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Review

59 years old male with local advanced adenoid cystic carcinoma of the hypopharynx – A case report and a review of the literature

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ABSTRACT

Minor Salivary Gland tumours are rare, representing 2% of the head and neck tumours and less than 1% of hypopharyngeal cancers. The most common subtype of minor salivary gland tumours is adenoid cystic carcinoma. Although there are several case reports discussed it, there are only two reports discussing hypopharyngeal presentation in the literature, and they were treated by surgical resection. Despite that, our case report still to be unique as it presents a case of irresectable locally advanced tumour subjected to a trial of induction therapy.

Introduction

Anatomically, the palate is the commonest site of minor salivary gland tumours, followed by paranasal sinuses, nasal cavity and the larynx [1]. Adenoid cystic carcinoma (AdCC) is the most common subtype of minor salivary gland tumours representing around 10% of all malignant salivary gland tumour [2]. The main cornerstone of treatment is the surgical resection while the role of radiotherapy and chemotherapy are still controversial [3]. Several prognostic factors have been identified such as free resection margin, nodal negativity and negative neural invasion [3].

However, AdCC of the hypopharynx is extreme rare disease, thus, extrapolating the clinical behaviour of laryngeal and pharyngeal counterparts is acceptable [2]. Generally, the disease is slowly growing but it has extensive invasiveness potential and high recurrence rate following resection. Likely as its counterparts in hard palate, adenoid cystic carcinoma is characterized by poor responsiveness to chemotherapy and radiotherapy [2,3].

The driver mechanism of adenoid cystic carcinoma is unknown. They arise between the fifth and sixth decade of life as submucosal mass and hence delayed diagnosis until advanced stages [3].

Case report

A 59-years old male smoker was presented with a history of cervical lymph node enlargement, stridor managed by urgent tracheostomy.

CT and MRI scanning revealed, large retro-laryngeal mass extending from the level of 4th cervical to 2nd dorsal vertebrae measuring $8 * 4.5 * 6$ cm in maximal Cranio-caudal, transverse and anteroposterior dimensions, respectively. The mass is infiltrating the internal Jugular, the thyroid and prevertebral fascia (see Fig. 1). CT chest and the abdominal US excluded metastatic spread.

His endoscopy revealed a large post-cricoid mass disturbing the laryngopharyngeal anatomy. Multiple biopsies were taken and examined pathologically showing low-grade uniform cells with adenoid cystic differentiation. Immune staining revealed positive CK, CD117 and S100 beside negative TTF-1 supporting the diagnosis of AdCC.

Due to the rarity of the disease and lack of supporting guidelines, the patient received a trial of Docetaxel 80 mg/m^2 [D1], Cisplatin 100 mg/m^2 [D1] and 5-fluorouracil 1000 mg/m^2 [D1-4] for two cycles. The first cycle was well-tolerated, however, in the eighth day following the second cycle, he developed uncontrollable neutropenia, mucositis and diarrhoea that is complicated by death.

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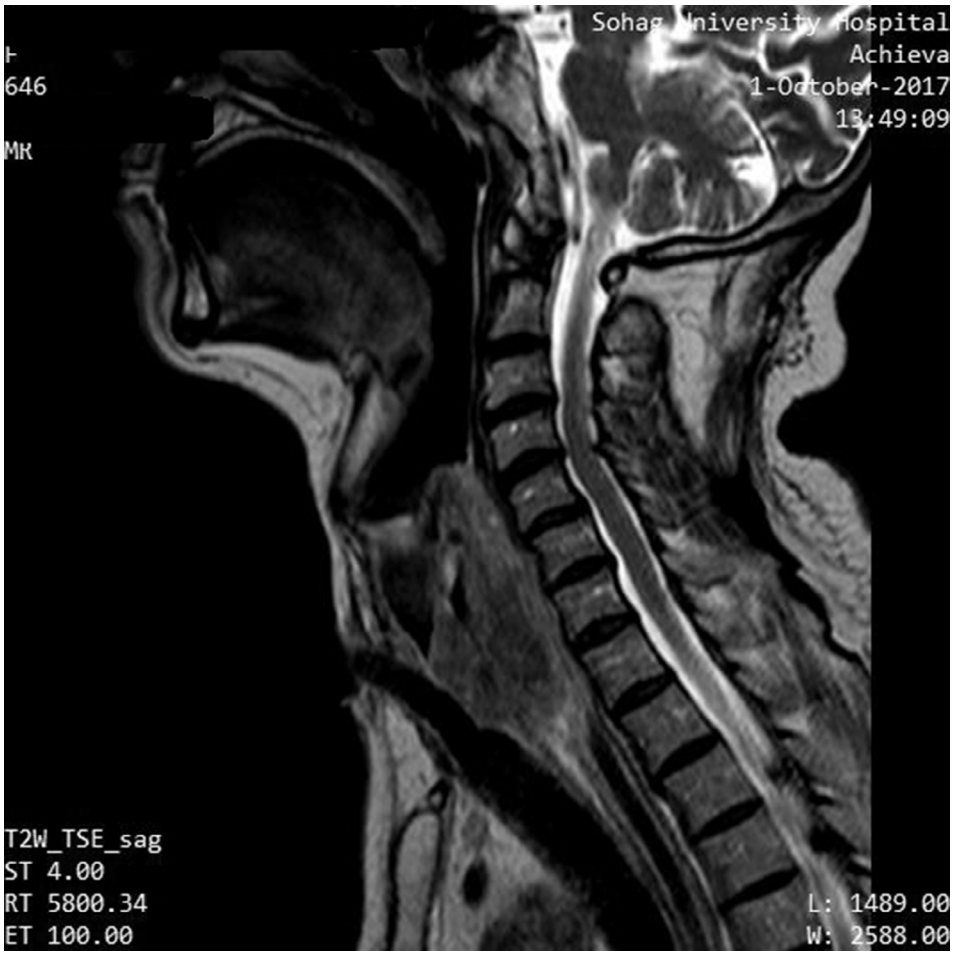

Fig. 1. Sagittal T2W MRI - infiltration of the pre-vertebral fascia.

Discussion

The most common tumours in the hypopharynx are the squamous cell carcinoma (90%) followed by other histological types. AdCC usually grows as a submucosal mass that is barely detected at earlier stages [1,2]. Adenoid cystic carcinoma occurs in old age groups with an equal male to female distribution. Symptoms are usually related to dysphagia, especially for solids.

AdCC usually recurs post-resection especially in patients with narrow resection margins. Several factors have been linked to improved 5-years survival rate; negative resection margin, nodal negativity and absence of neural invasion. Although surgical resection of AdCC is considered the standard whenever possible, other modality of treatment such as radiotherapy and chemotherapy still controversial. Although several publications pointed to the use of palliative chemotherapies; either taxane-based or anthracycline-based combination, none of them showed any survival benefit [4,5].

Few authors reported cases of AdCC in rare anatomical sites such as larynx, however, only two authors reported two cases of hypopharyngeal AdCC which were detected in an earlier stage and treated by surgical resection [3,6]. Our case is still to be unique since it represents a locally irresectable disease in the hypopharyngeal region and is treated by docetaxel, cisplatin and 5-fluorouracil combination. Despite the negative outcome; death of the patient, it is irrational to judge a regimen's efficacy and response from one case study.

Conclusion

Minor salivary gland cancer is one of the rare head and neck neoplasms. Adenoid cystic carcinoma is the most common histological subtype to be seen within that group. The commonest site for AdCC is the hard palate, paranasal sinuses and nasal cavity. Other sites such as larynx and hypopharynx are rare to be seen. Our case is unique and distinct since it represents a patient with irresectable late stage AdCC of the hypopharynx. Unfortunately, our trial of docetaxel cisplatin and 5-fluorouracil failed to show any response in that patients. Further studies of such disease are necessary for the proper definition of treatment guidelines.

Conflict of interest

The author(s) indicated no potential conflicts of interest.

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