

CASE REPORT

Adenoid Cystic Carcinoma of Larynx: A Case Report and Rare Experience

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ABSTRACT

Adenoid cystic carcinoma (ACC) accounts for less than 1% of laryngeal tumors. Surgery is the treatment of choice which can be combined with adjuvant radiotherapy. We present a rare case of ACC of glottis and subglottis presented to us with respiratory distress.

Keywords: Adenoid cystic carcinoma, Perineural, Subglottis, Surgery.

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INTRODUCTION

Squamous cell carcinoma is the most common histological type of laryngeal tumor. Adenoid cystic carcinoma (ACC) of the larynx is one of the rarest variety and it account for <1% of laryngeal tumors.^{1,2} The local recurrence and distal metastasis is the usual behavior of ACC. The causes behind it are early perineural invasion and blood-borne metastasis.³

We present a rare case of ACC of glottis and subglottis, presented to us with long history and with respiratory distress.

CASE REPORT

Twenty-four-year-old female patient presented with history of difficulty in breathing for the last 2 years. This

was insidious in onset, persistent and gradually progressive such that the patient presented to our emergency department with stridor. It was associated with hoarseness for the past 3 to 4 months, which was also persistent and progressive. There was no history of difficulty in swallowing, neck swelling, fever, weight loss, or loss of appetite. No history suggestive of any addiction. There was no significant family or past history of any chronic illness. On indirect laryngoscopic examination, multiple polypoidal tissue was filling endolarynx at the level of true cords. Clinical diagnosis of laryngeal papillomatosis was made and the patient was immediately shifted to operation theater for removal by direct laryngoscopy under general anesthesia. The ulceroproliferative growth was found under polypoidal mass and it involved glottis and 2 cm of subglottis (Fig. 1). The lesion was debulked and histopathology report showed feature of ACC. The patient underwent investigations, which include computed tomography scan of neck and it showed soft tissue density in left paraglottic space (Fig. 2). Magnetic resonance imaging (MRI) showed hyperintense lesion in left side of glottis and subglottis (Fig. 3). The mass lesion in endolarynx was not seen in radiology because of previous debulking of lesion. The patient was planned for wide field laryngectomy with partial pharyngectomy with left hemithyroidectomy under general anesthesia. Growth was involving left ventricle left vocal cord and left side of subglottis (Fig. 4). Postoperative period was

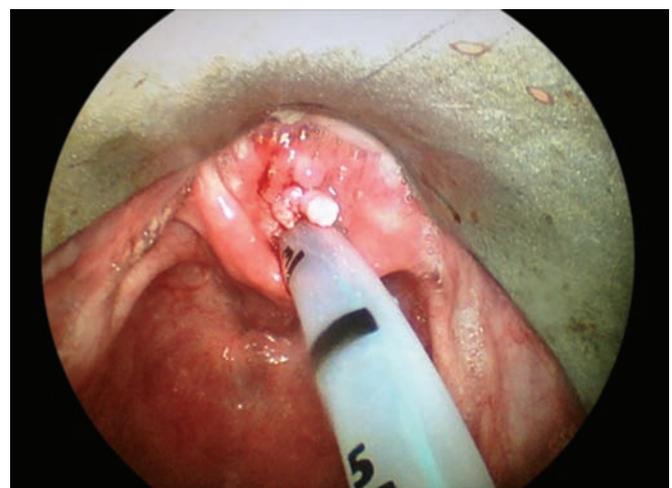


Fig. 1: Direct laryngoscopic photograph showing papillomatous tissue filling endolarynx

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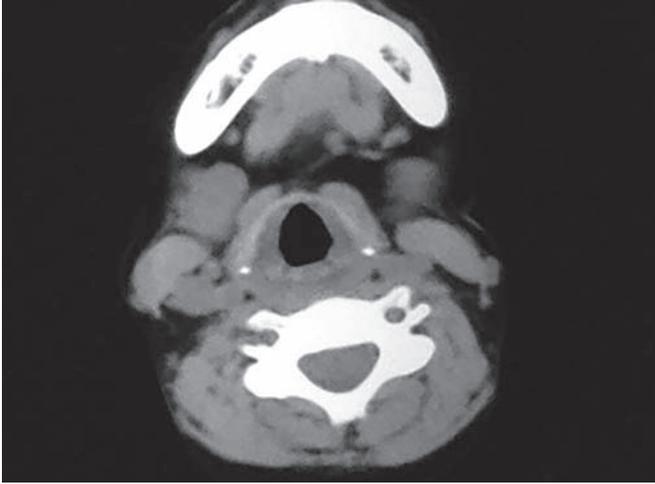


Fig. 2: The CECT showing edema in left paraglottic space

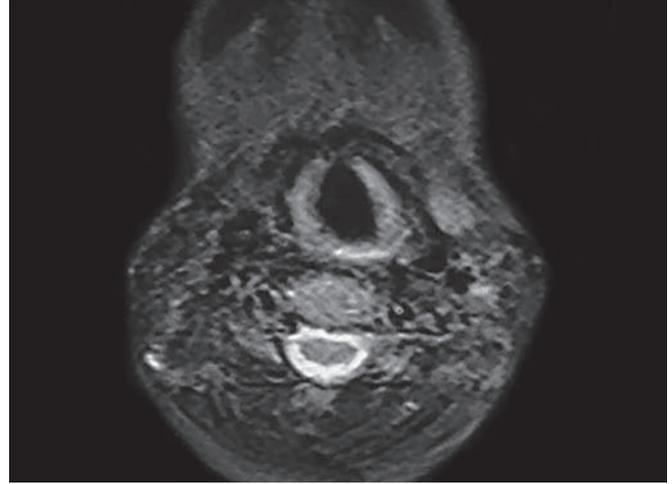


Fig. 3: The MRI showing isointense left paraglottic space



Fig. 4: Post laryngectomy specimen showing edematous left ventricular space

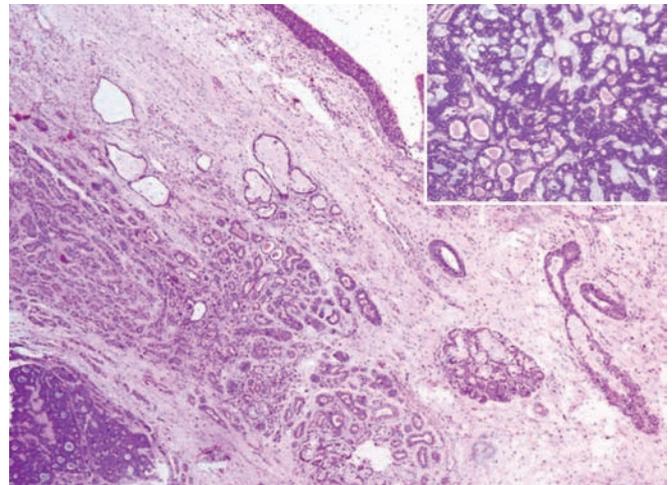


Fig. 5: Adenoid cystic carcinoma: Photomicrograph showing duct lining cell arranged in cribriform pattern with cyst-like spaces (H&E 40 \times). Inset showing higher magnification of tumor (H&E 100 \times)

uneventful. Oral trial was given on 10th postoperative day. The final histopathology report showed arrangement of basaloid epithelial cells in tubules and cribriform pattern, suggestive of ACC without epithelium and cartilage involvement with free resection limits and lymph nodes (Fig. 5). Patient received radical radiotherapy. She is disease-free at 6 months follow-up.

DISCUSSION

The minor salivary glands are found in low density in laryngeal mucosa. The salivary gland tumors are rare in larynx. Adenoid cystic carcinoma is the commonest salivary gland tumor of larynx.^{4,5} The density of seromucinous glands is reducing from supraglottis to subglottis.⁶ In spite of that, subglottis is the most frequent site of origin. As per the literature, the subglottis accounts for 64%, supraglottis accounts 25%, and glottis accounts 5% of total laryngeal ACC.⁷

The age range for ACC has been documented in the literature from 3 to 8 decades of life with peak at 5th decade with slight female preponderance, whereas our patient is relatively young.⁸ The risk factor specific for ACC is not mentioned in the literature.⁹

The clinical feature of laryngeal ACC is based on site and size. Supraglottic lesions have dysphagia whereas hoarseness is seen in glottis lesion. The subglottic lesion usually presented with dyspnea and stridor. The subglottic lesion is generally diagnosed late and they are in advance stage.¹⁰ Zvrko and Golubovic¹¹ proposed prelaryngeal pain is the only symptom even in advance stage.

The cribriform, tubular, and solid are three histopathological patterns of ACC. The cribriform have best prognosis whereas solid have worse prognosis.⁹

Precise mapping and staging are essential for planning and prognosis of disease. The contrast enhanced computed tomography (CECT) is the usual investigation

for mapping and staging of tumor. The ACC has characteristic submucosal extension and perineural invasion. In such suspected cases, MRI will be better modality.¹¹ The nodal extension is seen in 10 to 15% of cases.¹²

Surgery with or without adjuvant radiotherapy is the proposed management for laryngeal salivary gland tumors. Conservative laryngectomy can be possible in early stages.¹³ The characteristic feature of ACC is submucosal spread and perineural and lympho-vascular invasion. Total laryngectomy is the usual treatment of choice.⁷ Neck dissection is not recommended in N0 neck.^{3,7-9,14-16} Primary radiotherapy has a minor role in treatment.⁸ The prime indications for radiotherapy are positive surgical margins, perineural invasion, or high-grade tumors. The role of chemotherapy is in palliative care patient and as adjuvant therapy for high-grade lesions.

The patient requires long-term follow-up as the local and distal recurrence can appear even after many years.^{4,11,14,17} This patient is under follow-up for 2 years without local and distal recurrence.

CONCLUSION

This tumor is characterized by slow progression, multiple local recurrences, and distant metastasis. The treatment of choice is surgery and adjuvant radiotherapy with long-term follow-up.

REFERENCES

1. Fordice J, Kershaw C, El-Naggar A, Goepfert H. Adenoid cystic carcinoma of the head and neck: predictors of morbidity and mortality. *Arch Otolaryngol Head Neck Surg* 1999 Feb;125(2):149-152.
2. Batsakis JG, Luna MA, El-Naggar AK. Nonsquamous carcinomas of the larynx. *Ann Otol Rhinol Laryngol* 1992 Dec;101(12):1024-1026.
3. Ferlito A, Barnes L, Myers EN. Neck dissection for laryngeal adenoid cystic carcinoma: is it indicated? *Ann Otol Rhinol Laryngol* 1990 Apr;99(4 Pt 1):277-280.
4. Moukarbel RV, Goldstein DP, O'Sullivan B, Gullane PJ, Brown DH, Wang L, Irish JC. Adenoid cystic of the larynx: a 40-year experience. *Head Neck* 2008 Jul;30(7):919-924.
5. Idowu MO, Reiter ER, Powers CN. Adenoid cystic carcinoma: a pitfall in aspiration cytology of the thyroid. *Am J Clin Pathol* 2004 Apr;121(4):551-556.
6. Nassar VH, Bridger GP. Topography of the laryngeal mucous glands. *Arch Otolaryngol* 1971 Dec;94(6):490-498.
7. Dexamble P, Huth J, Rebufy M, Chabrol A. Carcinome adénoïdekystique du larynx. A propos de deux cas. *Ann Otolaryngol Chir Cervicofac* 2003 Sep;120(4):244-248.
8. Oloffon J, van Nostrand AW. Adenoid cystic carcinoma of the larynx: a report of four cases and a review of the literature. *Cancer* 1977 Sep;40(3):1307-1313.
9. Tincani AJ, Del Negro A, Araújo PP, Akashi HK, Martins AS, Altemani AM, Barreto G. Management of salivary gland adenoid cystic carcinoma: institutional experience of a case series. *Sao Paulo Med J* 2006 Jan 5;124(1):26-30.
10. Saraydaroglu O, Coskun H, Kasap M. Unique presentation of adenoid cystic carcinoma in postcricoid region: a case report and review of the literature. *Head Neck Pathol* 2011 Dec;5(4):413-415.
11. Zvrko E, Golubovic M. Laryngeal adenoid cystic carcinoma. *Acta Otorhinolaryngol Ital* 2009 Oct;29(5):279-282.
12. Ganly I, Patel SG, Coleman M, Ghossein R, Carlson D, Shah JP. Malignant minor salivary gland tumors of the larynx. *Arch Otolaryngol Head Neck Surg* 2006 Jul;132(7):767-770.
13. Alavi S, Calcaterra TC, Namazie A, Blackwell KE. Glandular carcinoma of the larynx: the UCLA experience. *Ann Otol Rhinol Laryngol* 1999 May;108(5):485-489.
14. Browne JD. Management of the nonepidermoid cancer of the larynx. *Otolaryngol Clin North Am* 1997 Apr;30(2):215-229.
15. Veivers D, Vito AD, Luna-Ortiz K, Brasnu D, Laccourreye O. Supracricoid partial laryngectomy for non-squamous cell carcinoma of the larynx. *J Laryngol Otol* 2001 May;115(5):388-392.
16. Javadi M, Bafrouee FM, Mohseni M, Asghari A. Laryngeal adenoid cystic carcinoma in a child: a case report. *Ear Nose Throat J* 2002 Jan;81(1):34-35.
17. Del Negro A, Ichihara E, Tincani AJ, Altemani A, Martins AS. Laryngeal adenoid cystic carcinoma: case report. *Sao Paulo Med J* 2007 Sep 6;125(5):295-296.