Case Report

Case report on adenoid cystic carcinoma of palate with review of literature

J S Lakshmi[1], Binitha T Thomas[2], Diana Sevi Jose[3]

From [1]Senior Resident, Department of Radiation Oncology, Sri Ramachandra Medical College and Research Institute, Chennai, Tamil Nadu, [2]Associate Professor, [3]Senior Resident, Department of Radiation Oncology, Government Medical College, Kottayam, Kerala, India

ABSTRACT

Adenoid cystic carcinoma (ACC) is a malignant tumor of minor salivary glands of the oral cavity, particularly the palate which accounts for about 5–10%. These tumors are slow growing with a high incidence of local recurrence, perineural invasion, and distant metastasis. Here, we report the case of a 69-year-old female who presented with swelling over the hard palate. Computerized tomography shows enhancing lesion along the undersurface of the left side of the hard palate, abutting the maxillary alveolus in the region of the second premolar and first two molars. There was no erosion of the inner cortex of the maxillary alveolus and no extension into the nasal cavity. Following the biopsy, she underwent a left hard palatectomy with upper alveolar resection. The final histopathological report confirmed ACC with no perineural invasion. Following she received adjuvant radiotherapy using an intensity-modulated radiotherapy technique. Now at 6 months of follow-up, there was no evidence of recurrence. Thus, our patient requires long-term follow-up to assess the benefit and survival outcome.

Key words: Adenoid cystic carcinoma, Cylindromas, Hard palate, Minor salivary gland, Oral cavity

Adenoid cystic carcinoma (ACC) also known as cylindroma is the most common malignant neoplasm of minor salivary glands of the oral cavity, particularly the palate [1-3]. It is a slow-growing tumor that accounts for about 5–10% of all salivary gland neoplasms. In general, these tumors are slow growing with a high incidence of local recurrence, perineural invasion, and distant metastasis [4]. Although ACC accounts for about 5–10% of all minor salivary gland malignancy, it constitutes <1% of all head-and-neck tumors.

In view of limited data on ACC, we report a case of ACC of the palate and a brief literature review on its clinical, histopathological, and therapeutic aspects.

CASE REPORT

A 69-year-old woman, non-smoker presented to our clinic with an 8-month history of swelling over the hard palate. The patient had no history of cough, dysphagia, or dyspnea. On general examination, the patient was oriented, moderately build, and nourished with high motor functions and normal cerebellar functions. Vitals were stable and other systems were within normal limits. On local examination, a 3 × 2.5 cm firm to hard swelling was seen over the left side of the hard palate extending to the left upper alveolus, medially not crossing the midline and posteriorly not extending to the soft palate. Clinically, no lymph nodes were palpable.

Biopsy reported the swelling as ACC. Computerized tomography (CT) of the head and neck with contrast medium showed heterogeneously enhancing lesion along the undersurface of the left hard palate. The lesion abuts the maxillary alveolus in the region of second premolar and molars on the left side. There was no erosion of the inner cortex of the maxillary alveolus and no extension into the nasal cavity. No significant lymph nodes were noted. The findings from the chest CT were normal.

The patient underwent a left hard palatectomy with upper alveolar resection and obturator reconstruction. The final pathology report confirmed ACC with negative margins and no perineural invasion was noted (Fig. 1). Thus, based on the American Joint Committee on Cancer 8th edition, the TNM stage was pT4a [5]. She received adjuvant radiotherapy of a total dose 60 Gy in 30 fractions using an intensity-modulated radiotherapy technique (Fig. 2). Now at 6 months of follow-up, there was no evidence of recurrence.

DISCUSSION

ACC is a rare tumor of the head-and-neck region of epithelial origin. It most often occurs in minor and major salivary glands.

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Correspondence to: Dr. J S Lakshmi, Department of Radiation Oncology, Sri Ramachandra Medical College and Research Institute, Chennai, Tamil Nadu, India. E-mail: jslakshmi1990@gmail.com

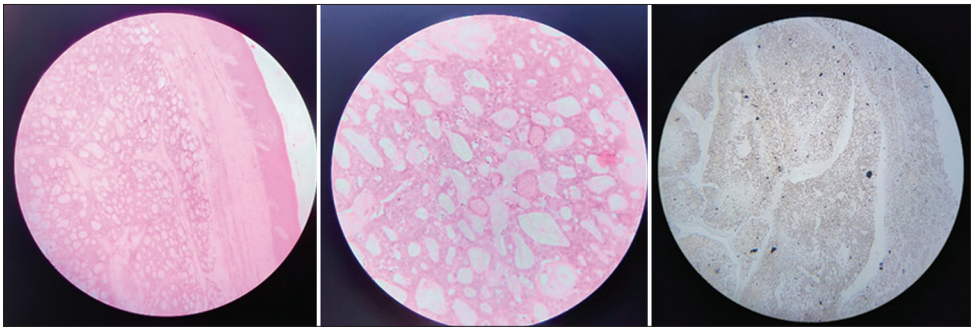

Figure 1: Histopathological findings of adenoid cystic carcinoma of hard palate

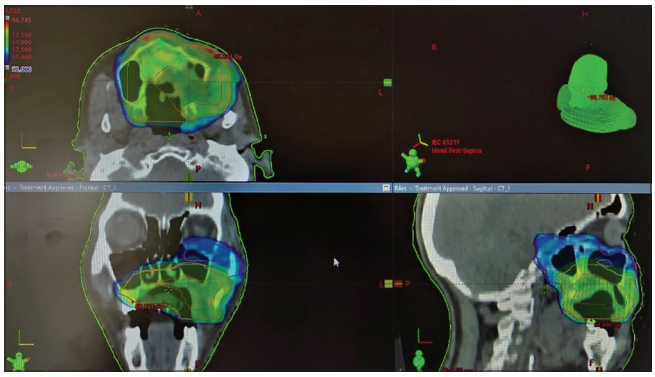

Figure 2: Radiation planning of ACC hard palate in post-operative setting

The palate is the most common site of intraoral lesion, the tongue being the second most common followed by the floor of the mouth and lip [6]. Rarely, it may present as intraosseous tumors of the maxilla and mandible. The terminology "cylindroma" was first coined by Billroth in 1859 due to its characteristic histology appearance [7].

The highlighted feature of this tumor is that the patient reports a long clinical course, with slow and progressive growth having a higher tendency to recur locally as well as metastasize regionally and systemically, sometimes at very late stages. In addition, it presents a strong neurotropism, which is a major route of tumor dissemination [1]. Frequently, the size of the tumor at the time of clinical presentation is reported to be usually 2–4 cm at its greatest diameter and intraoral lesion is seldom larger than 3 cm [8,9]. The incidence of cervical metastasis is low. As mentioned in the literature, our patient presented with 8 months clinical history of painless slow-growing tumor in the hard palate of size 3×2.5 cm, well lateralized with no significant neck nodes.

Histopathological confirmation is a must as the symptoms are similar to squamous cell carcinoma. ACC is classified into three distinct subtypes: Cribriform, tubular, and solid [10] of which cribriform is the most common, tubular carries the best prognosis, and solid type has the worst prognosis. In our case, we proceeded with laryngoscopy and biopsy for confirmation.

Imaging is a valuable part of pre-operative mapping and staging of the disease. It is highly essential in the assessment of primary tumor location, its extension, regional, and distant metastases. In our patient, we assessed with CT neck with a contrast medium that showed the presence of a submucosal mass in the hard palate. The added advantage of the CT scan was to rule out bony/cartilage involvement and suspicious neck nodes. Furthermore, these findings correlated with the final histopathologic findings.

In the present era, the various treatment options available for ACC are surgery, radiotherapy, and chemotherapy. These tumors are radiosensitive but not radiocurable. Hence, radiotherapy alone as a treatment modality is still debatable [11]. Proven benefits are noted in the adjuvant modality in case of positive margin, locally advanced stage, perineural or high grade in prolonging survival, and preventing local recurrence [12]. Thus, a combined modality treatment approach is preferred to prevent metastasis and improve the local control. The preferred multimodality approach is surgical resection followed by adjuvant radiation treatment. The role of systemic therapy (chemotherapy) is still controversial [13,14]. Our case underwent surgical resection followed by post-operative radiotherapy of a total dose 60 Gy in 30 fractions using an intensity-modulated radiotherapy technique. Since the perineural and hematological spread is common, regular close and long-term follow-up is mandatory. Distant metastases commonly occur in the lungs, liver, abdomen, lymph nodes, and bones.

CONCLUSION

ACC of the minor salivary glands has a better prognosis as compared to major salivary glands because the intraoral lesions are diagnosed and treated earlier. As per the literature, the ACC of the palate is said to have a favorable prognosis that the lesion elsewhere and better quality of life. Long-term follow-up is required to observe the local recurrence and metastasis pattern.

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