Case Report
Basal cell adenoma in the parotid: a bizarre myoepithelial-derived stroma rich variant

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Abstract: Basal cell adenoma (BCA) is a specific entity that lacks the myxochondroid stromal component of a pleomorphic adenoma. There are six histopathological types of BCA: solid, tubular, trabecular, membranous, cribriform, and myoepithelial-derived stroma rich. Myoepithelial-derived stroma rich variant is so rare, especially with cellular atypia. Herein we describe a rare case of BCA arising in the parotid on a 25-year-old man. A well-demarcated nodule arising in the parotid that was composed of basaloid cells, forming small duct-like or tubular structures containing basement membrane-like material, as well as highly cellular elongated cells with hyperchromatic, enlarged, pleomorphic, and bizarre nuclei. Immunohistochemically, S100 protein and p63 highlighted the basal aspect of the peripheral epithelial cells and peripheral spindle and bizarre cells, while CK7 expressed on the luminal cells. We made a diagnosis of “basal cell adenoma, myoepithelial-derived stroma rich variant, with bizarre myoepithelial proliferation”. The differential diagnosis includes cellular pleomorphic adenoma, basal cell adenocarcinoma, and carcinoma ex pleomorphic adenoma. After follow-up for 3 years, there was no evidence of recurrence. Further pathological characteristics of this disease are discussed.

Keywords: Basal cell adenoma, bizarre, myoepithelial, parotid

Introduction
Basal cell adenoma (BCA) was first described by Kleinsasser and Klein in 1967 [1], composed of cells with a basaloïd appearance, lacked the myxochondroid stromal component of a pleomorphic adenoma, which accounts for 1% to 2% of all salivary gland epithelial tumors. Less frequently, these tumors may be found in especially the upper lip and buccal mucosa, with the upper lip being the most common site. But BCA, myoepithelial-derived stroma rich variant, with bizarre myoepithelial proliferation was rarely reported reviewing the English literature [2]. We report a rare case of myoepithelial-derived stroma rich variant basal cell adenoma arising in the parotid, associated with bizarre myoepithelial proliferation, and discuss the similar entities.

Case report
A 25-year-old man was referred to our hospital for evaluation of a lesion of the right parotid. A physical examination revealed a hard-circumscribed nodule on his right parotid. Computed tomography showed a round, well-circumscribed heterogeneous mass measuring 2.8 × 2.3 cm in the right parotid gland. No calcification or cystic component was seen in the tumor. The mass was excised and submitted for the pathologic examination.

Pathological findings
Microscopically, low power microscopy illustrates the lesion was well encapsulated with fibrous tissue, which was arranged in anastomosing trabecular cords associated with sheets of spindle cells (Figure 1A). Frequently centrally located duct-like structures were observed. There was no cartilage formation, mucous stroma or necrosis in the tumor. The tumor comprised basaloid cells with nuclear palisading at the periphery and spindle cells emerged with glandular architecture. These basaloid cells were small, with scant cytoplasm and dense basophilic nuclei, which was surrounded by a
thick, eosinophilic basement membrane, on low magnification, appeared as pieces of a jigsaw puzzle at some areas (Figure 1B), but arranged in a Swiss cheese pattern similar to an adenoid cystic carcinoma at other sites. Glandular lumens were delineated from the "stroma," which consisted of highly cellular areas and bizarre cells at some foci (Figure 1C). What’s more, some bizarre and dense hyperchromatic cells were present at the periphery of basaloid architectures (Figure 1D). Mitotic figures were extremely rare. Immunohistochemically, the inner cells of the tubular components were positive for pan-cytokeratin (Figure 2A), cytokeratin 7 (Figure 2B), but negative for smooth muscle actin (SMA), calponin and S-100 protein, the peripheral cells were negative for CK7, positive for SMA (Figure 2C), and weakly positive for pan-cytokeratin and S100 (Figure 2E). Peripheral spindle cells and bizarre cells were highlighted by p63 (Figure 2D). The MIB-1 label of the spindle and basaloid cells was lower than 1% (Figure 2F).

Discussion

The term BCA was first described by Kleinsasser and Klein in 1967. BCAs are divided into 6 types of growth patterns: solid, trabecular, tubular, membranous, cribriform, and myoepithelial-derived stroma rich [3]. The most common type is the solid variant. According to previ-
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Figure 2. Immunohistochemical stains of the basal cell adenoma: A: The inner cells of the tubular components were positive for pan-cytokeratin. B: The inner cells of the tubular components were positive for cytokeratin 7. C: The peripheral cells were positive for SMA. D: P63 highlighted the basal aspect of the peripheral epithelial cells and spindle cells. E: S100 weakly expressed on the peripheral cells. F: The MIB-1 label of the spindle and basaloïd cells was lower than 1%.

Basal cell adenomas were originally classified as a type of monomorphic adenomas. But several studies have documented that the myoepithelial cells participate in the histogenesis of basal cell adenomas [2, 4-9]. BCA was a specific entity occurred at many sites, which had a distinct characteristic, different from pleomorphic adenoma and myoepithelioma. The precise cell of the origin of BCA has been previously studied by various methods. Most authors thought that BCA forms part of a spectrum of benign salivary adenomas that also includes pleomorphic adenoma, and that the ends of this spectrum are canalicular adenoma and myoepithelioma. We know the BCA could have myoepithelial cells. The component of BCA might be same to pleomorphic adenoma, except that pleomorphic adenoma has myxochondroid stroma. However, if neoplastic myoepithelial cells proliferated, how to make the distinction between pleomorphic adenoma and basal cell adenoma with prominent myoepithelial cell proliferation? It might be possible that some basal cell adenomas could develop a cellular myoepithelial component that contains tumor cells histogenetically and morphologically similar to pleomorphic adenoma, or some BACs with prominent myoepithelial cells may be pleomorphic adenoma.

Our case of BCA was unique that the lesion was composed of typical tubular, and cribriform type BCA, and also prominent spindle cells and bizarre cells around the glandular architectures, most exclusively at the peripheral areas and fascicles areas. The bizarre and spindle cells were typical positive for p63 and SMA, also weakly positive for S-100, which were typical immunophenotype of myoepithelial cells. Our case might be consistent with the hypothesis which was raised by Dardick et al [2] that it was possible that some basal cell adenomas could develop a cellular and nonmyxoid “stroma”. Prominent myoepithelial cell neoplastic proliferation, associated with typical BCA microscopical appearance, might suggest that it is a hybrid type between basal cell adenoma and myoepithelioma, and might be a variant type of pleomorphic adenoma.

Regezi and Batsaki [10] thought that the intercalated duct cells give rise to the acinar cells, other intercalated duct cells striated duct cell and myoepithelial cells. Most of the salivary gland tumors are considered to originate from...
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the intercalated duct cells [11]. We agree with Chaudhry et al [12] that undifferentiated stem cells possess an inherent ability to differentiate into myoepithelium and other cells. Our case might be explained by a phenomenon of multidirectional differentiation involving the stem cells. If the tumor purely composed of spindle myoepithelial cells, it defines as myoepithelioma; if it composed of spindle myoepithelial cells and also glandular epithelium at the myxochondroid stromal backgrounds, it might be sorted as pleomorphic adenoma. If both elements were neoplastic hyperplasia, it might be a hybrid tumor just like our case. Just like adnexal tumor, BCA might form part of a spectrum of benign salivary adenomas, which might be related to pleomorphic adenoma or be an early developmental phase of this tumor.

Myoepithelial-derived stroma rich variant of basal cell adenoma was so rare. As far as my knowledge, this case is the first case of myoepithelial-derived stroma rich BCA with prominent spindle cells and bizarre cells. There is limited experience about its prognosis. After 3 years follow-up, no recurrence has been detected in this case.

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Disclosure of conflict of interest

None.

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