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SIALADENOMA PAPILLIFERUM: THE FIRST REPORTED LARYNGEAL CASE

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ABSTRACT

Sialadenoma Papilliferum (SP) is a benign salivary gland tumor characterized by an exophytic and endophytic epithelial proliferation of mucosa of salivary gland origin. Typically, these tumors appear on the palate, with most cases involving the hard palate. Several cases have been described, on the soft palate, buccal mucosa, tongue and bronchi. We present the first documented case of laryngeal SP, associated with a previous history of recurrent laryngeal papilloma. Our case report is unique in two ways. Firstly, it is the first case reported in the larynx. Very few cases along the lower respiratory tract have been reported, but none on the vocal folds. Secondly, it is the first case associated with a previous history of squamous papilloma. High level of alertness is required in order to identify this rare diagnosis.

INTRODUCTION

Sialadenoma Papilliferum (SP) is a benign salivary gland tumor characterized by an exophytic and endophytic epithelial proliferation of mucosa of salivary gland origin(1). It was first described by Abrams and Flinck in 1969 (2) and although the first published report involved the parotid gland, most case reports since then describe intraoral lesions. They are probably the rarest of the salivary gland pathologies comprising 0.4% - 1.2% (2–5) of all minor salivary gland lesions. The SP typically manifests as a painless, exophytic papillary growth that is often interpreted clinically as a squamous papilloma. From a pathological aspect, squamous papilloma is composed entirely of squamous epithelium and lacks the endophytic growth pattern and glandular differentiation of SP (6).

The origin of the tumor is not clear. Abrams and Flick suggested that they derive from myoepithelial cells (2), but other authors have proposed that blocked glandular ducts cause hyperplasia (7). The fact that the lesion rarely exceeds 1 cm in size advocates for the hyperplastic nature. Nakahata et al. suggested that the tumor may arise from intercalated ductal cells (8). Gomes et al. performed an immunohistochemical study which revealed a strong positivity of the tumor cells to CK7 and CK8 (9). According to the authors this fact suggests a more distal origin.

Typically, SP appear on the palate, with most cases involving the hard palate. Apart from the first described case, three additional cases have been described affecting the parotid gland (9–12), but Fowler and Damm argue that these cases probably represent two examples of papillary cystadenoma and one example of Warthin tumor (13). These authors, attempted a review of all published SP cases and concluded that the palate is the most common anatomic site, with 80% arising from this site. Most of these cases were on the hard palate, followed by the junction of the hard and the soft palate, the soft palate itself, and buccal mucosa. In their review, the average age was 56.4 years and the male-to-female ratio was 1.7 to 1. One case of SP of the tongue has also been described in the international literature (14). Some rare cases have also been reported arising from the nasal cavity and nasopharynx (15–17).

Four cases of SP arising from the bronchi have been described (14–21)and these are the only cases of SP described in the lower respiratory tract. To our knowledge, no published data exist reporting a case of laryngeal SP.

Case Report

We present the first documented case of laryngeal SP. A 66 old female patient with symptoms of dysphonia underwent microlaryngoscopy, biopsy of an exophytic laryngeal lesion on the anterior aspect of the left vocal cord and LASER excision. The preliminary clinical diagnosis was papilloma and the histological analysis confirmed that. The patient had to undergo eight sessions of microlaryngoscopy in a period of five years because of recurrence of the lesion. In each case, a biopsy was taken, and the lesion was excised by use of CO_2 LASER. All biopsies confirmed laryngeal papilloma. In situ hybridization performed in one of these biopsies was negative for both low and high risk HPV subtypes. Her ninth biopsy for a lesion excised in 2018 from the anterior left vocal fold revealed the unusual diagnosis of SP.

The excised lesion, microscopically, demonstrated mixed exophytic and endophytic epithelial lesion, with alternating stratified squamous and columnar epithelium (ciliated and non-ciliated) lining the papillary projections, and proliferating ductal epithelium lining the endophytic component. Numerous scattered mucocytes were also present. Cystic degeneration was seen in the squamous areas. Scattered apoptoses and some koilocyte-like cells were identified in the squamous epithelium.

DISCUSSION

In our patient, the two pathological entities, that of squamous papilloma and that of SP co-existed in the lesion. During our microlaryngoscopy sessions, a biopsy is taken, and the rest of the lesion is vaporized by the use of CO_2 LASER. It is possible that an element of SP was previously present, but previous biopsies failed to include that element in the specimen. Information regarding the sizes of the prior biopsies were not available. Also, it was not possible to review all prior papillomas biopsies.

There has been no association of SP with Human Papillomatosis Virus (HPV) infection in the past and it is not clear whether the co-existence of the two pathological entities in our patient is incidental. Argyres and Golitz did perform polymerase chain reaction for HPV DNA on a case of SP with negative results (22). In situ hybridization performed in previous biopsies of our case was negative for both low and high risk HPV subtypes. Previous specimen were re-examined after the new diagnosis, but the diagnosis of squamous papilloma was not challenged.

In the literature, the recurrence of SP after assured conservative surgical excision is rare, with only 7.4% of the cases in the literature with follow-up information reporting a recurrence (13). Our patient demonstrated several recurrences of her squamous papilloma before eventually, the diagnosis of SP was made. We believe that the recurrence is attributed to the element of squamous papilloma and there has been no recurrence of the lesion to date.

Sialadenoma Papilliferum is generally considered a benign lesion. Solomon et al. described a case of "malignant SP" because of the size of the tumor, multiple recurrences and lymphnode metastasis (23). Several other authors have described cases of SP with potentially malignant features (20–27). These cases have ambiguous histopathological features and either the diagnosis of SP, or their malignant potential have been challenged. Our case did not demonstrate any malignant features.

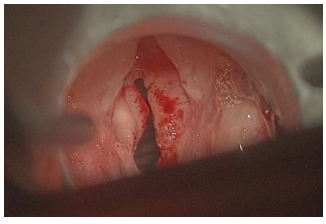

Figure 1. Macroscopic iage of Sialadenoma Papilliferum

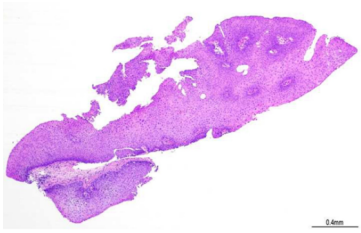

Figure 2. Squamous Papilloma (2016 biopsy)

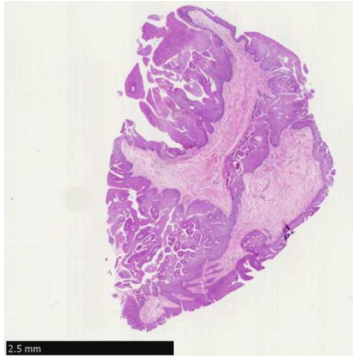

Figure 3. Sialadenoma Papilliferum (HE x 1.25, 2018 biopsy)

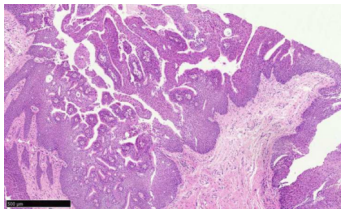

Figure 4. Sialadenoma Papilliferum HE x 5

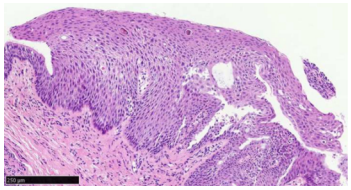

Figure 5. Sialadenoma Papilliferum HE x 10

CONCLUSION

Our case report is unique in two ways. Firstly, it is the first case reported in the larynx. Very few cases along the lower respiratory tract have been reported, but none on the vocal folds. Secondly, it is the first case associated with a previous history of squamous papilloma. Although the co-existence of the two pathological entities may be incidental, this discovery is very interesting. Given the rareness of the finding, it is possible that similar cases are misdiagnosed as squamous papilloma, or an element of co-existing SP is present in more than this case. High level of alertness is required in order to identify this rare diagnosis.

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