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Case Report

Adenoid Cystic Carcinoma of Mandible – A Rare Site of Presentation

Gurshinder Pal Singh

Associate professor, Department of ENT, Guru Gobind Singh Medical College, Faridkot, Punjab, India

Abstract

Adenoid cystic carcinoma (ACC) of mandible is very rare. We are presenting a case of ACC of mandible in 35-year-old female. Mandible is very rare site of this carcinoma. ACC is locally invasive tumor. In our case, it was involving mandible and extending into floor of mouth involving deep muscle of tongue. We did radical surgery by doing wide local excision with near total glossectomy, along with mid-arch mandibulectomy and neck dissection reconstruction with free fibula flap. The patient was given radiotherapy post-operatively.

Key words: Adenoid cystic carcinoma, Glossectomy, Mandibulectomy

INTRODUCTION

Adenoid cystic carcinoma (ACC) is malignant tumor of salivary glands. It accounts for 10 percent of all the salivary gland tumors.[1] It commonly occurs in minor salivary glands, which accounts for 60% of all the ACCs. Common sites of occurrence of ACC in the oral cavity include hard palate, lower lip, retromolar trigone, sublingual gland, and buccal mucosa. Extraorally, it is found in submandibular gland and parotid gland. ACC is a slow growing, locally invasive tumor. It is usually present as a painless swelling initially, which becomes painful in the advanced stage of the disease due to its perineural invasion. It can metastasize through lymphatics to the cervical lymph nodes and can also spread to the lungs through bloodstream. We, hereby, report are a case of ACC in the mandible, which is a rare site of its presentation.[2,3]

CASE REPORT

A 35-year-old female patient presented to us, with a swelling over left side of her face since 1 year. Initially, the swelling was painless, but it grew progressively, over a period of time to its current size. The patient also complained of pain over the swelling since the past 3 months accompanied with difficulty in swallowing. On extraoral examination, a 6 cm × 5 cm swelling was seen over the chin area. Intraorally, the swelling was observed to be arising from the lower alveolus in the midline, involving the floor of mouth, accompanied with restricted tongue mobility. On intraoral palpation of the swelling, an induration was present over the right dorsal surface of the tongue extending up to the ventral surface bilaterally. The base of the tongue was also involved, more over the left side, crossing the midline and involving the right side also.

Indirect laryngoscopy examination revealed normal valleculae, larynx, and hypopharynx. Neck examination demonstrated absence of palpable, involved lymph nodes bilaterally. A biopsy was taken from the mass and sent for histopathological examination which confirmed the diagnosis of ACC of the midline of the mandible. Contrast-enhanced computer tomography (CECT) of the swelling exhibited an ill-defined enhancing mass in the midline of the mandible involving the overlying and adjacent soft tissue. The tumor mass measured 6 cm × 5 cm × 4 cm in dimensions. The destruction of the cortex of mandible was evident, along with its posterior extension, along the floor of mouth on the left side with involvement of hypoglossal, geniohyoid, and genioglossal muscles [Figure 1]. Bilateral submandibular glands were normal. Few enhancing level 2 subcentimetric lymph nodes were present on the left side of the neck.

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Corresponding Author: Dr. Gurshinder Pal Singh, Department of ENT, Guru Gobind Singh Medical College, Faridkot, Punjab, India.

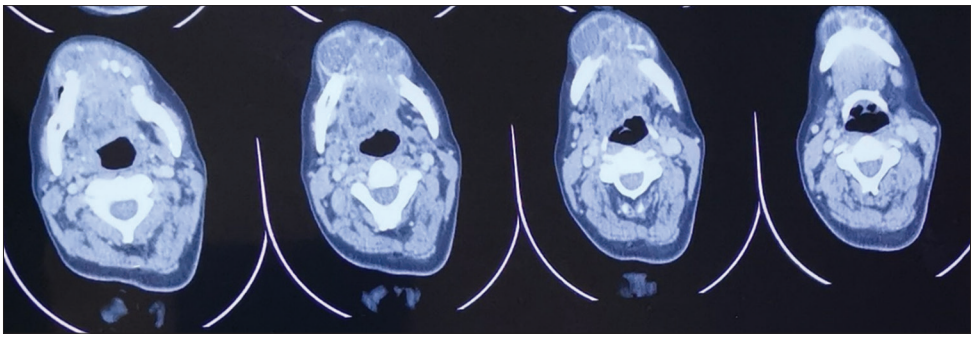

Figure 1: Computed tomography scan shows contrast enhancing mass involving mandible and extending to floor of mouth

On complete evaluation of the tumor mass, surgery with composite resection of the tumor mass, with near total glossectomy, along with mid-arch mandibulectomy was planned. Reconstruction of the post-operative defect with free fibula flap with left sided, supra-omohyoid neck dissection, was also decided. Surgery was performed, along with temporary tracheostomy. Post-operative healing was uneventful and the patient recovered completely. The excised specimen was sent for complete histopathological examination. The latter revealed an ACC with perineural invasion involving the mandible. All the bony and soft-tissue margins were free of tumor. Neck dissection specimen showed three positive nodes out of a total of 10 for the presence of tumor deposits. Consequently, the patient was referred for post-operative adjuvant radiotherapy, due to perineural invasion and lymph node metastasis.

DISCUSSION

ACC forms 10% of the all head and neck cancers.[1] It is a slow growing, locally invasive tumor mass which often presents with perineural invasion. Parotid gland is a major site of ACC, but majority of the cases arise from minor salivary glands in the oral cavity, nose, and paranasal sinuses. Mandible is a very uncommon site of this tumor. Even though, the origin of this particular type of tumor is still largely unknown, some theories have been put forward, to elucidate the reason of incidence of this malignant salivary gland tumor within the central jawbones. Some of them include entrapment of ectopic salivary gland tissues in jawbones; neoplastic transformation of epithelial lining of a cyst or sinus epithelium.[4]

Diagnostic modalities for detection of this tumor include fine-needle aspiration biopsy from major salivary glands, like parotid, but in vast majority of minor salivary gland tumors, a confirmatory biopsy becomes mandatory. Radiological examination is of utmost importance, to know the extent of the disease, invasion of the adjacent vital structures, and also to determine the perineural invasion. Computed tomography scan helps in determining the involvement of bony structures, while magnetic resonance imaging assists in defining the soft-tissue extent and perineural invasion. In our case, we did CECT scan because the tumor was arising from mandible.

The treatment of ACC of minor salivary glands requires wide local excision with neck dissection. In our case, we did wide local excision with near total glossectomy accompanied with mid-arch mandibulectomy. Left supraomohyoid neck dissection was then performed followed by reconstruction with free fibula flap.

Prognosis of ACC depends on the tumor site, stage, histology, perineural invasion, and lymph node metastasis. ACC of major salivary glands has a better prognosis when compared to minor salivary gland ACC. The stage of the tumor is a fine indicator of its prognosis. A higher stage of the tumor, usually, foretells a worse prognosis. There are three histological types of ACC; tumor mass with a predominant tubular pattern; with a greater cribriform pattern and a pronounced solid pattern. Tubular pattern has the best prognosis; cribriform pattern usually depicts a moderate prognosis, while the solid pattern is associated with the most adverse outcomes.[5,6] The overall 5-year survival rate is lower (48%) for the patients having lymph node metastasis compared to those without lymph node metastasis (77%).[7] Therefore, the lymph node metastasis is a significant predictor of prognosis of ACC cases in oral cavity.

Radiotherapy is always preferred, following a radical surgery of an ACC tumor mass, as it eradicates any residual microscopic disease, and thus, associated with a better outcome. Radiotherapy of 60 Gray (Gy) or higher is usually preferred on account of improved results.[8] Additional radiotherapy can be administered in cases exhibiting perineural invasion without positive margins for superior outcomes. The overall 5-year survival rates of patients with head and neck ACC have been reported to be approximately 90.3%. Our patient had a locally advanced ACC of mandible, with significant local invasion, along with lymph node metastasis and perineural invasion, but

all surgical cut margins were free. Hence, we referred the patient for post-operative radiotherapy.

CONCLUSION

Mandible is a rare site for occurrence of ACC. ACC in this location can significantly invade local structures and can metastasize to cervical lymph nodes. Therefore, a locally invasive ACC, post a radical surgery, should be certainly followed by post-operative radiotherapy for a superior prognosis and an enhanced outcome.

REFERENCES

1. Kim KH, Sung MW, Chung PS, Rhee CS, Park CI, Kim WH. Adenoid cystic carcinoma of the head and neck. Arch Otolaryngol Head Neck Surg 1994;120:721-7.
2. Giannini PJ, Shetty KV, Horan SL, Reid WD, Litchmore LL. Adenoid cystic carcinoma of the buccal vestibule: A case report and review of the literature. Oral Oncol 2006;42:1029-32.
3. Mahajan A, Kulkarni M, Parekh M, Khan M, Shah A, Gabhane M. Adenoid cystic carcinoma of hard palate: A case report. Oral Maxillofac Pathol J 2011;2:127-31.
4. Seetharaman D, Daniel MJ, Srinivasan SV, Jimsha VK. Adenoid cystic carcinoma of the mandible: A rare clinical entity. Trop J Med Res 2017;20:104-8.
5. Huang M, Ma D, Sun K, Yu G, Guo C, Gao F. Factors influencing survival rate in adenoid cystic carcinoma of the salivary glands. Int J Oral Maxillofac Surg 1997;26:435-9.
6. Chundru NS, Amudala R, Thankappan P, Nagaraju CD. Adenoid cystic carcinoma of palate: A case report and review of literature. Dent Res J (Isfahan) 2013;10:274-8.
7. Carrasco Ortiz D, Aldape Barrios B. Adenoid cystic carcinoma of the dorsum of the tongue: Presentation of a case. Med Oral Patol Oral Cir Bucal 2006;11:E417-20.
8. Umeda M, Nishimatsu N, Yokoo S, Shibuya Y, Fujioka M, Komori T. The role of radiotherapy for patients with adenoid cystic carcinoma of the salivary gland. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2000;89:724-9.

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