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Basal cell adenoma mimicking a mucocele of the lower lip- A rare occurrence

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Abstract

Basal cell adenomas are slow-growing and painless masses that account for approximately 1 to 2% of salivary gland adenomas. 70% of basal cell adenomas occur in the parotid gland, and the upper lip is the most common site for basal cell adenomas of the minor salivary glands. Here we present a very rare case of basal cell adenoma of the lower lip mimicking a mucocele.

Keywords: Mucocele, lower lip, basal cell adenoma, a rare occurrence

Case Description

A 22-year-old girl reported with an asymptomatic swelling on the inner aspect of the lower lip since 1 month. The patient gave a history of swelling which was insidious in onset, initially smaller in size, and gradually progressed to the current size. The swelling was not associated with any pain. The patient gave a history of discomfort while speaking as the swelling contacts the lower anterior teeth and also gave a history of lip bite in the region. There was no significant medical history. On intra-oral examination, a single, solitary, sessile, round swelling was noted on the right side of the lower labial mucosa measuring approximately 0.5x 0.5cm. The surface appeared to be keratinized. The swelling was soft in consistency and non-tender. No other oral anomalies were detected. Based on the clinical features and history of lip biting habits, the lesion was diagnosed as a mucocele. It was treated under local anesthesia using a scalpel by placing an incision circumferentially. The lesion was resected from the base and sent for

histopathological examination. Suture removal was done after 1 week. Microscopic examination of H & E stained lesion tissue showed multiple islands of epithelial cells supported by scanty connective tissue stroma. The peripheral cells were palisaded, hyperchromatic and columnar. The central cells showed pale staining nuclei with no cellular atypia. Lesional stroma showed various areas of hyalinization and fibrosis. Areas of salivary glands showing reactive changes and ductal hyperplasia were noted. Mixed inflammatory infiltrates predominantly lymphocytes were noted which was diagnosed as basal cell adenoma of the lower lip. On the 6-month follow-up, there was no history of recurrence of the lesion.

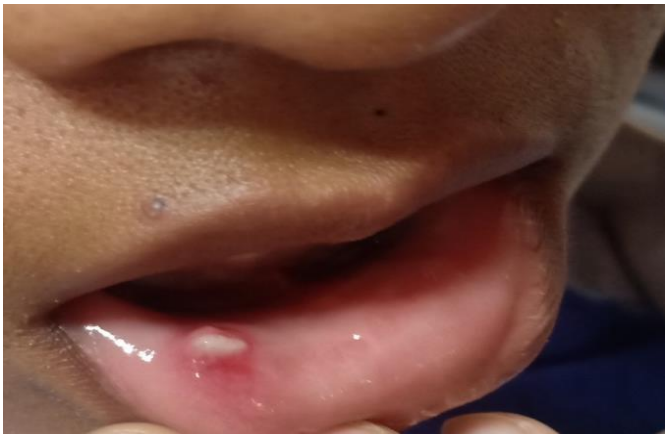

Figure 1

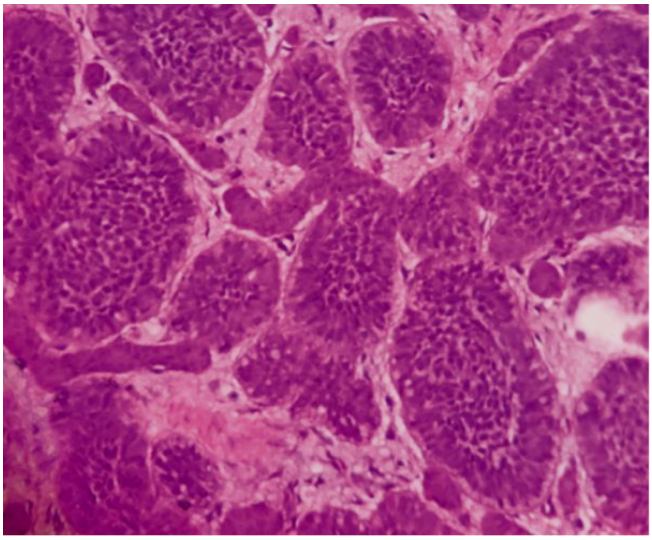

Figure 2

Introduction

Basal cell adenoma (BSA) is a rare benign neoplasm. It arises from the ductal epithelium of a salivary gland that is commonly present in the parotid gland. It rarely occurs in the oral region, such as the lip.[1] In 1967, Kleinsasser and Klein introduced the term “basal cell adenoma” which describes a benign salivary gland tumor comprised of uniform appearing basaloid cells arranged in different patterns such as solid, trabecular, tubular, and membranous. It has been recognized as one of the nine subcategories of salivary gland adenomas in the Second Edition of the Salivary Gland Tumors Classification of the World Health Organization (WHO). It accounts for approximately 1 to 2% of salivary gland adenomas. It is also called monomorphic adenoma as it lacks the chondroid mesenchymal and myxoid components seen in pleomorphic adenoma.[2]

No gender difference was noted in the incidence of oral BSA. In contrast, BSA arising from the parotid gland, females had a slightly higher predilection than males, with a male-to-female ratio of 1.0:1.5.[1] 70% of basal cell adenomas occur in the parotid gland, and the upper lip is the most common site for basal cell adenomas of the minor salivary glands.[3] Clinically, BSA may resemble a mucocele of the oral mucosa, but the mucocele usually appears in the lower lip of young people, whereas BSA appears in the upper lip of the elderly.[4] They are generally slowly growing, well-demarcated masses which are not fixed to the underlying structures.[1] The tumors are characterized by slow growth and are usually painless. They occur chiefly in adults, the average age of the patients is 57.7 years with the peak of incidence seen in the sixth decade. The tumor is treated by excision and has a high recurrence rate in the parotid gland but recurrence of oral BCA is rare if the tumor is resected completely.[1]

It presents with a good prognosis. The malignant transformation is rare among BSA but more common in the membranous type (around 4.3%).[4] It is most commonly seen in cases of basaloid type carcinoma, adenoid cystic carcinoma, basaloid carcinoma, or carcinoma ex monomorphic adenoma and basal cell adenocarcinoma. Kusafuka et al[6] reported a case of salivary duct carcinoma arising from a BSA of the parotid gland.

Mucocele is a mucus-filled cyst which is caused by the accumulation of saliva at the site of a traumatized or obstructed minor salivary gland duct. Mucoceles often present as painless, discrete, smooth-surfaced swellings which are usually asymptomatic but sometimes can cause discomfort by interfering with mastication, speech ,or swallowing.[7] Mucoceles are classified as retention and extravasation types. Mucous retention cysts are more commonly located on the palate or the floor of the mouth while extravasation mucoceles most frequently occur on the lower lip, where trauma is common. Surgical excision is the primary treatment for mucoceles, particularly to prevent a recurrence.[3]

In this case report, a 22-year-old female has been diagnosed with BSA clinically presenting as a mucocele in the lower lip which is a rare occurrence as it is seen more commonly in the adult population and occurs more prevalently in the parotid gland and the upper lip in minor salivary glands.

Discussion

In the present case report, we have described BSA of the lower lip in a young female patient. BSAs are often located in the parotid gland (superficial lobe) and rarely occur in the minor salivary glands[2] and when they do occur, they preferentially occur in the upper lip.[8] Tatehara et al reported a case of BSA occurring in minor salivary glands of the upper lip.[1] Similar cases were reported by Kudoh et al[9] and Karim et al[8] Apart from the upper lip, BSA has also been reported to occur in palatal mucosa. Gupta et al[2] reported a case of BSA occurring in the palate. This is the only case reported in literature where BSA has occurred in the lower lip.

BSA mostly affects older individuals as reported by Tatehara et al[1]. Similar cases were reported by Kudoh et al[9] and Karim et al[8] but in our present case, the patient is a young individual of 22 years. Gupta et al[2] has reported a similar case of a 32-year-old diagnosed with BSA.

In our present case, BSA has been shown to mimic mucocele clinically. Mucoceles most commonly occur in the lower lips of young patients (second and third decades). It has no gender predilection.[9] BSA can often be confused clinically with mucocele seen in 22% of cases, pleomorphic adenoma, and hemangioma. Some reports showed preoperative diagnoses of minor salivary gland tumors, including pleomorphic adenoma in 44% mucocele 22%, and vascular neoplasms in 11%. [1]

Two cases were reported by Antoniades et al[10] where the coexistence of mucous retention cysts and basal cell adenomas arising from the lining epithelium of the cyst was reported. It was further hypothesized that it was possible that the basal cell adenomas might have been developed from the lining epithelium of preexisting mucous retention cysts.

There have been reports of salivary gland tumours mimicking mucoceles. Madhavan NR et al[5] reported a case of canalicular adenoma of the upper lip mimicking a mucocele. A similar case was reported by Ata et al[11] of a parotid mucoepidermoid carcinoma mimicking a large mucocele. Another case was reported by Melo et al[12] of a mucoepidermoid carcinoma mimicking a ranula in the floor of mouth. In our cases, it is possible that the mucous retention cysts were developed due to

partial obstruction secondary to the presence of basal cell adenomas arising from excretory ducts.

Since benign salivary gland tumors cannot be discriminated clinically from malignant tumors, an early excision of all cystic tumors resembling mucous retention cysts is mandatory.[10]

Conclusion

This case report describes BSA in a very rare site, that is, the lower lip which is reported for the first time in literature. Hence, while considering the differential diagnosis for lesions of the lower lip, BSA should also be included and confirmed with histopathological examination.

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