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Sialadenoma Papilliferum with Inverted Pattern in a Young Patient: A Case Report

BDEF **Francisca Emanuel Reis de Sá Silva e Costa**
D **José Ramón Vizcaíno Vázquez**

Department of Pathology, Hospital de Santo António – Centro Hospitalar do Porto, Porto, Portugal

Authors' Contribution:

Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

Corresponding Author:

Conflict of interest:

This case report was presented as a poster at the 24th European Congress of Pathology, held in Prague, Czech Republic, from 8 to 12 September 2012 and the abstract was published in an abstract book which was a supplement to Virchow's Archive. Francisca Emanuel Reis de Sá Silva e Costa, e-mail: kikareis_med@hotmail.com

None declared

Patient: Male, 20
Final Diagnosis: Sialadenoma papilliferum with inverted pattern
Symptoms: Intraoral nodular mass
Medication: —
Clinical Procedure: Surgery – excision
Specialty: Surgery

Objective: Challenging differential diagnosis

Background: Sialadenoma papilliferum (SP) is a rare, benign neoplasm of salivary gland origin which manifests as an exophytic papillary excrescence of the mucosa. Indeed, SP is both an exophytic proliferation of papillary stratified squamous epithelium above the mucosal surface and an endophytic salivary ductal proliferation beneath the mucosa. It arises predominantly in minor salivary glands and usually affects patients in the age range of 32–87 years, with reports in young patients being exceedingly rare.

Case Report: We report the case of a previously healthy 20-year-old man diagnosed with a nodular mass in the upper lip buccal mucosa. The tumor was excised and submitted for microscopic examination. Histologic examination revealed a biphasic proliferation of papillary stratified squamous and salivary ductal epithelia, both underneath the mucosal surface.

Conclusions: In this unique case, as the classical SP, the tumor had a biphasic proliferation of squamous and ductal epithelia. However, unlike the classical SP, both epithelia grew under the mucosal surface. As a result, it did not manifest as an exophytic proliferation, but as a nodule. We excluded squamous papilloma, inverted ductal papilloma, intraductal papilloma and mucoepidermoid carcinoma, the principal entities in the differential diagnosis of SP, and concluded it was an SP with inverted pattern.

MeSH Keywords: Papilloma • Salivary Glands, Minor • Young Adult

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Background

Tumors arising in the salivary glands represent less than 1% of all tumors and approximately 3–5% of all head and neck neoplasms [1]. Minor salivary gland tumors are infrequent, representing 10–15% of all salivary gland neoplasms and are primarily located in the palate (50%), lips (15%), cheek mucosa (12%), tongue (5%) and floor of the mouth (5%) [1]. However, they constitute a heterogeneous group of neoplasms with a wide range of histological types and growth patterns.

Benign papillary lesions that arise from the ductal system of salivary glands (ductal papillomas) comprise intraductal papilloma, inverted ductal papilloma, and sialadenoma papilliferum (SP) [2–4]. All of these neoplasms originate from the excretory portion of a salivary gland duct [2]. These 3 tumors are considered to be relatively uncommon and are source of great interest because of their striking similarities but also variability in clinical and histological presentation [3]. They are most commonly found in the minor salivary glands, being rarely diagnosed in major salivary glands [3].

SP is a benign, exophytic, neoplasm of salivary gland origin, first described in 1969 by Abrams and Finck [5]. “Sialadenoma papilliferum” owes its name to the histological resemblance to syringocystadenoma papilliferum of cutaneous adnexal origin [6]. Since the first reports of SP, only 53 cases have been reported in the English-language literature. SP is, thus, a rare and distinctive neoplasm. Clinically, SP manifests itself as a white-colored, exophytic, papillary, excrescence of the mucosa [2,4]. Histopathologically, unlike the other tumors, it has biphasic growth: an exophytic papillary squamous component and a glandular component [7]. Indeed, SP is both an exophytic proliferation of papillary, parakeratotic, acanthotic, stratified, squamous epithelium above the mucosal surface and an endophytic proliferation of salivary ducts that form tortuous clefts and cystic spaces beneath the mucosa.

SP arises predominantly in minor salivary glands and usually affects patients in the age range of 32–87 years [2,5,8], with reports in young patients being exceedingly rare [8]. According to the literature, about 70% of the cases reported have occurred in the minor salivary gland of the palate, in middle-aged or older patients (age average, 56.8 ± 15.5 years), with a male:female ratio of 1.6:1 [4], the upper lip, retromolar pad, faucial pillars, and parotid gland accounting for sporadic cases [2].

SP generally runs a benign course, but lack of encapsulation around the proliferating ductal components may give an erroneous impression of malignancy [3,7]. Rare recurrent cases have been reported [9] and malignant transformation of SP has only recently been recognized [2,6,9].

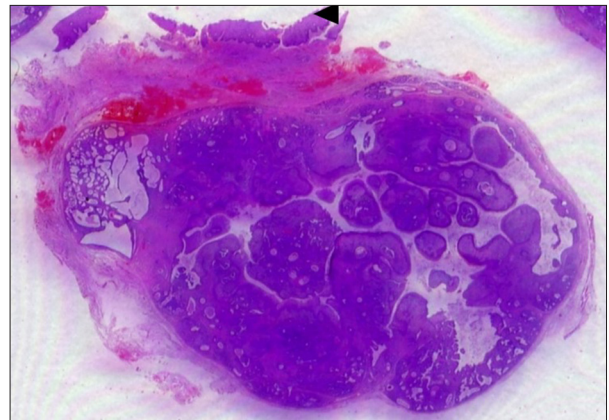


Figure 1. Sialadenoma papilliferum with inverted pattern. There was a well-defined, pseudo-capsulated lesion below the mucosal surface (arrowhead). Slide view, H&E.

The cell of origin of SP is still controversial. In fact, there are conflicting reports regarding this matter; some authors suggest that SP cells derive from excretory duct cells, while others concluded after ultrastructural analysis that the intercalated duct cell might give rise to this lesion [3]. Although the specific genesis of SP is still not universally accepted, it is unanimous that the cells of the excretory ducts are the likely point of origin.

Case Report

We report the case of a previously healthy 20-year-old man diagnosed with a nodular mass in the upper lip buccal mucosa, with unknown duration. The tumor was excised under local anesthesia, with a suspected clinical diagnosis of mucocele, and submitted for histopathological examination.

The clinical information and follow-up data were obtained from the records. The excised tissue specimen was fixed in buffered formaldehyde (4% formalin) and embedded in paraffin. Sections were stained with hematoxylin and eosin.

On gross examination, the specimen consisted of a nodular mass, measuring $1.6 \times 1.3 \times 0.7$ cm, partially covered by a mucosal fragment of 1.2×0.4 cm. The cut surface was yellowish-white, firm, with a centrally located cystic area of 0.3 cm in diameter.

Histologic examination revealed a biphasic proliferation of papillary stratified squamous and salivary ductal epithelia, both located underneath the mucosal surface.

Microscopically, there was a well-defined pseudo-capsulated lesion which grew in an endophytic way, creating a submucosal cystic space (Figure 1). The luminal cavity was filled with papillary projections and folds of keratinizing stratified squamous epithelium supported by fibrovascular cores containing

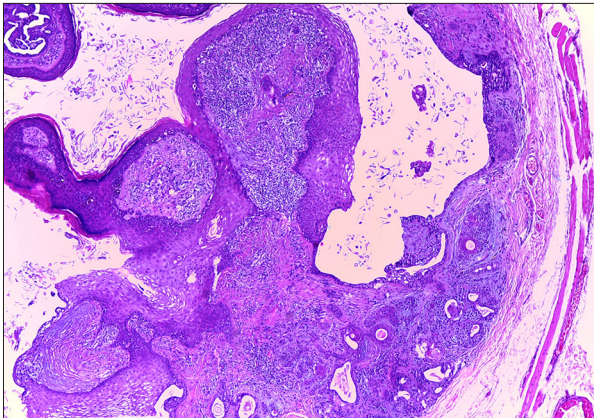


Figure 2. Sialadenoma papilliferum with inverted pattern. The papillary projections were composed by keratinizing stratified squamous epithelium supported by fibrovascular cores containing an infiltrate of lymphocytes and plasma cells. H&E, 40×.

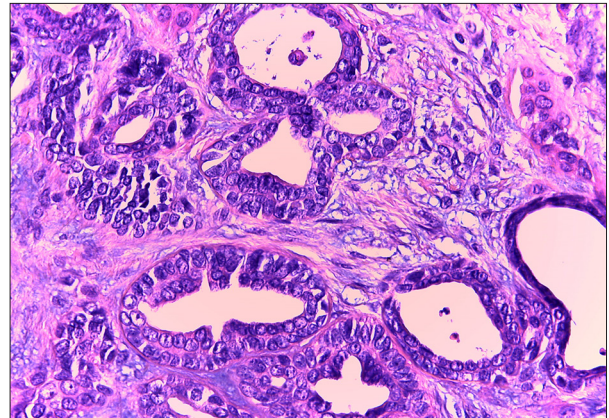


Figure 4. Sialadenoma papilliferum with inverted pattern. Beneath the papillary projections, there was a proliferation of small ducts lined with a double row of cells showing a basal layer composed of cuboidal cells and a luminal layer of columnar cells. H&E, 400×.

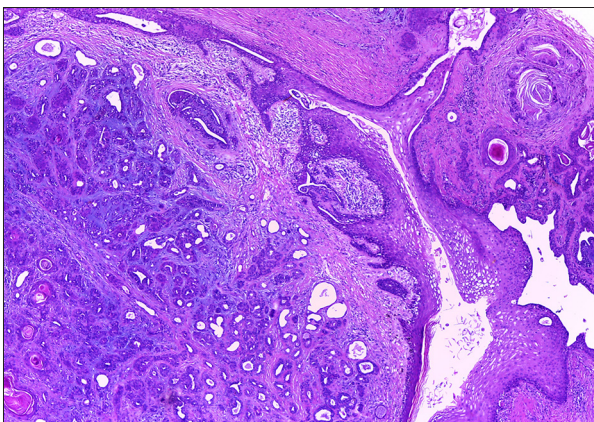


Figure 3. Sialadenoma papilliferum with inverted pattern. The lesion was composed by a biphasic submucosal proliferation of papillary stratified squamous and salivary ductal epithelia. H&E, 40×.

an infiltrate of lymphocytes and plasma cells (Figure 2), beneath which there was a proliferation of small and ectatic ducts (Figure 3). The ducts were lined with a double layer of cells: a basal layer composed of cuboidal cells and a luminal lining layer of columnar cells (Figure 4).

So far, our patient has a follow-up period time of 48 months, with no evidence of recurrence.

Discussion

In this unique case, as the classical SP, the tumor has a biphasic proliferation of squamous and ductal epithelia. However, unlike the classical SP, both epithelia were growing under the mucosal surface. As a result, and contrasting markedly with

classical SP, it did not manifest as an exophytic proliferation, but as a submucosal nodular swelling. We excluded squamous papilloma, inverted ductal papilloma, intraductal papilloma, and mucoepidermoid carcinoma, the main entities in the differential diagnosis of SP [3], and concluded it to be a SP with inverted pattern. In other words, our case was to an SP which grew inward, probably representing a rare variant. In addition to the striking architectural features of this case, it is worth noting that it occurred in a patient in an age group not usually affected.

As with SP, conservative local excision was the treatment of choice, although follow-up at regular intervals is required once there is a small percentage of recurrent cases of classical SP [3].

Conclusions

SP with inverted pattern should also be considered in the differential diagnosis of oral submucosal/endophytic masses.

To the best of our knowledge, this is the first case of SP with inverted pattern to be reported in the English-language literature.

Conflicts of interest

The authors declare that they have no conflicts of interest.

Acknowledgements

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