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Case Report

Adenoid cystic carcinoma: A rare late presentation of the mobile tongue

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ABSTRACT

Adenoid cystic carcinoma (ACC) is an infrequent malignant neoplasm of the salivary glands. We present a case of a 70-year-old male patient with a swelling over the dorsal and ventral surface of anterior two third of the tongue which was causing him difficulty in mastication since 10 months. Ultrasound and magnetic resonance imaging were done following which the surgical excision of the lesion was performed and histopathological diagnosis of ACC was achieved. It was rare to find ACC in such an old man with such a large lesion presenting so late in the rare site of the mobile tongue. ACC is a slowly growing, highly invasive cancer with a high recurrence rate and chances of metastases, so surgery is the choice of treatment with mandatory long-term follow-up.

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1. Background

Adenoid cystic carcinoma (ACC) is an infrequent malignant neoplasm that represents approximately 1–2% of all malignant neoplasms of the head and neck and 10–15% of all malignant salivary gland neoplasms. It can originate in the minor or major salivary glands.¹ It is more common in females and characterized by slow indolent growth, affinity for nerve invasion and potential to produce distant metastases, mainly to the lungs and bones.² One of the least frequent sites of presentation is the mobile tongue, the incidence of only approximately 3% having being reported.³

2. Case report

A 60-year-old man presented with a growth on the tongue causing him oral discomfort and masticatory difficulties for the past 10 months. There was no significant medical history or addiction. On examination, a mass was noticed on the tongue extending antero-posteriorly from the tip to the posterior one third and involved both dorsal and ventral surfaces, measuring about 50 × 30 × 20 mm [Fig. 1]. It was larger on the right side, involving and crossing the midline. The mass was firm, of the same colour as that of the surrounding mucosa, had a smooth surface and was slightly

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Abbreviations: ACC, adenoid cystic carcinoma; FNA, fine needle aspiration; MRI, magnetic resonance imaging.

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Fig. 1 – Preoperative clinical presentation and MRI (axial view) of tongue.

tender on palpation. There was no evidence of cervical lymphadenopathy.

Ultrasonography was advised and reports were suggestive of haemangioma. Magnetic resonance imaging (MRI) revealed an ill-defined moderate sized soft tissue lesion with altered signal entities involving the dorsum of tongue with invasion suggestive of benign neoplasm (Fig. 1). Fine needle aspiration results were inconclusive. Laboratory blood investigations were within normal limits. With provisional diagnosis of schwannoma of tongue, the patient was planned for surgical excision of lesion under GA.

The patient was positioned supine and general anaesthesia was given through nasal intubation. Following aseptic measures, incision was placed over right lateral border of tongue from tip of tongue to the posteriorly till 2/3rd of tongue. After separating the dorsal and ventral surfaces of tongue, the lesion was identified and dissected superiorly inferiorly and medially. A lobulated firm mass with irregular borders was

identified with a thick posterior-inferior pedicle, subverting more inferiorly towards base of the tongue. The pedicle was dissected meticulously, ligated with 2-0 silk and divided with scalpel. Haemostasis was achieved. The specimen was tagged over margins and sent for histopathologic examination. Macroscopically, the mass was brown in colour with an irregular surface, measured $80 \times 40 \times 20$ cm and was firm in consistency [Fig. 2]. The histopathologic study showed multiple pseudocystic spaces of variable sizes surrounded by cuboidal cells with scarce cytoplasm and oval nuclei, filled with eosinophilic material and hence was diagnosed as cribriform ACC [Fig. 2]. There was no evidence of perineural infiltration on serial sections and the surgical margins were negative. Immunohistochemistry analysis showed expression of p16 which further confirmed the diagnosis (Table 1).

Post-operatively, the patient has been on regular follow-up to note for any recurrence and three years after treatment, he is well without any evidence of relapse.

Fig. 2 – Excised lesion with histopathology and postoperative view of tongue.

Table 1 – Table of latest reviews of ACC of tongue.

Name of author	Year	Observations on ACC of tongue
Fine et al.	1960	2 Cases, 2.5% among minor salivary gland neoplasms
Leafstedt et al.	1971	11 Cases on tongue among 56 ACCs
Spiro et al.	1974	26 Cases on tongue out of 171 ACCs
Main et al.	1976	3 Cases of ACC tongue of 112 minor salivary gland neoplasms
Isacsson, Shear	1983	1% ACC tongue of 201 minor salivary gland neoplasms
Eveson and Cawson	1985	3 ACC tongue out of 336 minor salivary gland neoplasms
Andersen et al.	1991	3% Cases out of minor salivary gland neoplasms
Spiro and Huvos	1992	19.8% On tongue, 85% of these at the base of tongue
Huang et al.	1997	10 Cases reported, 20.8% incidence
Cheuk et al.	1999	Poorly differentiated adenocarcinoma with cervical lymph nodes, bone and lung metastasis.
Emre Üstündag	2000	12 Year girl (youngest reported patient)
Spiro	2005	3.4% to 17.1%, 2.9% over mobile tongue
Jaber	2006	3 Cases, 4% incidence among minor salivary gland tumours
Ortiz and Barrios	2006	Case with carcinogenic embryonic antigen, epithelial membrane antigen, glial fibrillary acidic protein, Ki67 positive and recurrence in 2 years
Wang et al.	2007	20 Cases reported, 2.7% of total 737 minor salivary gland neoplasms
Eduardo C. S. Soares	2008	Base of tongue lesion with no metastases
Luna-Ortiz et al.	2009	Clinicopathological study and survival analysis in 8 cases
Gavin A. Falk	2011	Late metastasis to the pancreas
Jonas Osher	2011	Tongue base lesion treated with ultrasound-guided interstitial photodynamic therapy
Pavitra Baskaran	2012	Anterior dorsum of tongue
Toshitaka et al.	2013	p53 Abnormalities in low grade lesions
Sengupta et al.	2013	Dorsum of the tongue presentation in 45 year old
Jie Li et al.	2014	Development and characterization of salivary adenoid cystic carcinoma (ACC) cell line
Massimo Mesolella et al.	2014	Treatment of c-kit positive ACC of the tongue
Young Jo Sa	2014	Tongue ACC 27 months after successful pulmonary ACC resection
Sawmik Das et al.	2015	Liver metastasis

3. Discussion

The term ACC was first used by Spies in 1930. It is a malignant epithelial neoplasm of salivary gland origin was originally described by Robin and Laboulbène in 1853.⁴ It mostly affects adults in the 4th–6th decades and occurs more frequently in females.⁵ The frequency in which the tongue has been the site of origin of ACC ranges from 3.4% to 17.1%^{6,7} and 2.9% for the mobile tongue.⁶ Histopathologically, ACC may present in three different patterns: solid, tubular, and cribriform. Perineural invasion is common and has been reported in almost half of the cases. Necrosis and vascular invasion are present at a much lower rate.³

When located in the tongue, ACC is usually asymptomatic due to gradual submucosal growth, which may prevent early diagnosis, but such large size is rare. It is also rare to come across lesions such as these which pose as diagnostic dilemmas. Immunostaining with p16 is reported in literature for ACC and aids in diagnosis affirmation.⁸ Also, it is rare to find large-sized ACCs with late presentation as tumours located in the mobile tongue tend to cause profound functional alterations thus generally being diagnosed earlier.⁹

Due to the slow growth pattern of the tumour, there is much controversy regarding the treatment of ACC. However, due to local recurrence and late metastasis, surgery remains the mainstay of management with or without radiotherapy¹⁰

which is indicated in the presence of positive resection margins, infiltrative growth pattern or perineural invasion.¹

Although few authors state that the solid variants of the tumour have the worst prognosis, the histological classification is mostly not given benefit, and a correlation between microscopic appearance and prognosis is denied.⁷ Although our patient with cribriform pattern of ACC is alive and well after 36 months of surgery, it cannot be considered a definitive cure, due to the common occurrence of late metastasis.⁹ Most studies have shown that ACC has a bad prognosis and it is therefore necessary to carry out long-term follow-ups, irrespective of the therapeutic method applied for management.

4. Conclusion

Early diagnosis and proper treatment of this infrequent neoplasm of the tongue are important factors from a functional point of view, since ACC are slowly growing and may produce diffuse invasion. Surgery is still the cornerstone of treatment and radiotherapy is indicated in the presence of compromised surgical margins. Long-term follow-up is mandatory.

Consent

Patient's written informed consent has been obtained for publication of details and images.

Conflicts of interest

The authors have none to declare.

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