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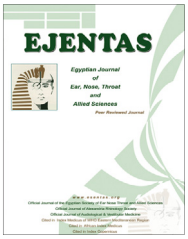

CASE REPORT

2 Case reports of sinonasal adenoid cystic carcinoma: Review of the literature on surgical approaches

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Abstract Adenoid cystic carcinoma (ACC) of sinonasal is an uncommon tumour that progresses slowly and generally manifested in advanced stage. Surgical management of sinonasal ACC can be challenging to the attending surgeon as they need to outweigh the decision between tumour clearance and morbidity of the patient. Multimodality treatment is the mainstay of treatment for sinonasal ACC and prognosis depends on the histological subtype of tumour, patient factor and treatment modalities opted. We are presenting 2 cases of sinonasal ACC manifested in different age groups in our centre with different types of surgical approaches opted and their outcomes.

1. Introduction

Sinonasal malignancy accounts for 1–2% of all malignancies. Adenoid cystic carcinoma (ACC) is the 3rd commonest sinonasal malignancy. It is common in females compared to males with a ratio of 2:1 and the peak incidence at age of 5th to 6th decade of life. The most common site affected is in the maxillary sinus followed by nasal cavity.[1] Tumors of the sinonasal tract commonly present with symptoms that are identical to those caused by inflammatory sinus disease, such as nasal obstruction, nasal discharge, epistaxis, headache, facial pain and cheek swelling.

ACC exhibits extensive local tissue infiltration and perineural spread, which results in a high rate of recurrence despite aggressive surgical resection. Classically, ACC has a distinct natural history of slow and indolent growth with late development of distant metastasis but tends to have local recurrence.[2] A few studies suggested that subtype of histopathology of

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ACC such as solid, tubular, cribriform and mixed types may determine the prognosis of the survival of a patient.

In general, tumours with perineural invasion, cervical lymph node metastasis, solid histological features and distant metastasis are associated with increased treatment failures and recurrences.[3] Tumour stage according to TNM staging has a significant correlation to the type of modality treatment opted.[4] In view of these tumours being rare and often presenting at an advanced stage, controversies exist as to the most appropriate treatment suitable. Until today, most sources agree that aggressive treatment of these tumours is necessary. Radiation therapy (RT), surgical resection, and combined modality treatments of these tumours have been reported and suggested.

2. Case report

2.1. Case 1

A 23 year-old Malay lady presented with left epistaxis for 8 months. There was no haematemesis and anaemic symptoms. It was associated with persistent left unilateral nasal obstruction associated gradual painless worsening left cheek disfigurement for 2 months. There were no allergic rhinitis symptoms and smell disorder. There was no cheek numbness, loosening of teeth, blurring of vision, or limited mouth opening. There were also no headache, throat or ear symptoms and no associated constitutional symptoms.

Generally patient was stable. There was blunted left nasolabial fold with vague mass overlying it. All cranial nerves examinations were unremarkable. The nasoendoscope examinations revealed a mass in the left nasal cavity which were dry, friable and bleed easily. The mass was sandwiched in between the septum and lateral nasal wall. Otherwise, the right nasal cavity was normal and both the Fossa of Rosenmueller (FOR) was clear.

Computed tomography (CT) scan revealed a soft tissue lesion at anterior left nasal cavity causing mild nasal septum deviation to the right. There was heterogenous enhancement seen post contrast and no soft tissue mucosal thickening seen at bilateral maxillary, ethmoidal, frontal and sphenoid sinuses without bony destruction. There was no turbinate hypertrophy, bilateral osteomeatal complexes are patent (Fig. 1). There were no neck nodes or distant metastases.

Magnetic resonance (MRI) scan showed a well-defined enhancing mass occupying the anterior aspect of left nasal cavity with a tiny hypointense lesion with no enhancement on post-gadolinium T2 weighted image. There was evidence of deep aspect of the overlying nasal ala subcutaneous tissues involvement but preservation superficial subcutaneous fat and skin of the nasal ala. No enhancing flow void was seen within the mass to suggest for an enlarged vessels within. The mass is in contact with the nasal septum. There was minimal focal deviation of the nasal septum to the right. There was no extension to the paranasal sinuses or focal intracranial lesion. There was no perineural invasion.

Biopsy of the mass revealed multiple fragment of tumour tissues, a few of the fragments are lined by stratified squamous epithelium. The tumour cells are predominantly in solid nest (60%) and cribriform pattern (40%). It is composed of small basaloid cells with scanty cytoplasm. Mitosis is frequently

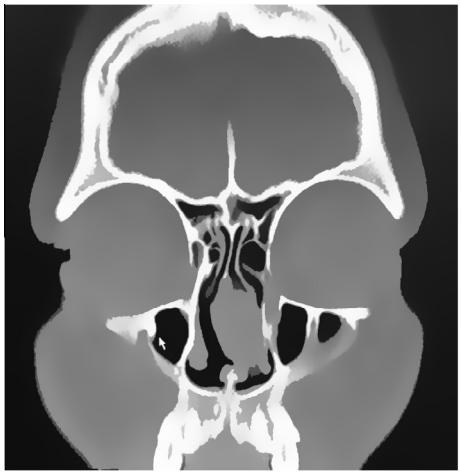

Figure 1 Soft tissue heterogenous enhancement lesion at anterior left nasal cavity causing mild nasal septum deviation to the right. There was heterogenous enhancement seen post contrast and no soft tissue mucosal thickening seen at bilateral maxillary, ethmoidal, frontal and sphenoid sinuses. There was no turbinate hypertrophy, bilateral osteomeatal complexes are patent, orbit and visualized brain is normal. No bony destruction seen.

seen. The lumina contain PAS positive materials that are suggestive of adenoid cystic carcinoma (Fig. 2). All blood investigations were in normal range.

Prior surgery, disease was at stage T3N0M0. Patient underwent tumour resection via combined endoscopic and midfacial degloving, with sublabial incision approach and transfixes incision at interseptal cartilage under general anaesthesia. Intraoperative revealed a mass arised from the anterosuperior septum which was about 1 cm anterior to middle meatus. Both inferior, middle turbinate and OMC were normal. The mucosa of septal cartilage and nasolabial fold which are infiltrated by the mass are resected. Histopathological (HPE) specimen revealed a positive margin of tumour septal root, septal mucosa and nasolabial subcutaneous tissue. The patient was finally diagnosed as having ACC of nasal septum. She then underwent radiotherapy 33 fractions for total 66 Gy. 6 months after surgery follow up showed no recurrence of local and regional of disease.

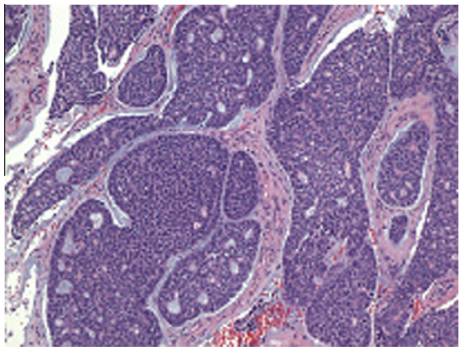

Figure 2 Tumour cells are predominantly in solid nest (60%) and cribriform pattern (40%).

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