A rare tumor of salivary gland: Diagnostic Dilemma on fine needle aspiration cytology

ABSTRACT
Salivary gland cytopathology is a diagnostically challenging area with overlapping cytomorphological features between benign, malignant, and metastatic tumors. We report the case of a 45-year-old male who presented with two swellings in right retroauricular and infraauricular region along with a palpable single right cervical lymph node. On ultrasonography of the neck, a possibility of malignant lesion was given. Contrast enhanced computed tomography of the head showed a large well-defined space occupying lesion in right temporoparietal region eroding the skull bone with both extra and intracranial extension. Fine needle aspiration was performed from both swellings and cervical lymph node. Based on cytological features and clinicoradiological examination, a possibility of metastasis from epithelial malignancy (adenocarcinoma) was suggested. The retroauricular region swelling was excised, and a diagnosis of salivary duct carcinoma was given on histopathology. In this article, we discuss the diverse presentation, cytomorphological features, and differential diagnosis of this rare salivary gland tumor.

Key words: Fine needle aspiration cytology; retroauricular and infraauricular; salivary duct carcinoma

Introduction
Fine needle aspiration (FNA) of salivary gland represents one of the most challenging areas of cytopathology due to a wide variety of reactive and neoplastic lesions. Pathologically, salivary gland tumors have been described as one of the most heterogeneous groups of human tumors. There is marked cytomorphologic diversity and overlap between benign, malignant, and metastatic tumors.

Case History
A 45-year-old male complained of bloody discharge from the right ear with progressive weakness of the right side of the face since 2 months (right facial nerve palsy). On examination, a diffuse, firm swelling measuring 4 × 3 cm was present in right retroauricular region since 1 year which was fixed to the underlying occipital bone. Another swelling was present in the infraauricular region since 3 months measuring 2 × 1 cm which was well-defined, globular, soft to firm, with glistening red surface. Patient also had right cervical lymphadenopathy. Ultrasonography (USG) of the neck showed a well-defined mass in posterior auricular region, eroding the mastoid bone, and a possibility of malignancy was given. Few enlarged lymph nodes with normal thyroid and bilateral salivary glands were seen. Contrast enhanced computed tomography (CECT) of the head and neck showed similar mass in right temporoparietal region with both extra and intracranial extension.

Charu Agarwal, Manju Kaushal, Minakshi Bhardwaj
Department of Pathology, PGIMER, Dr. RML Hospital, New Delhi, India

Address for correspondence: Dr. Charu Agarwal, PGIMER, Dr. RML Hospital, New Delhi, India. E-mail: dr.charu.ag@gmail.com

How to cite this article: Agarwal C, Kaushal M, Bhardwaj M. A rare tumor of salivary gland: Diagnostic Dilemma on fine needle aspiration cytology. J Cytol 2017;34:107-9.
FNA was performed from both right retroauricular and infraauricular swellings along with lymph node. Blood mixed cellular aspirate was obtained. Cytological smears from both the swellings were stained with Giemsa and Papanicolaou stains and showed similar morphology. Smears were highly cellular with cells arranged in large flat sheets, three-dimensional papillae, and cohesive clusters [Figure 1a, inset]. At places, adenoid pattern was seen. Cells were round-to-oval showing mild-to-moderate nuclear pleomorphism, low nuclear–cytoplasmic ratio, eccentric nucleus, prominent nucleolus, and granular abundant cytoplasm [Figure 1a]. Few cells showed vacuolated cytoplasm with blood in the background. Smears from enlarged cervical lymph node showed tumor cells with similar morphology.

In view of the normal salivary gland on USG, absence of normal salivary gland tissue on smears and presence of pleomorphic round cells arranged in papillae and adenoid pattern with bony and cranial extension, a possibility of metastasis from epithelial malignancy (adenocarcinoma) was reported on cytology. An excision biopsy with immunohistochemistry was advised.

Magnetic resonance imaging (MRI) of the brain, reported later, showed lytic destruction of the right occipitotemporal bone with intracranial component. Another lesion was seen in the auricular region, which was inseparable from the deep lobe of the right parotid gland. A possibility of neoplastic lesion was given.

Patient was taken up for surgery and the retroauricular region swelling was excised. However, the infraauricular swelling that was adherent to the underlying deeper structures was not excised. The tumor was hard, vascular, extradural, adhered to dura, and involved the temporal bone.

Subsequently, a single soft-to-hard tissue mass measuring 9 × 6 × 3 cm was received. External surface was hemorrhagic. The cut section was gray white with multiple pinpoint cystic areas. The undersurface was bony hard.

Sections stained with hematoxylin and eosin (H and E) showed a partly encapsulated tumor with cells arranged in solid-cystic, glandular, and cribriform patterns [Figure 1b inset]. The cysts were lined by single-to-multilayered (2–3 cells) apocrine-like cells and lumen showed mucin and muciphages [Figure 1b]. These cells had abundant eosinophilic granular cytoplasm, eccentric nucleus, vesicular chromatin, and prominent nucleolus. Both typical and atypical mitosis were noted. Tumor cells showed infiltration into the bone. The resection margins were free of tumor. Based on the histomorphological features, the following differentials were considered: Salivary duct carcinoma, oncocytic carcinoma, low grade cribriform cystadenocarcinoma, mucoepidermoid carcinoma, and metastatic adenocarcinoma.

Mucicarmine and an immunohistochemistry (IHC) panel comprising of cytokeratin (CK) 7, CK20, panCK, epithelial membrane antigen (EMA), thyroid transcription factor (TTF-1), HER2/ neu, S100, smooth muscle actin (SMA), carcinoembryonic antigen (CEA), and p63 were performed. Mucicarmine was negative excluding mucoepidermoid carcinoma. CK7 and EMA were positive [Figure 2a and b] whereas the rest of the markers were negative.

In view of the presence of cysts lined by multilayered cells along with cribriform pattern, possibility of oncocytic carcinoma was excluded. Possibility of metastasis was excluded on the basis of immunonegativity for CK20, CEA, and TTF-1. Low grade cribriform cystadenocarcinoma was excluded because cysts were multilayered with apocrine-like cells and markers like S-100 and SMA were negative. A final diagnosis of salivary duct carcinoma was given and the patient was referred for radiotherapy.

Discussion

Salivary duct carcinoma is a distinctive primary neoplasm of salivary gland, which was first described by Kleinsasser et al. in 1968.[2] Approximately 200 cases have been reported in the English literature. It is a rare tumor with an estimated
incidence of 1–3% of all malignant salivary gland tumors.[3] Nearly 85% of the cases occur in the parotid gland followed by submandibular gland.[2] Three-fourth of the patients are males, with peak incidence in sixth to seventh decade. Twenty-five percent of the patients exhibit symptoms related to facial nerve involvement. Lymph node metastasis are common. This tumor is considered to be high grade, with mortality up to 70%.[4] In the present case, facial palsy and lymph node metastasis were present, however, the patient was relatively young. Normal salivary gland tissue and necrosis were absent.

Histomorphologically, this tumor shows similarities with intraductal cribriform and comedocarcinoma of the female breast, thus, known as “salivary duct carcinoma.”[3,5,6] Understanding this entity is necessary to avoid false interpretation.

The cellular yield varies from low to high, depending on the degree of desmoplasia and necrosis. The cells are arranged singly in loosely cohesive groups, three-dimensional clusters, flat sheets, papillae, and cribriform pattern. On account of its apocrine features and high nuclear grade, several primary and metastatic neoplasms are included in the differential diagnosis.[2] Nevertheless, if cribriform groups are noted in a salivary gland aspirate, the diagnosis of salivary duct carcinoma should at least be considered.[7] However, in the present case, cribriform pattern was not noted cytologically. The absence of cribriform or papillary groups suggests an inconclusive diagnosis and the need to establish a differential diagnosis with other salivary gland tumors, in particular with adenocarcinoma not otherwise specified and high-grade mucoepidermoid carcinoma.[8]

Despite the fact that the histomorphologic features are always necessary for its characterization, several authors have reported that the cytopathological approach can establish the final diagnosis.[4] Treatment modalities are nonconsensual, however, some authors advocate the necessity of aggressive approach.[3]

**Conclusion**

To conclude, the preoperative diagnosis of this type of neoplasm may warrant a more extensive radiological work-up.[5] At present, FNA has gained wide acceptance as a first line procedure in the evaluation of salivary gland mass lesions.

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

There are no conflicts of interest.

**References**

1. Faquin WC, Powers C. Salivary Gland Cytopathology. New York, NY: Springer publishers; 2008. p. 1-16.
2. Kinnera VS, Mandyam KR, Chowhan AK, Nandyala R, Bobbidi VP, Vutukuru VR. Salivary duct Carcinoma of parotid gland. J Oral Maxillofac Pathol 2009;13:85-8.
3. Mlika M, Kourda N, Zidi Y, Aloui R, Zneidi N, Rammeh S, et al. Salivary duct carcinoma of parotid gland. J Oral Maxillofac Pathol 2012;16:134-6.
4. Valeri RM, Hadjileontis C, Skordalaki A, Pandidou A, Vahtsevanos C, Destouni H. Salivary duct carcinoma of the parotid gland: Report of a rare case with a comparative study of aspiration cytology and histomorphology. Acta Cytol 2005;49:61-4.
5. Elsheikh TM, Bernacki EG Jr, Pisharodi L. Fine-needle aspiration cytology of salivary duct carcinoma. Diagn Cytopathol 1994;11:47-51.
6. Khurana KK, Pitman MB, Powers CN, Korourian S, Bardales RH, Stanley MW. Diagnostic pitfalls of aspiration cytology of salivary duct carcinoma. Cancer 1997;81:373-8.
7. Gilcrease MZ, Guzman-Paz M, Froberg K, Pambuccian S. Salivary duct carcinoma. Is a specific diagnosis possible by fine needle aspiration cytology? Acta Cytol 1998;42:1389-96.
8. Gracia-Bonafe M, Catala I, Tarragona J, Tallada J. Cytologic diagnosis of salivary duct carcinoma: A review of seven cases. Diagn Cytopathol 1998;19:120-3.