Case Report

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Dentistry Section

Sialadenoma Papilliferum of Hard Palate Mimicking Squamous Cell Carcinoma: A Case Report

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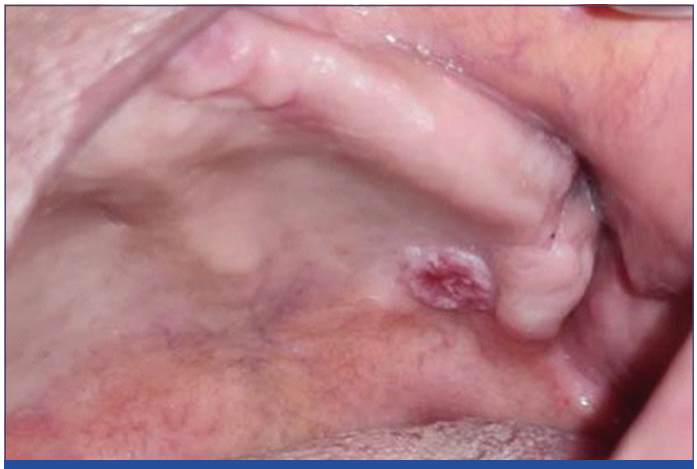
ABSTRACT

The occurrence of unusual ulcers in the oral cavity has fundamental value regarding oral and systemic diseases and abnormalities, as it can show clinical evidence that differs from other lesions, contributing to the diagnosis process. A 64-years-old female patient reported with a non healing of an oral lesion since two months (seeking clinical treatment for an atypical hard palate ulcer). A 64-years-old female patient was presented with a two month history of an oral lesion that did not repair. Her medical record indicated past smoking habits and hypertension. During clinical evaluation, an exophytic ulceration located at the left posterior hard palate, with a rounded form and elevated edges, was observed. An incisional biopsy was performed to confirm the diagnosis. Histopathological complementary exams revealed biphasic papillary projections, extending from the epithelium through the connective tissue, that are considered ductal structures. The final diagnosis was sialadenoma papilliferum, and total surgical excision was performed as a treatment. Follow-up showed no recurrence over 14 months. Sialadenoma papilliferum is a rare benign salivary gland tumour, mostly known for its papillary aspect and categorized as a ductal papilloma. Sialadenoma papilliferum origin is not precisely defined, nor associated with a single cause, given that different investigators pointed to multiple variables to Sialadenoma papilliferum origin. Being one of the rarest salivary gland tumours, SialP's clinical aspects and its resemblance to other benign salivary gland tumours can lead to a wrong diagnosis if not properly and microscopically analyzed, hence the importance of knowing its features.

Keywords: Head and neck neoplasms, Oral ulcer, Oral ulcer, Salivary glands

CASE REPORT

A 64-years-old female patient came to the Dental Clinic complaining of a non healing wound, lasting approximately two months. The patient had a medical history of hypertension, treated with a daily 20 mg dose of Losartan, in addition to smoking habit from the last 20 years (smoking four cigarettes per day). The clinical intraoral inspection revealed the presence of a round-shaped lesion with elevated edges, ulcerated, and with an exophytic appearance. Intraoral palpation revealed that the lesion was firm. Colour ranged from pinkish to white on the edges and red in the center. The lesion was located in the posterior region of the left hard palate [Table/Fig-1].



[Table/Fig-1]: The intraoral aspect of the lesion located on the left hard palate.

Initially, the only diagnostic hypothesis was Squamous Cell Carcinoma (SCC), because of the patient´s past smoking habits, and the clinical feature of an ulcer with elevated edges. This provisional diagnosis of SCC was considered only after the clinical intraoral inspection and the patient´s history, and no histopathological evidence had been analyzed at this point. To better analyse the lesion, an incisional biopsy was performed under local anaesthesia, and the removed fragment was sent for histopathological microscopic analysis.

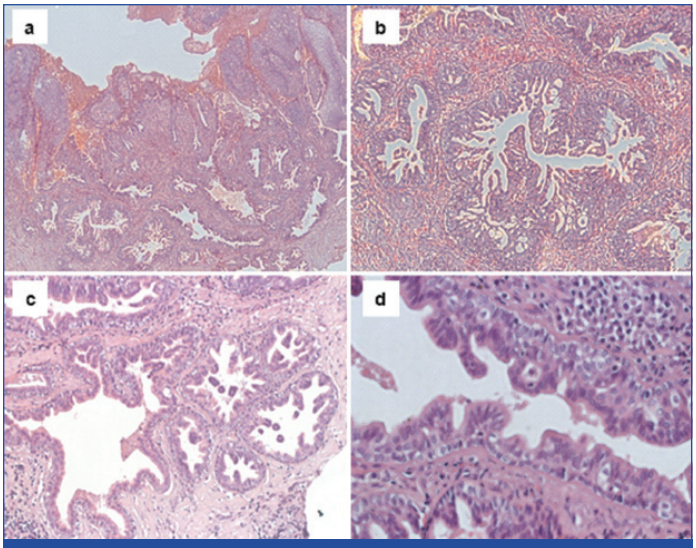
The histopathological analysis revealed biphasic papillary projections, with an exophytic superficial component of stratified squamous epithelium with incomplete keratinisation, this epithelium being contiguous to endophytic proliferations that form ducts in the submucosal region [Table/Fig-2a]. In addition, the endophytic projections form a ductal structure with an irregularly shaped lumen, which some authors [1,2] defined as a fissure shape, with multiple branching [Table/Fig-2b].

At higher magnification, one can observe the ductal structure of the papillary projections, composed of a double layer of different cells [Table/Fig-2c]. This structure has basal layers composed of squamous epithelium and a luminal layer of cells, ranging from cuboidal to columnar [Table/Fig-2d].

Based on the histological findings, the final diagnosis was determined as sialadenoma papilliferum. As treatment, the patient was put under local anaesthesia and total surgical excision of the lesion was performed resecting the surgical margin of 1mm from the normal tissue. After the surgery, the only postsurgical management mode necessary was follow-up, and the patient attended, initially, monthly visits to the clinic, which showed that she had no recurrence nor any changes at the biopsy site for 14 months. After a year of follow-ups, the patient´s follow-up frequency was modified, and she has been attending the clinic every 6 months.

DISCUSSION

Sialadenoma papilliferum is a condition that mostly occurs in the intraoral mucosa, especially in the hard palate [1]. Based on previous studies, sialadenoma papilliferum consists of a rare benign



[Table/Fig-2]: Histological features of Sialadenoma papilliferum. a): squamous surface characterizing exophytic growth extending into the submucosa as endophytic ducts (H&E staining; 5X) b): ductal lumen with irregular branches (H&E staining; 10X) c): ductal lumen with a double layer of cells (D) ductal endophytic structure with a basal layer of squamous cells and a luminal layer of cuboidal to columnar cells (H&E staining; 40X).

tumour of the salivary glands, considered a ductal papilloma [3]. The ductal papilloma classification of sialadenoma papilliferum is related to several characteristics that can occur in salivary glands tumours: histologically, papillary exophytic projections can be observed, much like subjacent ductal proliferations that extend into the connective tissue [4]. According to the fourth edition of the World Health Organization (WHO) Classification of Head and Neck Tumours, the overall characteristics of sialadenoma papilliferum cases reported in the literature show that sialadenoma papilliferum is a tumour mostly located in the hard palate and especially found in male adults [2].

Salivary glands are anatomic structures in which the glandular tissue commonly discharges in ducts, producing fluids known for differing in the type of secretion produced (serous, mucous, and mixed), and they are studied based on different criteria, however, commonly sorted into two categories: major and minor salivary glands [5]. The vital importance of the salivary gland in healthcare becomes evident, with the necessity of amplifying studies that aim to explore common abnormalities of these structures. Salivary gland tumours, such as sialadenoma papilliferum, are fundamental in the Oral and Maxillofacial Pathology comprehension as a whole, considering that those signs can turn into new paths in the search for a more effective diagnosis [3].

In contrast to that, the knowledge of malignant salivary gland tumours has increased at a much higher level than benign salivary gland tumours, given that possibilities in diagnosis and pathogenesis are widely discussed regarding malignant tumours [6]. This is extremely harmful to the correct diagnosis of salivary gland and overall oral cavity abnormalities since many benign salivary gland lesions exist nowadays and are uniquely classified, with different diagnoses, treatments, and features [6].

Sialadenoma papilliferum was first described in the literature by the authors Abrams AM and Franck FM, in 1969, who reported two previously unknown clinical cases, a mass located in the region of the parotid gland and another at the junction of the hard and soft palate [7].

According to data from the Armed Forces Institute of Pathology (AFIP) , sialadenoma papilliferum to approximately from 0.4% to 1.2% of all neoplasms present in minor salivary glands. Regarding the general characteristics of Sialadenoma papilliferum, its predominant location is intraoral, with the hard palate being the most common region, followed by the buccal mucosa [4]. The highest occurrence of sialadenoma papilliferum is in older adult males, especially between age group of 40 to 80 years [2,8,9]. Usually, sialadenoma papilliferum is described as a lesion of slow progression [8,9], which is not observed in the present case report, since the patient sought dental care during a 2 month progression period.

Clinically, sialadenoma papilliferum can be found as a papillary exophytic growth, especially in minor salivary glands [3]. In microscopic analyses, it has been noted that the most common histological features of sialadenoma papilliferum are biphasic projections that characterize an exophytic proliferation of the squamous epithelium, contiguous with the formation of papillary ducts in the submucosal area [1-4].

The most common treatment for sialadenoma papilliferum is total surgical removal, with rare recurrences [2,3]. Koç AK et al., reported the first successful case of Transoral Robotic Surgery (TORS) - a recent technique used in head and neck surgical treatments - which had no recurrence and was proven to extend surgical performance and technical control during the treatment [10].

There is still no established consensus on the possible cause or pathogenesis of sialadenoma papilliferum, and a possible relationship with human papillomavirus (HPV) has not been proven [11]. Freedman PD and Lumerman H, proposed that SialP cells can originate from excretory duct reserve cells [12], in contrast to Abrams AM and Franck FM, who stated a possible myoepithelial origin.

Some factors, such as the clinical and histological appearance of the lesion, may resemble other head and neck abnormalities that can be the differential diagnosis, such as squamous papilloma, intraductal papilloma, and inverted ductal papilloma [1,3,4,9,13]. Although squamous papilloma has similar clinical features, it does not have biphasic projections, since it presents only squamous proliferation of the epithelium [4]. In addition, ductal papillomas such as intraductal papilloma and inverted ductal papilloma have an endophytic component; however, it is not possible to observe exophytic proliferation in these cases [7].

CONCLUSION(S)

In conclusion, sialadenoma papilliferum is a benign neoplasm that occurs in salivary glands, especially in minor salivary glands, which affects mostly the hard palate region. It has clinical features of an exophytic lesion and can be histologically differentiated from other ductal papillomas by its biphasic component. Total surgical removal is an effective treatment and the prognosis is good.

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