication only with prednisone 20 mg 1cd/24 hours, later the dose will be reduced to 5 mg cd/24 hours until total remission of the lesions, as for the oral melanosis, strict clinical and photographic control will be carried out to ensure that the number of lesions does not increase and also that the blackish color does not intensify. She is being treated for Chagas disease with the internal medicine and cardiology services. Regarding the carious lesions, she is under comprehensive dental treatment.

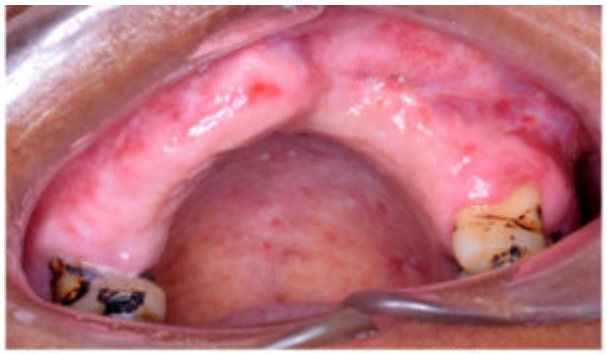

Figura 8: Imagen posoperatoria a 21 días.

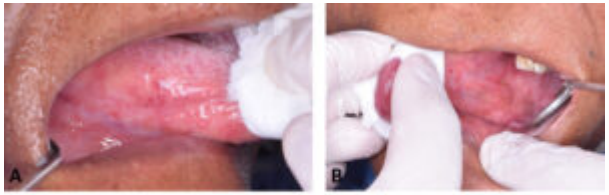

Figura 9: Control de lesiones a 45 días de terapia con corticosteroide tópico A)Borde lateral de lengua derecho B)Borde lateral de lengua izquierdo.

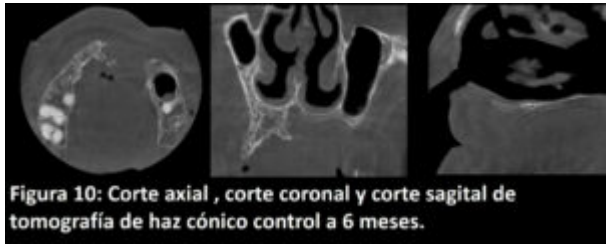

Figura 10: Corte axial , corte coronal y corte sagital de tomografía de haz cónico control a 6 meses.

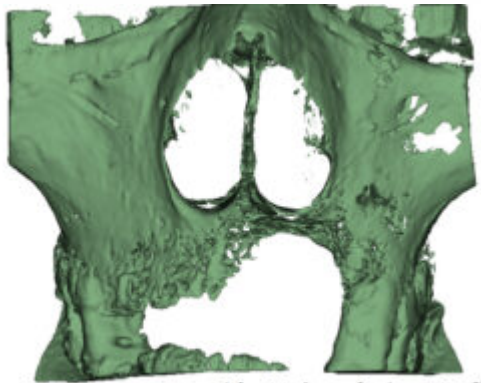

Figura 11: Reconstrucción Volumétrica 3D final.

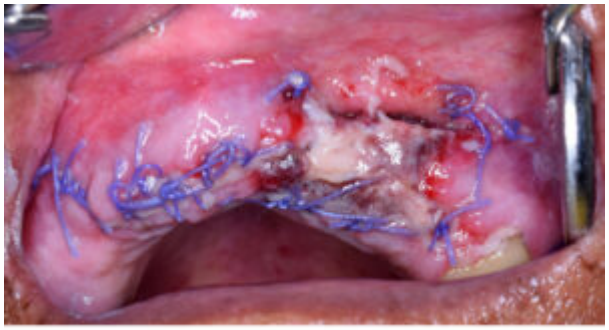

Figura 7: Imagen Posoperatoria a 7 días.

Discussion

It is important to make the differential diagnosis of the residual cyst with other pathologies such as odontogenic keratocyst, nasopalatine cyst, Gorlin's cyst or tumor lesions of odontogenic origin. It has also been reported that the residual cyst can infrequently transform into squamous cell carcinoma, which is why it is important to perform a total enucleation of the lesion to avoid recurrences.

These cysts can also cause significant bone loss, resorption and displacement of vital areas such as the inferior alveolar canal, maxillary antrum and nasal cavity as shown in the present clinical case. The probable factors for flap distress were: poor or no bone support remaining after enucleation and the Actinomyces infection already described. The factors that contributed to adequate wound healing were: preservation of the periosteum, collateral irrigation from the major palatine arteries and coronary arteries and finally the topical use of 0.2% chlorhexidine digluconate gel plus hyaluronic acid.2% plus hyaluronic acid at 1% with its characteristics of 1) antiseptic effect on the damaged tissues thus preventing the growth of oral biofilm and 2) regularization of the inflammatory process 3) easy application due to the high viscosity, all these conditions are advantageous for an adequate and accelerated healing of the surgical wound.

Probably the Chagas disease, not being under medical treatment, could generate a state of immunosuppression and reactivate the autoimmune disease of pemphigus of the oral mucous membranes. Taking into account the risk habits practiced by the patient, such as chewing tobacco with coca leaf and being a chronic smoker, the possibility of malignization of the oral lesions cannot be ruled out, so they require strict control.

Conclusion

Enucleation treatment with curettage is the most recommended for the treatment of residual cysts since it decreases the probabilities of recurrence. The topical use of 0.2% chlorhexidine digluconate gel with hyaluronic acid contributes to the repair of surgical wounds with poor bone remnant.

Other relevant documentation

Thanks to Dr. Alison Calle Mansmith, specialist in Oral and Maxillofacial Radiology, and Dr. Roberto García Rejas, specialist in Oral and Maxillofacial Pathology, for their collaboration in the clinical case.
The author has no conflicts of interest.

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