CASE REPORT

Tubular basal cell adenoma of submandibular salivary gland: A rare case report with an unusual presentation

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Abstract
Basal cell adenoma is an uncommon salivary gland tumor. Its occurrence is mainly noted in the parotid gland and rarely in the submandibular gland. Owing to therapeutic and prognostic implications, differential diagnosis with adenoid cystic carcinoma is mandatory. The tubular variant is the least common histologic type and its occurrence in the submandibular salivary gland makes this case interesting to document.

Keywords
Adenoid cystic carcinoma, basal cell adenoma, submandibular salivary gland

Introduction
Basal cell adenoma (BCA) is a rare neoplasm of the salivary gland, which occurs in the sixth and seventh decades of life. Histologically it shows basoid morphology of the tumor cells. It chiefly occurs in the parotid gland and upper lip, its occurrence in the submandibular gland has been infrequently reported.[1]

The incidence of BCA is only 1% of all salivary gland tumors.[2]

It usually presents as a slow-growing, asymptomatic, freely mobile mass which is usually <3 cm in diameter and well circumscribed.[3]

Histologically four characteristic patterns have been described as solid, trabecular, tubular and membranous. It is characterized by the presence of a basoid cellular layer with a stockade pattern bounded by a hyaline substance.[4]

Differential diagnosis with adenoid cystic carcinoma and basoid squamous cell carcinoma has to make due to their close resemblance as the treatment and prognosis varies markedly.

Case Report
A 40-year-old male visited the dental clinic with a chief complaint of a swelling in the left side of the jaw for the past 3 months.

On examination, a well-defined swelling was seen on the left side of the jaw in the submandibular region measuring 4 cm × 3 cm; it extended from the inferior border of the mandible to 3 cm below it. Antero-posteriorly it extended from the submental region to the anterior region of the ramus. On palpation, the swelling was non-tender, immobile and firm to palpate.

A provisional diagnosis of pleomorphic adenoma was arrived at unilateral lymphadenopathies were also considered.

The lesion was excised under general anesthesia and the tissue was subjected to histopathological examination.

Microscopic examination showed a capsulated lesion with basoid cells arranged in a cribriform pattern.

Proliferation of basoid cells was seen in the form of ducts with eosinophilic coagulum and areas of hyalinization [Figure 1a and 1b]. These features were indicative of an adenoid cystic carcinoma.
When serial sections were closely analyzed the proliferating strands and cords were lined by an inner layer of cuboidal cells and a peripheral layer of tall columnar cells. Centrally located round pale staining cells were also observed [Figure 2a and 2b]. Tumor nests were clearly delimited from the surrounding stroma and a sharp demarcation between the neoplastic epithelial cells and the surrounding connective tissue was evident [Figure 3]. These features are absent in adenoid cystic carcinoma.

A final diagnosis of tubular BCA was concluded. A follow-up of 1 year revealed no recurrences.

Discussion

BCA of the salivary glands is a rare tumor, first described by Kleinsasser and Klein in 1967. Batsakis was credited with reporting the first case in the American literature in 1972, and suggested that the intercalated duct or reserve cell is the histogenetic source of the BCA. The Salivary Gland Tumors Classification of the World Health Organization recognized it as an independent entity.[5]

The commonest location is the parotid gland. Other less common sites are the palate, lips and buccal mucosa, respectively. Occurrence in the submandibular gland is extremely rare.[6] The occurrence is usually in the sixth and seventh decades of life.

A female predilection has been noticed.[7] The present case occurred in a male patient in the third decade and in the submandibular salivary gland, which is a rare location for this tumor.

Histopathologically, four patterns have been described namely solid, trabecular, tubular and membranous. In the solid pattern the basaloid cells form islands and cords that have a broad, rounded, lobular pattern. These cells are sharply demarcated from the connective tissue stroma by basement membrane. The trabecular type has the same cytological features as the solid type, but the epithelial islands are narrower and cord like and are interconnected with one another, producing a reticular pattern. The membranous type is a distinct variant of BCA characterized by the presence of abundant, thick, eosinophilic hyaline layer which surrounds and separates the epithelial islands. Electron microscopy has shown that this hyaline material is reduplicated basement membrane. The tubular variant exhibits multiple small, round duct like structures. These tubules are lined by two distinct layers of cells, with inner cuboidal ductal cells surrounded by an outer layer of basaloid cells. It is also characterized by the presence of a basaloid cellular layer with a stockade pattern and bounded by a hyaline substance with the absence of myoepithelial cells. The present case was diagnosed as a tubular variant, which is the least common variant encountered.[8] The tubular variant is the least common type.

Differential diagnosis from similar appearing neoplasms is a diagnostic dilemma. Adenoid cystic carcinoma shows the most striking histologic similarity to the BCA.[9]

Differentiating this lesion from adenoid cystic carcinoma is of prime importance since the treatment and prognosis for benign and malignant salivary gland tumors differ drastically.

The basal cell adenocarcinoma is another malignant tumor that shares histologic features with the BCA. Both exhibit myoepithelial differentiation and reactivity patterns indicative of the ductal epithelium. Basal cell adenocarcinoma is distinguished from BCA by the histologic features of invasion, mitotic activity, and neural or vascular involvement.[10]

The present case did not show any invasion, mitotic figures, neural or vascular invasion. Features suggesting high-grade
malignancy were not observed, and findings are primarily consistent with a benign neoplasm.

Treatment of BCA is simple surgical excision. Recurrence is rare. So, pre-operative histopathologic diagnosis is very useful for preventing aggressive therapy such as post-operative radiotherapy in case of adenoid cystic carcinoma.

**Conclusion**

BCA of the salivary gland is a rare neoplasm consisting of a monomorphic population of basaloid epithelial cells. Four distinct histopathological types are recognized with the tubular variant being the least common. A rare case of tubular BCA occurring in the submandibular salivary gland has been reported here, which is rare and unusual in presentation. Due to therapeutic and prognostic implications it should be differentiated from adenoid cystic carcinoma which can be a challenge to the pathologist.

**References**
