Case Report

Adenocystic Carcinoma of Hard Palate with Computed Tomographic Features: A Case Report and Review of Literature

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Abstract

Adenoid cystic carcinoma (ADCC) is a rare tumor that begins in the minor salivary glands, where the palate is the most common site. <1% of head and neck cancers and 10% of salivary gland tumors are ADCC. The most commonly affected area of the oral cavity is the palate, followed by the parotid and submandibular glands. They are often asymptomatic; grow slowly, with perineural infiltration, and invasive growth as their hallmarks. This publication reports a case of glandular cystic carcinoma in a 60-year-old woman with swelling in the left side of the palate, including the hard palate, for 2 months and was histopathologically diagnosed. The purpose here is to highlight the clinical, radiological, and histopathological aspects of adenocystic carcinoma.

Keywords: Adenoid cystic carcinoma, CT scan, Malignant salivary gland tumor, Maxillary sinus, Palate, Perineural invasion.

Introduction

Adenoid cystic carcinoma (ADCC) is a malignant neoplasm that can affect either large or small salivary glands.[1,2] It first described by three Frenchmen (Robin, Lorain, no Laboulbene) in two articles published in 1853 and 1854 who described the cylindrical appearance of this tumor. In 1859, Billorth first described the ACC under the term "cylindroma," for which a cribriform appearance formed by cylindrical tumor cells pseudolumina or pseudospaces, with the high tendency to recur.[3] In 1930, Spies proposed the term ADCC instead of cylindroma and the name became widely accepted. Until the 1940's, this tumor was considered a benign variant of a mixed tumor of the salivary glands. In 1943, Dockerty and Mayo give emphasis on the malignant nature of this tumor.[4] This tumor signifies 1% of all malignant tumors of oral and maxillofacial region and 22% of all salivary gland epithelial malignancies.[5]

ADCC is an inactive tumor with slow progression and distant recurrences.[6] ADCC with high grade transformation is a very rare variant. It is more aggressive and has more local recurrences and metastasis.[7] It is currently being recognized ADCC remains the most difficult disease to treat. Conley and Dingman explained it"as one of the most damaging and unexpected head and neck tumours."[8] Clinically, it shows slow growth, local recurrence, perineural invasion, and distant metastasis.[9] A characteristic feature of ADCC is perineural invasion, even in the early stage of tumor.[10] We have reported a case of adenocystic carcinoma of minor salivary gland of palate and a brief review on its clinical, radiological, histopathological, and therapeutic aspects.

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Case Report

A 60-year-old female patient came with the chief complaint of swelling in the upper left back tooth region from last 2 months. According to the patient's history, the swelling began with small size and gradually grew to its present size patient already went for extraction of 27 from a general dental clinic 2 years back due to decay, even discomfort persisted after extraction. In addition, there was no relevant medical and tobacco consumption history. Extraoral examination revealed no facial asymmetry or lymphadenopathy.

Intraoral examination revealed a single erythematous diffuse swelling extending anteroposteriorly from the upper left canine to maxillary tuberosity region and buccolingually extending

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from buccal vestibule to midline of hard palate without crossing. Swelling was associated with the mobile root stump of 27. No surface discharge was present. Overlying mucosa of swelling shows ulceration on the involved tooth. On palpation swelling was non-tender, non-fluctuant, and soft to firm in consistency (Figure 1).

On the basis of chief complaint and clinical examination, provisional diagnosis was made as radicular cyst. The clinical differential diagnosis included a benign or malignant neoplasm of minor salivary glands and a neoplasm of maxillary sinus. Radiological and histopathological investigation was done to rule out the differential diagnosis. Routine blood investigations were found to be normal.

Panoramic radiograph showed irregular radiolucency with poorly defined border extending from maxillary left 1st molar to maxillary tuberosity region involving left maxillary sinus (Figure 2) and computed tomography (CT) scan revealed an opacified, destructive, and obliteration of left maxillary sinus along with invasion of pterygopalatine fossa (Figure 3a-c). An incisional biopsy was performed from palatal and maxillary sinus lining and histopathology confirmed it as ADCC of tubular pattern shows neoplastic cells arranged in multiple small tubules or ducts lined by stratified cuboidal epithelium (Figure 4).

Figure 1: Intraoral photograph shows the presence of diffuse swelling of size 3 × 2 cm with an ulcerative lesion on the surface involving the hard and soft palate but not crossing the midline

Figure 2: Panoramic radiograph shows radiolucency with ill-defined borders, involving left maxillary sinus

Discussion

ADCC comprises approximately 29.6% of cancers of minor salivary glands. Palate is most frequent site of involvement, followed by tongue; floor of the mouth and lip.[1] ADCC is commonly seen in minor salivary gland and submandibular salivary glands but less commonly seen in parotid and sublingual salivary glands. Other unusual sites include the gastrointestinal tract, lachrymal glands, and adnexal cutaneous glands.[11] Mucoepidermoid carcinoma (21.8%) is the most common malignant neoplasm of minor salivary glands followed by polymorphous low-grade adenocarcinoma (PLGA) (7.1%) and ADCC (6.3%), respectively.[12]

ADCC can be found in any age group population, but a majority of cases are seen between 24 and 78 years of age. ADCC of major salivary glands is seen in the younger generation (mean 44 years) than in minor salivary glands (mean 54 years) with a female predilection. (F: M − 1.2:1). Clinically, ADCC is a

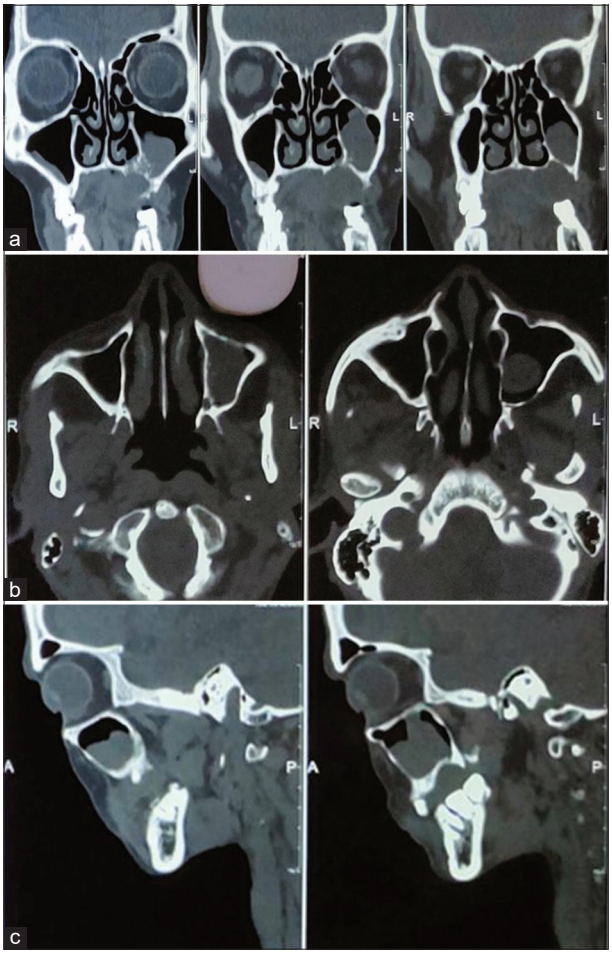

Figure 3: (a) Coronal view-unenhanced computed tomography (CT) image shows an opacified sinus with destructive maxillary sinus tumor. (b) Axial view-unenhanced CT image reconstructed with bone algorithm shows an extensive bony defect of the posterior maxillary sinus wall. (c) Sagittal view shows pterygopalatine fossa invasion accompanied by perineural spread

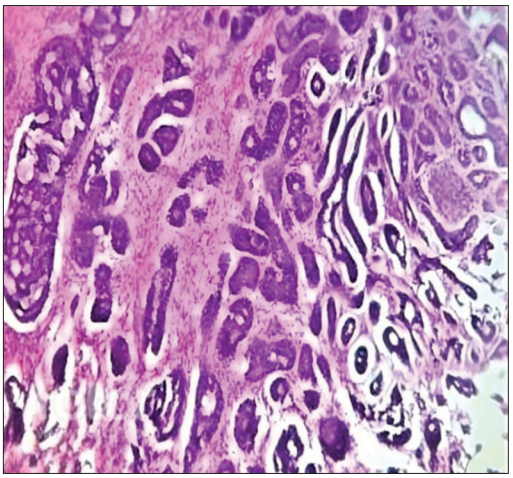

Figure 4: Photomicrograph shows intensely basophilic ductal neoplastic cells arranged in tubular pattern with numerous microcystic spaces with hyaline material. The neoplastic cells show perineural invasion

symptomatic slow-growing mass with a smooth or ulcerative overlaying surface, soft to firm in consistency on palpation rarely larger than 3 cm in diameter. Radiologically, ADCC shows bone destruction.[12]

ADCC is usually encapsulated which shows infiltration and invasion of underlying bone. Through the bloodstream, distant metastasis occurs to the bones and lungs. The cause of death has been reported due to direct expansion of lesion at the base of the skull. Cervical metastases are barely common.[13] ADCCs of the minor glands have been reported to have worse prognosis than those of the major salivary glands. Tumors involving the nose, paranasal sinuses, and maxillary sinus have the worst prognosis as they are usually detected with higher stages at the time of diagnosis.[14]

The best CT feature for diagnosing ADCC in the palate with intact mucosa is the enlargement of greater palatine foramen (GPF). The most distinctive characteristic features of ADCC are PNS signs, such as GPF enlargement and involvement of the foramen rotundum and pterygopalatine fossa. Clinically, if tumors originate in the palate, GPF are the first site among PNS to be affected, followed by the pterygopalatine fossa and the foramen rotundum.[13]

Histopathologically, ADCC is consist of three patterns such as cribriform characteristic feature ("swiss cheese pattern"), tubular and solid pattern; cribriform pattern is the most common variant, in which the neoplastic epithelial cells are arranged in multiple cylindrical spaces which give pseudocystic appearance, and many of these pseudocystic space contain a hyaline material. The solid variant of ADCC has a poor prognosis as compare to cribriform variant which has a better prognosis.[15] Perineural invasion, and to a lesser extent, intraneural invasion, is a prevalent and noticeable characteristic of ADCC. Beyond the tumor's clinically visible borders, tumors can travel a long way through nerves. PLGA, salivary duct cancer, basal cell adenoma, and basaloid squamous cell carcinoma are among the possible diagnoses.[15]

ADCC has various histological patterns and clinical behavior; therefore, distinct treatment modalities can be include such as surgery, radiation, chemotherapy, and combined therapy in most situations. Due to significant differences in histological grading and clinical classification, it is imperative to have a thorough understanding of anatomy and physiology in addition to pathology skills while diagnosing ADCC.[15]

Conclusion

ADCC is an invasive and uncommon salivary gland malignancy and palate is the most common site of involvement which can invade to the maxillary sinus and other surrounding anatomic structure. CT is a very important modality and helpful for the early detection and extension of the lesion to other important anatomic structures such as maxillary sinus, GPF, pterygopalatine fossa, and foramen of rotundum involvement of which can cause death. CT scan is also important for surgical treatment which will improve the prognosis and quality of life.

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Conflicting Interest

None.

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