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Review Article

Intraoral Sialadenoma Papilliferum: A Comprehensive Review Of The Literature With Emphasis On Clinical And Histopathological Diagnostic Features.

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Abstract

Context. Diagnosing Sialadenoma papilliferum (SP), a rare benign epithelial tumor of salivary gland origin, can be difficult. It was initially identified as an analog of the cutaneous syringocystadenoma papilliferum by Abrams and Finck in 1969. Through the analysis of previously published cases and the reporting of new cases, this thorough review seeks to highlight the clinical and histological diagnostic characteristics of intraoral SP. Techniques. Up until February 2022, Medline, Scopus, and Web of Science were searched using the entry word “sialadenoma papilliferum.” Only English-language studies were considered, and there were no time restrictions. Only mouth-related cases were covered. Letters to the editor, personal correspondence, and conference proceedings were not included. outcomes. Overall, The palate, especially the hard palate, was the area most impacted. There have been reports of four cases with unclear malignant characteristics. The preferred treatment for SP is conservative excision because of its low growth potential, unusual histological and immunohistochemical characteristics, and somewhat nonspecific clinical symptoms (such as submucosal edema with ulceration). Conclusions.Even though SP is uncommon, it should be considered when making a differential diagnosis for intraoral swellings, especially those that are on the palate. Typically, there is an erythematous patch inside a mucosa that is otherwise normal (Figure 1A). Based on the keratotic appearance of the papillary surface, papilloma is the most common differential diagnosis. Soft tissues neoplasms, pyogenic granulomas, palatal fistulas, and other benign and malignant small salivary gland tumors are additional clinical diagnostic theories.

Keywords : *sialadenoma papilliferum; salivary gland tumors; oral pathology; oral medicine; oral surgery.*

INTRODUCTION

Stensen’s and Wharton’s ducts, as well as minor and sublingual salivary glands, can be the site of benign or malignant intraoral salivary gland tumors. Sialadenoma papilliferum (SP) is a very uncommon benign lesion, and diagnosing it can be difficult [1]. SP is classified as a benign epithelial tumor in the most recent World Health Organization (WHO) classification of salivary gland tumors [2].Since Abrams and Finck’s 1969 [1] initial report of SP, there have only been 63 intraoral instances published in the English literature. Waldrom et al. claim that SP is responsible for 2% of benign salivary gland tumors and 1.1% of small salivary gland cancers [3]. The term comes from its histological resemblance to syringocystadema papilliferum, a rare benign tumor of the sweat glands that predisposes to the forehead and scalp [4].Here, we present a thorough analysis of the literature with a focus on intraoral SP’s clinical and histological features.

MATERIALS AND METHODS

The search term “sialadenomapapilliferum” was used to search the Medline, Scopus, and Web of Science databases. The database was screened through February 2022. Only English-language studies were taken into consideration, and there were no time restrictions. Only mouth-related cases were covered.Letters to the editor, personal correspondence, and conference proceedings were not included.Titles and/or abstracts were screened at the first level, and in contentious cases, the complete text was assessed. Reviews’ references were examined to find any works that might have slipped through the cracks of the database search.Title, authors, year of publication, number of reported cases, oral subsite, and lesion size were among the retrieved data (Table 1). Table 1: Information from the 42 publications (64 cases) that are part of this review Scheme 1 shows the flowchart used to choose the 42 papers that were reviewed. 3. Findings After

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removing duplicates, the list of 234 papers that the database search produced was whittled down to 99. 42 publications were selected for additional data extrapolation and analysis after being evaluated in full text and abstracts as particularly reporting on SP of the mouth. Just to note, four instances of SP affecting the parotid were discovered and, based on the criteria employed, were disqualified [1,46–48]. Since the lesion was first described by Abrams and Finck [1] in 1969, 64 cases have been found (Table 1).3.1. Sex and Age With the exception of two cases involving a guy aged 18 and a male aged 20, all patients were older than 30 years, with ages ranging from 18 to 87 years, with a mean age of 57.2. The current review has shown that SP is more common in men (39 men and 25 women; M/F ratio: 1.6:1). 3.2. Medical Characteristics With 48 (75%) cases, the palate was the most frequently implicated site. Of these, 37 (58%) were on the hard palate, 6 (9.5%) were on the hard and soft palate junction, 4 (6%) were on the soft palate, and 1 (1.5%) was on an unidentified palatal localization. Additional locations were the upper lip mucosa (3; 5%), buccal mucosa (6; 9.5%), Figure 1 shows a 48-year-old female patient's hard palate with sialoadenoma papilliferum (A); surgical excision performed under local anesthetic (B); and a surgical specimen (C). Tumor sizes in the patients included in this analysis ranged from tiny (0.3 cm) to large tumors up to 4 cm, with an average of 0.79 cm.Abram's case is one of four examples that have been reported to date, and larger cases up to 7 cm can occur in the parotid [1,46].3.3. Characteristics of HistologyThe histopathologic pattern of SP was relatively characteristic, and the histological descriptions of the cases assessed in this analysis were rather consistent.The surface part of the salivary glands' excretory ducts appears to be where the tumor started. Convoluted clefts and gaps are finally formed by the development of papillary processes. A core of fibrovascular connective tissue supports two or three layers of cells that line each papillary projection. Squamous epithelial lining surrounds the lesion's most superficial sections, while deeper regions mostly display cuboidal to columnar cells that are frequently oncocytic in appearance (Figure 2). Similar to a squamous papilloma, the mucous membrane covering it becomes papillary or verrucous as growth continues. Several studies have used light microscopy, electron microscopy (EM), and immunohistochemistry (IHC) to try and determine the cell of origin of SP [41]. The majority of researchers have proposed excretory duct or excretory duct reserve cell origin, although these methods have produced inconsistent results [9–12,19,24,29,31,41,44]. Other authors proposed that myoepithelial cells [1,29,41] or intercalated duct cells [12,49] could be the source.Basal cells on the ductal structures were shown to be immunoreactive for the myoepithelial immunophenotypes p63 and p40 by Fowler and Damm [41]. It was also discovered that there was variable responsiveness between smooth muscle actin (SMA) and the basal cell layer. The luminal cells inside the ductal structures were immunoreactive to the epithelial membrane antigen (EMA) in every instance they described in their research. According to our findings, the convoluted ductal structures of SP are made up of two cell types: a luminal layer of ductal epithelial cells (EMA+) and a basal layer of myoepithelial cells (p40+, p63+, and SMA+) (Figure 3A,B). Figure 2 shows an example of SP's histological characteristics (this case differs from the one shown in Figure 1):glandular processes beneath the mucosa and stratified squamous epithelium covering the exophytic papillary structure (H&E—50×). Figure 3. (A, B) Positive results for smooth muscle actin (SMA) and p63 by focal immunohistochemistry.p63 immunoreactivity in ductal structure basal cells indicating a myoepithelial immunophenotype. Additionally, ductal structures are clearly positive; (C,D) ductal luminal cells have positive immunohistochemistry expression for cytokeratins 7 and 14 (CK7 and CK14), which may validate the epithelial origin (100× and 200×).According to a recent immunohistochemical investigation published by Atarbashi-Moghadam et al., vimentin and smooth muscle actin are negative, but cytokeratins 13, 14, 7, 8, and 19 are positive. This immunoprofile resembles that of the salivary gland's excretory ducts [44] (Figure 3C,D). 3.4. Treatment The preferred course of action appears to be conservative excision (Figure 1B,C). Since SP is so uncommon, no clinical guidelines have been put forth on the potential length of follow-up. Van der Wal & van der Waal recommend that follow-up appointments be made on a regular basis [22]. 3.5. The outlook Only two cases out of 64 cases of SP, a benign neoplasm with limited development and potential for local aggression, were reported to return within three years. 3.6. Potential Malignant Development The presence of a malignant version of the tumor or the eventual malignant transformation of SP are unknown. Four cases with unclear malignancy have been recorded, based on this review.Others have contested the diagnosis (Ellis, G.L. and Auclair, P.L., 1991), although Solomon et al. reported a case with a potentially malignant SP [10]. An SP with potentially malignant characteristics, including fast and destructive growth, radiographic resorption of the underlying bone, and unusual histological findings, was described by Ide et al. [36]. Squamous cell carcinoma was the clinical diagnosis made after Santos et al. described a case of SP on the tongue with clinical features that seemed to be malignant [40]. Shimoda and associates. In other words, it is still unclear if SP has the potential to be malignant.

DISCUSSION

Although they are relatively rare in the minor salivary glands, salivary gland tumors are a morphologically and clinically heterogeneous group of neoplasms that mostly

attack the large salivary glands. When all malignancies of the salivary glands are taken into account, the annual incidence worldwide is roughly one case per 100,000 [49].The two most prevalent benign and malignant minor salivary gland tumors are pleomorphic adenoma and mucoepidermoid carcinoma, although SP accounts for only 1.1% to 1.6% of cases [3,9].The extremely varied clinical and radiographic characteristics, along with the relatively large variety of histological subtypes, are common challenges in the treatment of small salivary gland tumors. The majority of intraoral salivary gland tumors manifest as submucosal nodular swellings, with or without superficial ulceration, and SP shares this exophytic development pattern. Other lesions, such as exophytic ductal papilloma, verrucous hyperplasia, and squamous papilloma, share same clinic pattern.Verrucous hyperplasia and squamous papilloma can be clearly distinguished from SP since they exclusively exhibit squamous epithelial growth. A ductal proliferation beneath the epithelium is absent from an exophytic ductal papilloma, despite the presence of exophytic papillary ductal epithelial proliferation. Rather, an exophytic proliferation of papillary stratified squamous epithelium and a concurrent endophytic salivary ductal growth underneath are distinctive histopathologic hallmarks of SP [50].SP usually appears as an erythematous lesion with erosion or ulceration that could indicate a malignant lesion such sarcomas or verrucous carcinoma [38]. Since the majority of salivary gland tumors do not exhibit a gender preference and the mean age for SP in the current review is comparable to that reported for other salivary gland tumors (45 years, with a range of 11–74) [51], age and gender are not very useful in the differential diagnosis from an epidemiological perspective. With an average size of 0.79 cm at diagnosis, SP seems to have a limited capacity for growth, making conservative surgery an option. Despite the fact that four examples of alleged malignant transformation were documented, there is not enough evidence to establish SP's malignant potential.The cells of origin of SP are now the subject of research, however this question has not been resolved despite immunohistochemistry studies.In conclusion, although though SP is uncommon, it should be included in the differential diagnosis of intraoral swellings, especially those on the palate, and further research is required to fully comprehend its biology.

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