Ultrasound features of acinic cell carcinoma of the tongue: a rare case report

Dr. Tejashree Digambar Ghode
Department of Oral Medicine, Diagnosis and Radiology, M. A. Rangoonwala College of Dental Sciences, Azam Campus, Pune, Maharashtra, India

Correspondence
Dr. Tejashree Digambar Ghode, Senior Lecturer, Department of Oral Medicine, Diagnosis and Radiology, M. A. Rangoonwala College of Dental Sciences and Research Centre, 2390-B, Hidayatullah Road, Azam Campus, Pune - 411001, Maharashtra, IN
Tel: (020)26430960, (020)26430961
Fax: (020)26430962
E-mail: tejashree11@gmail.com

Key Clinical Message
Histopathological diagnosis should be considered mandatory in benign-appearing lesions, and the treatment choice should be surgical removal with adequate margins.

Keywords
Acinic cell carcinoma, malignant minor salivary gland tumor, oral cancer, tongue carcinoma, ultrasonography.

Introduction
In this study, a rare case is presented in which a tumor was found on the tip of the tongue. Ultrasound was performed on the lesion. Findings indicated a benign nature, but histopathological evaluation after surgical excision showed it to be a malignant minor salivary gland tumor of the tongue.

Salivary gland tumors represent a significant proportion of oral tumors and are the second most common neoplasm of the maxillofacial region after squamous cell carcinoma. The annual incidence of salivary gland tumors throughout the world ranges from about 1–6.5 cases per 100,000 people [1]. About 73.8% of these neoplasms are located in the major salivary glands, and the remaining 26.2% are found in the minor salivary gland [2]. About 88.2% of minor salivary gland neoplasms have been found to be benign in nature and about 11.8% are malignant [3].

In this study, we describe a rare case in which a nodular and freely movable tumor was found on the tip of the tongue; ultrasonographic (USG) findings indicated a round hypoechoic lesion without infiltration into the tongue musculature. These findings suggested that the lesion was benign. However, histopathological evaluation showed that the tumor was malignant acinic cell carcinoma. This case is notable because of the rarity of the tumor and its location. The use of ultrasound for such type of lesions is suggested as it is a readily available imaging modality as compared to MRI, and it enables to appreciate the pattern of the enlargement, the shape, border, and the vascularization and helps establish the exact location and relation with the surrounding structures. The aim of this study was to highlight the importance of histopathological analysis, regardless of the clinical and USG findings suggesting benign behavior.

Case History
A 42-year-old man presented with the chief complaint of swelling on the tip of his tongue, associated with mild discomfort when speaking. The swelling had been present for the past 4 months. It was not associated with numbness or discharge. He had noticed the swelling when it was about the size of a peanut and it had increased gradually.
Oral examination revealed a solitary, ill-defined nodular swelling on the right ventral surface of the tongue, 1 cm from the tip and not crossing the midline (Fig. 1). The well-demarcated swollen area was about 1.5 cm in diameter, with diffuse borders. The overlying mucosa was intact and normal in color. The swelling had caused elevation of the right dorsal surface of the tongue. Slight asymmetry of the tongue and widening of its right anterior third were observed in the relaxed state (Fig. 2). Palpation revealed a firm nodular mass, about 1 cm in diameter, beneath the submucosa; it was nontender and freely movable, with a positive slip sign. The patient had been edentulous for 6 months. His medical history was noncontributory, with no significant extraoral finding. Lymphadenopathy was not evident. A provisional diagnosis of a benign salivary gland tumor was made, and routine blood investigations, fine-needle aspiration cytology (FNAC), and radiographic and USG examinations were performed. A mandibular occlusal radiograph did not show the presence of any foreign body. A B-mode ultrasound examination of the tongue was performed; coronal and transverse sections confirmed the presence of a well-defined, rounded, iso- to hypoechoic lesion measuring 9 mm at the tip of the tongue, slightly on the right side, with a relatively hypoechoic margin suggestive of a capsule but no infiltration into the tongue musculature or subepithelium (Figs 3 and 4). The gray-scale examination was followed by the color Doppler sonography (CDS) examination. The lowest wall filter value and highest color sensitivity available on the machine were used to depict intratumoral blood flow. Doppler imaging showed
the absence of intralesional blood flow. USG examination of the cervical nodes, carotid vessel, and parotid and submandibular salivary glands yielded normal findings. The diagnosis, based on USG examination, was a benign-appearing semisolid lesion 1 cm posterior to the tip of the tongue.

Surgical treatment was planned, and preoperative FNAC yielded negative findings. Excisional biopsy was performed on a 1-cm-diameter encapsulated nodule with smooth, rounded borders, and firm consistency was removed. The ultrasound report was correlated with the histopathological report.

Considering the clinical presentation and localization of the lesion, we included minor salivary gland tumors such as mucoepidermoid carcinoma, Warthin’s tumor, and clear cell carcinoma. Lesions such as giant cell fibroma, focal fibrous hyperplasia, deep-seated mucocele and granular cell tumor, neurofibroma, neurilemmoma, subgemmal neurogenous plaque, intraoral rhabdomyoma amyloid tumor were also included in the differential diagnosis.

**Histopathology**

Grossly, the specimen measured about 1.7 x 1.7 cm; it was a well-circumscribed, circular, soft gray/white mass (Fig. 5). Microscopically, a histopathological section showed a lobular pattern of partially encapsulated tissue composed of centrally placed cells, resembling acinar cells, with granular eosinophilic cytoplasm and basally placed nuclei. The central cells were arranged in sheets, whereas intercalated duct-like cells with peripherally placed hyperchromatic nuclei predominated in surrounding tissue. The intercalated duct-like cells were columnar to cuboidal, and some places showed small duct-like spaces lined with cuboidal cells and mucous pools within the lumen. Clear cells with peripheral nuclei were present in many places; lymphoid aggregates were also noted in three or four places. Supporting stroma appeared to be scanty, with vascular proliferation (Fig. 6). Peripheral muscle infiltration was noted, and the lesion was diagnosed as solid-variant acinic cell adenocarcinoma of low-grade malignancy.

**Discussion**

Acinic cell carcinoma is a malignant epithelial neoplasm of the salivary glands in which the neoplastic cells show acinar cell differentiation. It is classified as a low-grade malignant neoplasm [3]. Most authors have agreed that tumors with infiltrative borders or local invasion are more likely to behave aggressively [4, 5]. Acinic cell carcinoma is defined by cytological differentiation toward...
serous acinar cells, the characteristic feature of which is
cytoplasmic periodic acid–Schiff-positive zymogen-type
secretory granules [6].

In the Armed Forces Institute of Pathology (AFIP) data
for salivary gland neoplasms, acinic cell carcinoma repre-
represented 17% of primary malignant salivary gland epithelial
neoplasms, after mucoepidermoid carcinomas and adenocarci-
nomas. In these data, acinic cell carcinoma comprised 6% of all salivary gland tumors [3, 6]. However, some studies have shown the occurrence to be 2.7–4.6%
of all salivary gland neoplasms [5, 7]. Although this
tumor has been reported to occur chiefly in the parotid
gland (80–97% of cases), it occasionally occurs in other
major salivary glands and about 0.2–8% of cases have
been reported in the minor salivary glands [6–10]; about
0.6% of cases occur in the minor salivary glands of the
tongue [6]. Acinic cell carcinoma occurs primarily in
middle age (average, 44 years), but it can occur at virtu-
ally any age; it has been found in children up to 11 years
and adults up to 77 years. Women are affected more
often than men (ratio, 3:2) [10, 11]. In the present case,
the lesion originated in the minor salivary glands of the
tongue tip of a 42-year-old male patient, which makes it
a rare occurrence.

The tumor usually arises as a slowly enlarging, asympto-
tomatic, fixed mass. In about 78% of cases, it appears as
a painless mass [12]. Most untreated patients had been
aware of the lesion for <1 year, and 5–10 years had
elapsed before a physician was consulted in about 7% of
cases [8]. Tumors of this type are fairly well circum-
scribed and encapsulated, with some degree of micro-
scopic invasion in 56% of cases. About 8% of cases show
facial paresis. Tumor size ranges from 0.7 to 7 cm, with a
firm rubbery consistency and a tan–gray cut surface.
Lymph node involvement has been observed rarely
[10–15].

Histologically, acinic cell carcinoma is composed
mainly of differentiated glandular cells bearing secretory
granules identical to those of the normal serous salivary
acinus [16]. Tumors in the minor salivary glands are his-
tologically similar to those arising in the major salivary
glands [10]. Four growth patterns have been described:
solid, papillary cystic, follicular, and microcystic. The
solid variant is characterized by numerous well-differen-
tiated acinar cells arranged in a pattern resembling nor-
mal parotid gland tissue [6, 12, 13, 15, 16]. A solid
growth pattern is the most easily recognized and domi-
nant (38% of cases) morphological variant of acinic cell
carcinoma; it is considered to be the “classic” pattern
because it is typified by numerous well-differentiated aci-
nar cells and closely resembles a normal parotid gland
[6]. Findings in our case were consistent with this classic
solid variant. In the differential diagnosis, we considered
clear cell carcinoma, mucoepidermoid carcinoma,
Warthin’s tumor, and oncocytoma.

In the present case, it was useful in determining the
tumor site and size; the sharp borders and homogeneity
suggested the noninvasive nature of the lesion. Sharp bor-
ders have also been noted in other acinic cell carcinomas.
The echostructure of the tumor is not an indicator of
malignancy. Whether a tumor is homogeneous or not, on
the whole, gives no indication of its nature, as around
50% of both benign and malignant tumors have been
found to be nonhomogeneous, which means that the
echostructure of the tumor is not an indicator of malig-
nancy. It was also concluded that vascularisation alone
cannot determine the benign or malignant nature of a
tumor [17]. In such cases, elastography could provide
useful information in differentiating benign and malig-
nant behavior. Considering the apparently benign nature
of the tumor, our patient underwent surgical enucleation
and was followed closely. Surgical resection is the treat-
ment of choice for acinic cell carcinoma [18].

Conclusion

This case is notable because of the rarity of the tumor
and its location. The use of ultrasound for such type of
lesions is suggested as it is the more easily available imag-
aging modality as compared to MRI and it enables us to
appreciate the pattern of the enlargement, the shape, bor-
ders and the vascularisation and helps establish the exact
location and relation with the surrounding structures.
Considering the clinical and ultrasound correlation, the
malignant tumor in this case was initially diagnosed as
benign. We suggest that no lesion should be underesti-
imated; histological diagnosis should be considered
mandatory, and the treatment of choice in benign-appear-
ing malignant lesions should be surgical removal with
adequate margins.

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Conflict of Interest

None declared.

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