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An atypical presentation of gigantiform torus palatinus: A case report

Atypical tori palatine and surgical management

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ABSTRACT

INTRODUCTION: Torus palatinus (TP) is a slow growing maxillary exostosis, commonly found in the in the hard palate on either side of the median raphe of the palatine bone. Its aetiology is still unclear, but it has been attributed to genetic events and environmental agents, with a slight predilection for the female gender.

PRESENTATION OF CASE: The aim of this case report was to describe an unusual presentation of nodular TP in younger female patient, with medical history of hysterectomy when she was 25 years old. The TP manifested insidious progression that affected functional activities such as eating, swallowing and normal pronunciation. Furthermore, we describe our experience during pre-planning the surgery by using 3D-scan-printing, and the conservative surgical approach performed.

DISCUSSION/CONCLUSION: TP may be associated with epidemiological, environmental and systemic disorders of patients suffering from this condition. Conservative surgical removal continues to be the first choice treatment when the TP must be removed. Further studies are considered necessary to elucidate the etiological factors.

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1. Introduction

Torus palatinus (TP), also known as maxillary exostosis, commonly localized in the region of the hard palate as excessive bone tissue formed slowly, is characterized by a highly dense and strictly limited group of bone marrow, covered by a thin mucosa, with little vascularization. Its growth is slow, and in most cases asymptomatic, with the exception of some cases in partially edentulous or completely edentulous patients, in whom wearing dental prostheses is made difficult and sometimes impossible without removal of the lesion [1].

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The aetiology of the torus is still uncertain; however, the genetic profile is considered one of the factors most associated with development of the pathology. On the other hand, among others, temporomandibular disorders, bruxism, malocclusion, eating habits and a deficient diet of vitamins and medications, are associated causes that would lead to disorders in calcium metabolism [2,3].

Clinically, this case usually is diagnosed on an occasional and sudden basis, since there are no painful symptoms – taking into consideration that the majority of patients do not report any clinical manifestation. When seeking clinical care, patients usually report complaints of phonetics, some ulceration in the underlying mucosa; or when they need to have a prosthesis made or inserted, this makes it necessary to have the lesion removed [4].

The present case report aimed to describe the atypical presentation of maxillary bone exostosis (torus palatine) and to show our experience with the use of prototyping to perform pre-surgical planning and clinical results obtained by means of conservative surgery. This study was reported in line with the SCARE criteria [5].

2. Case report

The patient, a 46-year-old woman, arrived to our outpatient Oral and Maxillofacial Surgery clinic, complaining of pain in the hard palate while eating and swallowing, in addition to speech difficulties. With regard to the patient's medical history, she was submitted to hysterectomy at 25 years of age, and told us that her grandmother and sister had presented smaller TP lesions than the one in her condition.

On intraoral clinical examination, an extensive lesion of solid consistency was observed. It was fixed and covered by normal mucosa that showed isolated areas of ulceration in the central portion of the lesion, as related by patient. In addition, it extended from the anterior region of the hard palate until it invaded the space up to 1 cm from the limit of the soft palate. The lesion measured approximately 5 cm in the anteroposterior direction, 4 cm and 2.5 in the transversal plane, in the posterior and anterior region of the hard palate, respectively (Fig. 1).

The patient reported a slow and insidious growth over the course of many years (she reported that it appeared when she was approximately 5 years old) and that in the last 5 years, the lesion had increased considerably in volume and thickness. Moreover, a change in its initial color was perceived, with whitish and ulcerated areas due to the increase in size and the patient's constant attempts to adapt to the clinical condition.

Relative to imaging assessment, in the initial panoramic radiograph, a well-defined radiopaque area located posterior to the maxillary central incisors was observed (Fig. 2A). However, computed tomography (CT) revealed a rounded hyperdense area that started in the anterior region of the hard palate and extended to the posterior portion of the hard palate, compromising the soft palate. In the caudal direction, the lesion extended from the floor of the nasal cavities to the level of the occlusion plane in the posterior region. Finally, the sagittal sections showed a large amount of bone



Fig. 1. Nodular exophytic mass localized in hard palate presenting with solid consistency, fixed and sessile base from anterior region towards soft palate.

tissue, with a thickness of approximately 3 and 4 mm, with few areas surrounded by lamellar bone (Fig. 2B).

In the 3D reconstruction, the large size of the bony protuberance was detected, which occupied approximately 70% by volume of the hard palate. In addition, the following features were confirmed: the protuberance was fixed and well-defined within a sessile base, and showed a slight subdivision in the anterior region, closer to the nasopalatine duct (Fig. 2C).

After confirmation of the anatomopathological analysis by incisional biopsy taken, in which the presence of a compact bone with areas including osteocytes, Haversian canal and lack of cancellous bone were shown, these features were considered compatible with Torus Palatinus (TP) as shown in Fig. 3A. Thus, surgical excision was chosen, with surgical pre-planning based on the 3D scan printing (Fig. 3B).

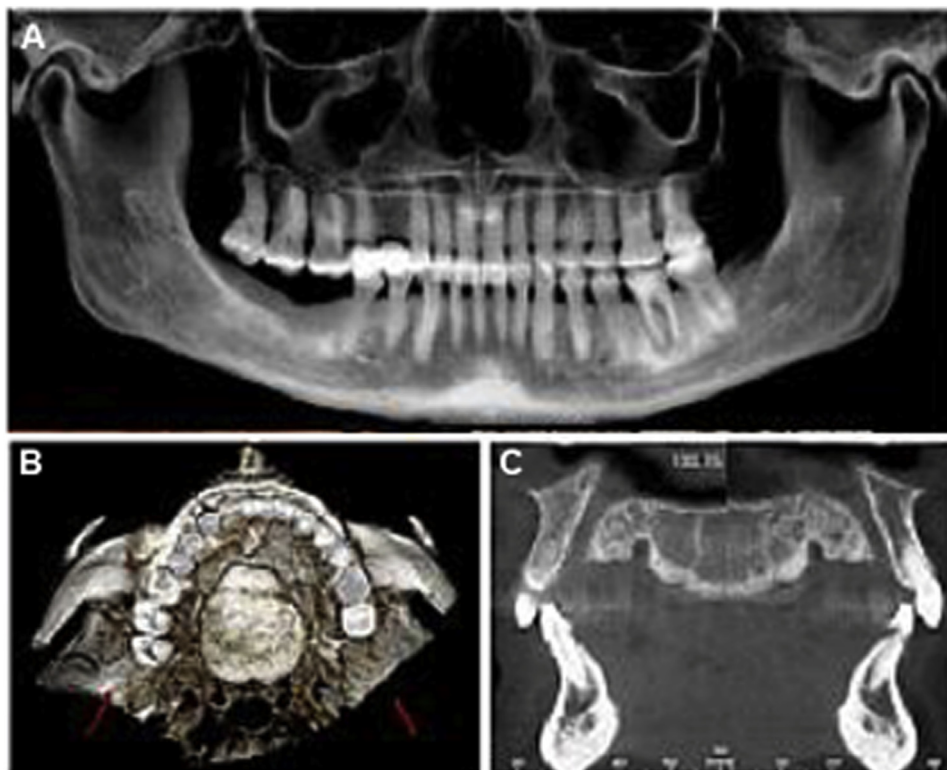


Fig. 2. Imaging analysis with A) panoramic radiography (orthopantomography), B) 3D Reconstruction and C) Computed Tomography (CT) sagittal section showed the size, extension and length of TP.

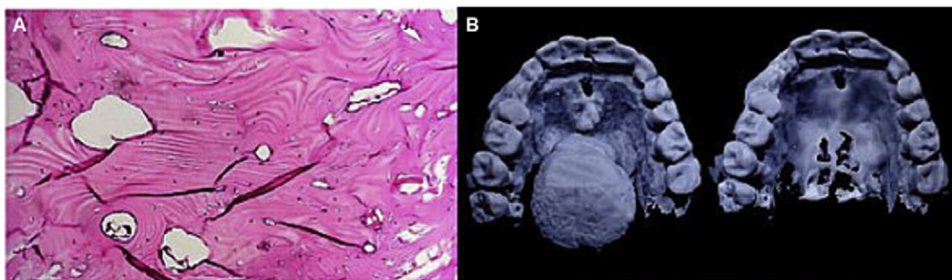


Fig. 3. A) Histopathological assessment showed cortical bone, isolated Haversian ducts and absence of cancellous bone; B) 3D scan prints after technique performed.

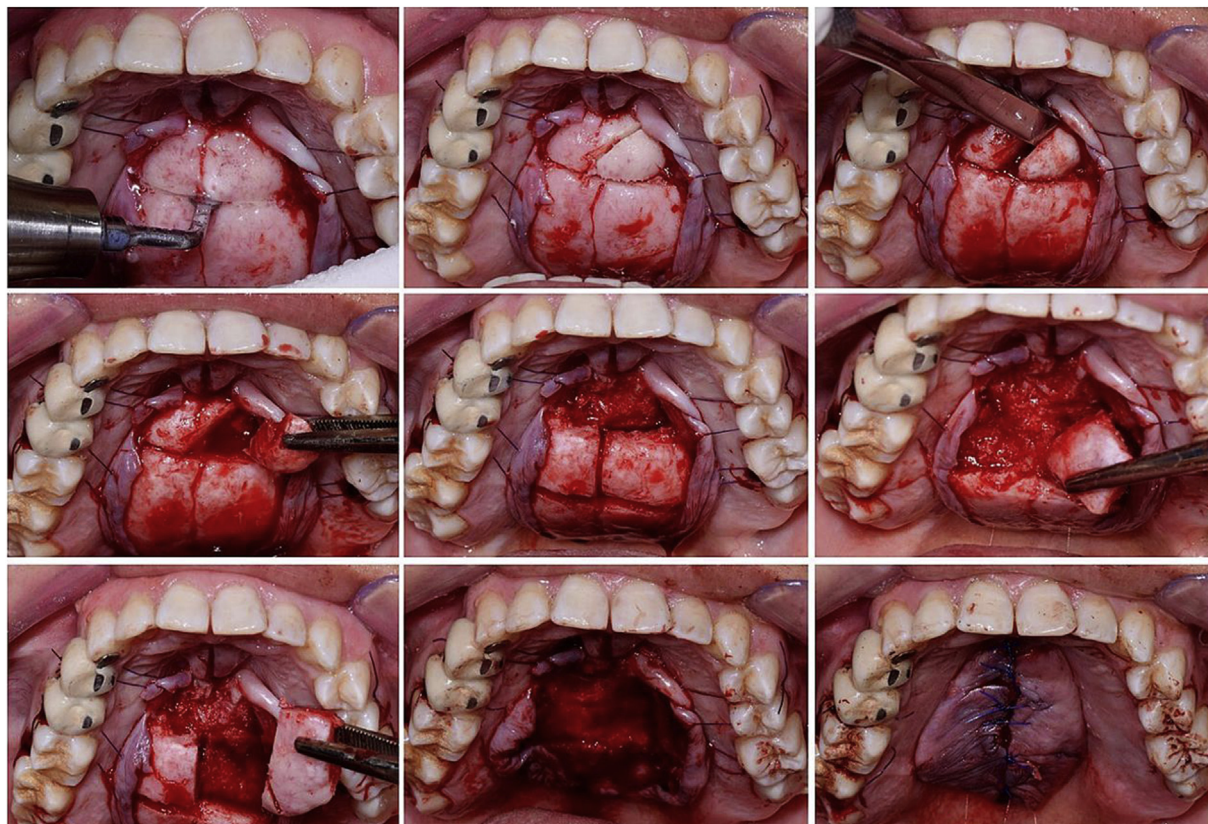


Fig. 4. Conservative surgical management of TP by osteotomy using piezoelectric instruments was performed, concluding with primary wound closure.

With reference to the technique, which consisted of obtaining an impression of the lesion in acrylic resin, by printing it on the AccFad-D1 Dental 3D printer, with a speed of 40 mm/h, a layer thickness of 25 microns, and the source used was a 405 μm UV LED light, Model OD01, and SHINING DENT photo-activated resin. Acquisition of the impressions served to determine the actual size of the lesion, to define the limits, and thereby perform more accurate surgery with predictable clinical results. After printing was performed, invasion of the lesion into the oropharynx region was confirmed, according to which a projection of the osteotomy was planned, which covered the base of the exostosis itself.

The surgery was performed under general anesthesia (GA) using a conservative approach, for excision of exostosis and remodeling of the remaining bone tissue. An incision was made with a scalpel 15, along the entire length of the palatal raphe, approximately 6 cm in length, from the posterior region of the central incisors towards posterior limit of the hard palate. Subsequently, mucosal tissue and the underlying periosteum were detached in order to expose the

subjacent lesion to the largest possible extent. Then, suspension points were placed; these were fixed laterally onto the teeth with 4.0 suture.

The surgical technique itself was based on the initial osteotomy using piezoelectric instruments and abundant irrigation with 0.9% saline solution; this was complemented manually with osteotomy and osteotomy with chisel and hammer. Bone remodeling was carried out immediately with gubia forceps and burs placed in a high speed hand-piece in order to provide irregular areas with a uniform aspect. Finally, the excess covering mucosa was removed (approximately 2 cm), and primary closure of the surgical bed was performed with simple sutures (Fig. 4).

In the immediate postoperative period, antibiotic, anti-inflammatory and analgesic therapy was indicated, and the suture was removed 15 days after the procedure, without dehiscence or signs of inflammation, but with abundant granulation tissue (Fig. 5). Follow-up performed after 6 months of surgery showed no signs of recurrence.

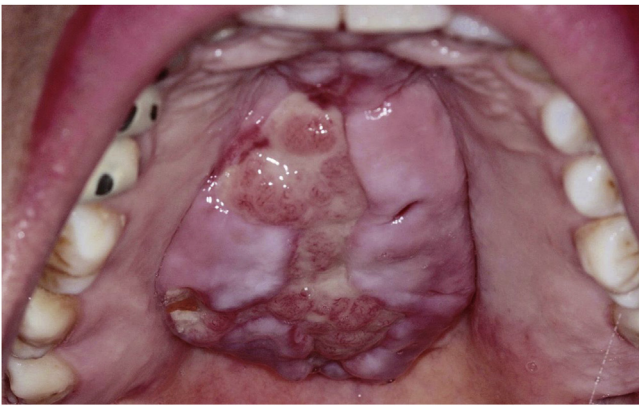


Fig. 5. Wound appearance fifteen days after surgical removal without signs of infection and higher level of surrounding granulated tissue.

3. Discussion

TP has been described as an anatomical variation rather than a pathological condition, which more frequently affects the female gender and young population [6,7]. Its aetiology is still unclear, but is commonly attributed to genetic and environmental factors [6–8], with the majority of cases having a causal factor resulting from environmental agents (70% of cases) [7] rather than from some type of genetic mutation on an autosomal dominant inheritance pattern [8]. However, recent studies have shown that the genetic and epidemiological profile of the population influences the appearance of this clinical condition according to variations in morphology and ethnicity, with a prevalence of 0.0%–66% for TP [7].

A diverse morphological variation can be seen such as flat, nodular, lobular and spindle shaped, the prevalence of which depend on the epidemiological profile of each population [8]. Whereas surgical removal is very rarely performed; however, in some cases it has been considered. Otherwise, when specifically indicated in order to reduce changes in phonetics, eating functions or interference in the ability to wear dental prostheses, conservative surgical treatment must be performed, as shown in this case report of nodular gigantiform TP [9,10].

Various techniques have been used in the conventional surgical treatment of these lesions [4–10] since the technique will depend on the extent and location of the lesion, and the epidemiological profile of the patient. There are cases in which pre-surgical planning with the aid of auxiliary tools such as prototyping is necessary, in order to reduce the risks inherent in surgery when the lesion is medium to large, and favor a clinically adequate postoperative period for patients.

Lastly, laser surgery for removal of TP lesions with reduced volume has shown favorable clinical results [9]. However, in cases in which the volume and extent of the lesion become limiting factors that should be taken into account for surgical removal, the use of auxiliary 3D techniques could be necessary to obtain satisfactory clinical results in both the perioperative (mainly during surgical technique performed) and postoperative stages, as described in the present study. Planning of the surgery by predetermining the exact limits of the lesion, based on the 3D molding obtained, favored the surgery performed and ensured a better postoperative outcome, as described in this case.

Finally, the present case report showed an unusual presentation of TP because its size was shown to be without associated comorbidities, in a younger patient, who had a previous history of hysterectomy at 25 years of age, and had noticed an increase in volume of the exostosis in the last 05 years. The possibility of considering the surgical data in this case as a possible, special sys-

temic triggering factor for progression of growth of the lesion in the patient, was in fact uncertain, because subsequent change in the patient's bone metabolism after hysterectomy had not been properly recorded.

Therefore, propose that wider scientific investigations should be encouraged in these types of cases, in order to determine the exact repercussion of changes in bone metabolism caused by systemic disorders. This should also apply to surgical events that involve hormone alterations or other systemic alterations, as was currently described in a study with a group of patients that received dialysis treatment (hemodialysis and peritoneal dialysis) for a long time. In the cited study, the peritoneal dialysis group showed a higher prevalence of TP when compared with hemodialysis group [7].

4. Conclusion

TP may be associated with the epidemiological profile, environmental features and systemic disorders of patients suffering from this condition. Conservative surgical removal is still the first choice treatment when the lesion must be removed. New auxiliary technologies such as 3D scan printing could be considered to improve surgical planning. Further studies are necessary to elucidate the etiological factors that play a role in the occurrence and progression of disease.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

This report was exempt for ethical approval because of the use of medical data records of the patient without exposure of her identity or photos that shows her facial identification or characteristics. We are including a document entitled "Patient Release Form" that consists in to explain details of further publication to the patient and about the Journal. It was assigned which represents the agreement of patient.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

All authors have contributed in the each step for writing this paper, participating in the whole process to retrieve medical information, review of the literature, and writing of each issue included.

Wilber Edison Bernaola-Paredes has written and selected the topics for structuring this case report. Moreover, he did and worked in the Introduction and Discussion issues. On the other hand, Thayná Albuquerque, Amanda Mesquita, Ivan Solani and Flavio Lima has done the literature review in order to support our discussion and introduction. Kleber Vallejo-Rosero did the case report (presentation and details about it) and took the all pictures during the surgery performed and follow-up in the clinical attendance.

Kleber Vallejo was the first oral and maxillofacial surgeon who performed lesion removal. Wilber, Ivan and Flavio Lima participated as members of the team.

Registration of research studies

NA.

Guarantor

Wilber Edison Bernaola-Paredes/Kleber Vallejo-Rosero.

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