International Journal of Surgery Case Reports 94 (2022) 107055

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr

Case report

Adenoid cystic carcinoma of maxillary antrum: A case report

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ARTICLE INFO

Keywords:
Adenoid cystic carcinoma
Biopsy
Case report
Maxillary sinus

ABSTRACT

Introduction: Adenoid cystic carcinoma rarely involves the nose and paranasal sinuses. It is a slowly growing, aggressive malignant tumor with a predisposition for perineural invasion. The study highlights the importance of timely diagnosis and management of adenoid cystic carcinoma.

Presentation of case: Herein, we report a case of adenoid cystic carcinoma of maxillary antrum in a 69 years old female, who presented with an insidious onset of painful right cheek swelling for three months. Biopsy showed the glandular pattern of tumor cells with perineural invasion, suggestive of adenoid cystic carcinoma. Contrast-enhanced computed tomography of paranasal sinuses showed a well-differentiated, irregularly outlined, non-enhancing heterogeneous lesion in the right maxillary sinus with bony destruction, suggesting the malignant nature of the lesion. The mass was surgically excised and histopathological examination further confirmed the diagnosis.

Discussion: Adenoid cystic carcinoma is a rare malignancy of the nose and paranasal sinus, although it is quite common in salivary glands. It should be considered in the differentials in a patient presenting with sinonasal mass, pain, nasal obstruction, epistaxis, and auditory symptoms.

Conclusion: As the tumor can be missed clinically owing to its rarity in paranasal sinuses and can present at an advanced stage, early diagnosis and management of adenoid cystic carcinoma are important.

1. Introduction

Adenoid cystic carcinoma (ACC) is an uncommon malignant tumor arising from the epithelium of mucus glands, usually from salivary glands and rarely from the nose and paranasal sinuses [1,2]. It contributes to less than 1% of all head and neck malignancies and around 5% of paranasal sinus malignancies [1,3]. It is common in the fourth to sixth decade, with a slight female predominance [4]. It is a slow-growing tumor with a predilection for perineural invasion and has a poor prognosis, making the diagnosis and management challenging [1,2]. As it is less commonly encountered, the diagnosis is often missed, leading to improper management [5]. So, timely diagnosis and management are necessary. It should be considered in the differentials in a patient presenting with sinonasal mass, pain, nasal obstruction, epistaxis, and auditory and visual symptoms [5].

Herein we report a rare case of adenoid cystic carcinoma of maxillary antrum in a 69 years old female. The study highlights the importance of an early diagnosis and management of adenoid cystic carcinoma. The case report has been reported in line with the SCARE criteria [6].

2. Presentation of case

A 69 years old female presented to the Ear, Nose, and Throat (ENT) outpatient department (OPD) with an insidious onset swelling of the right cheek for three months measuring four by three cm, with a vague ill-defined margin. The swelling was progressive in nature, firm in consistency, and was associated with pain. The skin over the swelling was tense and the nasolabial fold was found to be distended while the orbit and nose were normal without any distension. There was no associated history of rhinorrhea, cough, otorrhoea, dysphagia, blurring of vision, fever, headache, difficulty in breathing, and epistaxis. She denied the history of tooth extraction in the past. There was no history of trauma to the face. There was no history of similar illnesses in the family.

On intraoral examination, teeth were absent in the upper right

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https://doi.org/10.1016/j.ijscr.2022.107055
Received 12 February 2022; Received in revised form 5 April 2022; Accepted 5 April 2022
Available online 7 April 2022

maxilla and the hard palate on the right side was found to be pushed downward. Biopsy was taken from the right upper alveolus and the histopathological examination using hematoxylin and eosin stain showed a glandular arrangement of tumor cells (Fig. 1) with perineural invasion (Fig. 2), features suggestive of adenoid cystic carcinoma. Contrast-enhanced computed tomography (CECT) of paranasal sinuses showed well-differentiated, irregularly outlined, non-enhancing heterogeneous lesion in the right maxillary sinus and alveolar process of right maxilla with bony destruction suggestive of the malignant nature of the lesion (Fig. 3).

The mass was excised en block with a free margin by Weber-Ferguson incision and was sent for histopathological study. The palate defect was repaired by mobilizing the temporalis muscle flap and the right midfacial defect was repaired by a split-thickness skin graft from the lower limb. Histopathology confirmed the lesion to be adenoid cystic carcinoma of maxillary antrum with perineural invasion. She had no complications after the surgery.

3. Discussion

Adenoid cystic carcinoma is a rare malignancy of the nose and paranasal sinus, although it is quite common in salivary glands [1]. The peak incidence of this tumor is from the fourth to sixth decade, being slightly more common in females, which is similar in our case too [4]. The symptoms of this tumor are non-specific however patients may present with complaints of swelling, pain, nasal obstruction followed by epistaxis, auditory symptoms, nerve symptoms, nasal discharge, and visual symptoms [5,7]. Pain occurs due to perineural invasion of tumor cells and is a common finding in adenoid cystic carcinoma [8]. The facial swelling in our case was also associated with pain, probably due to the perineural invasion. The tumor may involve the nasal cavity alone, nasopharynx alone, maxillary sinus alone, or a combination of all of these [5].

This is a slow-growing malignant tumor that is characterized by wide local infiltration, perineural spread, predilection to local recurrence, and distant metastasis [3]. The growth pattern of this tumor can be an expansile type with minimal bony defect or a destructive type with extensive bony defect [9]. It may be aggressive but indolent typically presenting at the advanced stage reflecting a poor prognosis of a patient [7].

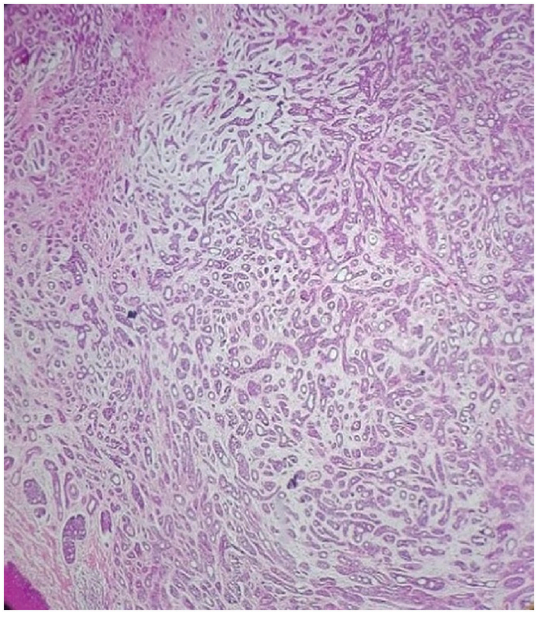

Fig. 1. Low power microscopic view showing the glandular pattern of tumor cells.

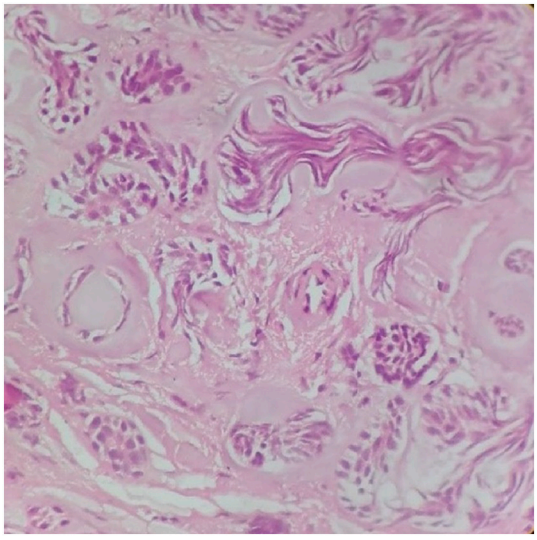

Fig. 2. High power microscopic view showing the perineural invasion by tumor cells.

Histopathologically, the tumor can be invasive with bony, neural, and lymphovascular invasion, consisting of a variety of growth patterns including cribriform, tubular, and solid. [5,8]. The cribriform type is related to a better prognosis than the solid type [8]. Multiple modalities of diagnosis have been described for this tumor including computerized tomography (CT Scan) and magnetic resonance imaging (MRI) which may show local expansile and destructive patterns of the tumor as well as distant metastasis [9]. Other modalities include fine-needle aspiration cytology (FNAC) and histopathological examination. CT scan and histopathological examination with hematoxylin and eosin stain were used in our case for the diagnosis of adenoid cystic carcinoma. It can also be confirmed immunohistochemically by the presence of a positive reaction for pan-cytokeratin, CK7, CK5/6, CAM5.2, and EMA, with myoepithelial reactivity with SMA, P63, Calponin, S100 protein, SMMSC, CD117, CEAGFAP, and P16 [5]. Polymorphous low-grade adenocarcinoma, basaloid squamous cell carcinoma, adenosquamous carcinoma, and small cell neuroendocrine carcinoma can be considered in the differential diagnosis of adenoid cystic carcinoma [10].

Among the various treatment modalities for this tumor, surgery followed by radiation therapy remains the first choice of treatment, providing the best overall survival and control [1]. The surgical procedures may be open or endoscopic. The choice of surgery depends upon the extent of the tumor, accessibility, and the bone that has to be removed. However, functional endonasal sinus surgery, a minimally invasive technique is the preferred surgical approach in the treatment of adenoid cystic carcinoma of the nose and paranasal sinuses [11]. The tumor in our case was excised en block with a free margin by Weber-Ferguson incision along with the repair of the palatal defect and midfacial defect.

The outcome of the treatment and recurrence of the tumor depends upon staging, skull base involvement, solid histology, perineural, bony, and lymphovascular invasion [5]. The overall and disease-free 5-year survival rate with surgery and adjuvant therapy is 63% and 43% respectively [1]. The prognosis of the patient depends on the clinical stage, histologic subtype, treatment approach, and distant metastasis [1]. Solid histology, advanced stage, perineural invasion, and positive surgical margins were associated with poor prognosis [11,12].

4. Conclusion

Adenoid cystic carcinoma, being an uncommon tumor in the paranasal sinuses can be missed clinically and can present in an advanced

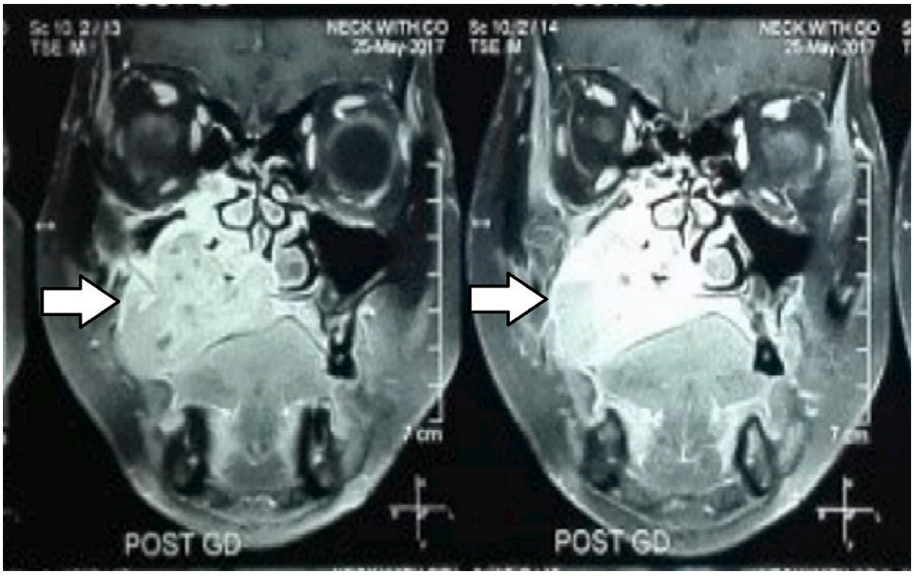

Fig. 3. CT scan showing involvement of right maxillary sinus and extension to the hard palate.

stage reflecting a poor prognosis. So, timely diagnosis and management of the case are crucial. Hence, it should be considered in the list of differentials when a patient presents with a sinonasal mass, pain, nasal obstruction, epistaxis, and auditory symptoms.

Funding

None.

Ethical approval

Not applicable.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in Chief of this journal on request.

Author contribution

Author 1: Led data collection, contributed to writing the case information.

Author 2: Literature review, prepared initial manuscript draft.

Author 3: Literature review, contributed to writing an introduction.

Author 4: Literature review, prepared final manuscript draft.

Author 5: Literature review, revising, and editing the manuscript.

All authors were involved in manuscript drafting and revising, and approved the final version.

Research registration number

Not applicable.

Guarantor

Saurab Karki.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Declaration of competing interest

None.

Acknowledgement

None.

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