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ADENOID CYSTIC CARCINOMA OF TONGUE: A RARE ENTITY

Oral & Maxillofacial Surgery

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ABSTRACT

Adenoid cystic carcinoma is one of the rarest salivary gland carcinoma that predominantly arises from the minor salivary glands, the palate being the most common site. The tumor is typically slow-growing compared to other carcinomas with perineural invasion and distant metastasis are common findings. This neoplasm is defined by its distinctive histological appearance. We have reported a case of adenoid cystic carcinoma involving the right anterior two third of tongue. The aim here is to highlight the importance of diagnosis; its treatment planning and long term follow up of the patient.

KEYWORDS

Adenoid cystic, Cylindroma, CEMRI, basaloid cells, honeycomb

INTRODCUTION

The adenoid cystic carcinoma is an unusual and well-recognized salivary gland malignancy that may affect either the minor or major salivary glands of the oral cavity.(1) However it can also occur in other parts of the body, such as the breast, cervix and prostate gland. (2) ACC was first described by Robin, Lorain and Laboulbene in 1853. Billroth in 1859 suggested the name Cylindroma because of its distinctive histopathologic features. This term is still being used sometimes as a synonym for this neoplasm. In 1930, Spies suggested the term adenoid cystic carcinoma to replace cylindroma and this name has been widely accepted.(3) Initially the tumor was thought to be a benign variant of the mixed salivary gland, but Dockerty and Mayo emphasized the malignant nature of this tumor. (4,5)

Adenoid cystic carcinoma is one of the rarest carcinoma that predominantly arises from the minor salivary glands, the palate being the most common site and the tongue being the rarest. Perineural invasion and distant metastasis are common findings in adenoid cystic carcinoma.(6)

CASE REPORT

A 33 years old male patient reported to our department with a chief complaint of swelling on the right side of his tongue since 3 months. No significant extra-oral findings were recorded. On intra-oral examination, a 2.5 X 2 cm ulcero-proliferative lesion extending anteroposteriorly 1cm from the tip of the tongue to the distal cuspal margin of mandibular right second premolar, Supero-inferiorly; involving the dorsal margin of right lateral border of tongue to 2 cm above floor of the mouth (figure 1).

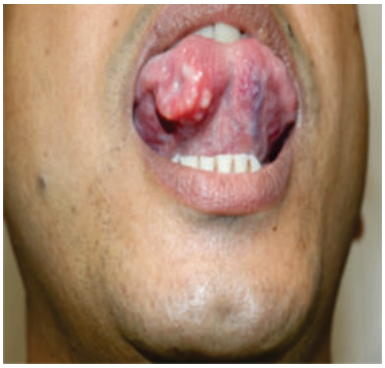

Fig 1: Clinical Picture Of Adenoid Cystic Carcinoma Involving The Ventral Surface Of The Anterior 2/3rd Of Tongue.

The lesion had an irregular with diffuse margins, firm in consistency, non-tender with an indurated base. The patient's cytomorphological report showed occasional epithelial clusters with atypia for which biopsy was strongly recommended. Biopsy was performed and the report showed that the tumor was composed of ductal and myoepithelial cells arranged predominantly in cribriform pattern along with some pattern of tubular pattern. The Myoepithelial cells are having angulated nuclei and scant cytoplasm. There was no evidence of necrosis and peri-neural invasion.

CEMRI of the patient showed a signal intensity alteration involving the tip of the right anterior two-third of the tongue reaching upto the median raphe without any contralateral extension. The lesion was infiltrating the intrinsic muscles of the tongue and anterior fibres of right geniohyiod and genioglossus muscle (figure 2).

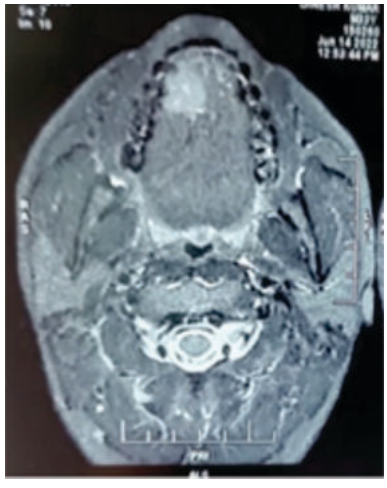

Fig 2 Cemri Of Tongue Shows A Hyper Intense Signal Of The Right Anterior 2/3rd Of Tongue Involving The Intrinsic Muscle Reaching Upto The Median Raphe Without Contralateral Extension.

Treatment

The entire procedure was performed under general anaesthesia. Intra-operatively, the lesion was resected from its two-third of the tongue with 1 cm safety margin (figure 3). The resected specimen was sent for frozen section biopsy and the report was free of any positive margin. Primary closure of the tongue was done.

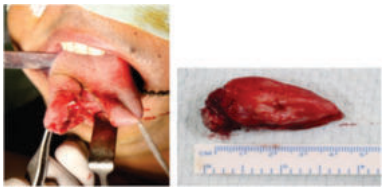

Fig 3: Intraoperative Picture Showing The Excision Of The Lesion And The Resected Specimen

Histolpathological examination, showed an infiltrating tumour with >80% of cribriform pattern and are composed of basaloid cells with dark blue, angulated nuclei and scant cytoplasm. (Figure 4 (A) and (B): H&E x 40). Immunohistochemistry showed tumour cells display strong cytoplasmic expression for CD117 (figure 4 (C): DABx200). p40 was negative in tumour cells, however the myoepithelial cells of adjacent unremarkable duct display immunoexpression (figure 4 (D): Dabx200).

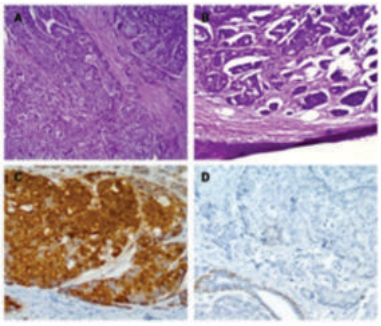

Fig 4: Tumour Nests Are Showing >80% Of Cribriform Pattern With Scant Cytoplasm.

The patient was followed on a regular follow-up on the 3rd and 6th months respectively to track the evidence of any recurrence. The patient was overall satisfied with the treatment. (Figure 5)

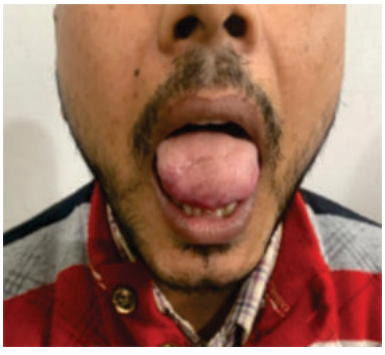

Fig 5: Showing The Post Operative Picture

DISCUSSION

Adenoid cystic carcinoma can occur in any salivary gland site, but approximately 50% develop within the minor salivary glands. It is a relatively rare carcinoma, constituting only 2% to 3% at all tumors. It is also relatively common among palatal salivary neoplasms with 8% to 15% of all such tumours. The lesion is most common in middle-aged adults and is rare in people younger than age 20. There is a fairly equal sex distribution, although some studies have shown a slight female predilection.(3)

Three major histologic patterns of growth have been recognized. The cribriform pattern: "Honeycomb" or Swiss cheese" pattern characterised by islands of basalaid epithelial cells that contain multiple cylindric, cyst like spaces often contain a mildly basophilic mucoid material. The Tubular pattern; the tumor cells are similar but occur as multiple smalll ducts or tubules with in a hyalinized stroma. The Solid variant consists of larger islands or sheets of tumor cells that demonstrate little tendency toward duct or cyst formation (3, 7). Unlike the former patterns, cellular pleomorphism and mitotic activity as well as focal necrosis in the center of the tumor islands may be observed. The solid variant has the worst prognosis.(8). Distant metastasis occurs through blood stream to lungs and bones. Direct extension of the lesion to the base of skull have also been reported.(5)
The choice of treatment for adenoid cystic carcinoma is surgical resection with a post-surgical radiotherapy (not less than 60 Gy) which enhances local and regional control and may slightly improve patient survival in some cases. The role of chemotherapy however is still controversial (8,9). The prognosis of the lesion is poorest for tumors arising in the maxillary sinus and submandibular gland.

CONCLUSION

Adenoid cystic carcinoma shows a variety of histologic and clinical manifestation with a possibility of distant metastasis. Although ACC is rare, it is important to rule out the differential diagnosis in patients presenting with pain, swelling, bony destruction and paraesthesia involving either the major or minor salivary glands through a thorough assessment. The correct diagnosis of the disease will help to determine the treatment planning for better outcome.

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Research involving human participants and/or animals: yes and ethical clearance approved by the ethical committee

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