World Journal of Advanced Research and Reviews
eISSN: 2581-9615 CODEN (USA): WJARAI
Cross Ref DOI: 10.30574/wjarr
Journal homepage: https://wjarr.com/

(CASE REPORT)

Definitive chemoradiotherapy for adenoid cystic carcinoma of the trachea: A case report

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World Journal of Advanced Research and Reviews, 2023, 20(02), 1082–1086

Publication history: Received on 25 September 2023; revised on 18 November 2023; accepted on 20 November 2023

Article DOI: https://doi.org/10.30574/wjarr.2023.20.2.2355

Abstract

Adenoid cystic carcinoma is a malignant tumour rarely found in trachea, and even less so in our sub-region. In addition to a slow clinical course, this histology has a predilection for perineural invasion and a tendency for local and distant recurrence. Most patients present with dyspnea and symptoms often mimic those of asthma or chronic bronchitis. Although surgical resection is the mainstay of treatment, tumour size and location, comorbidities and patient reluctance may prevent surgery. Furthermore, the recognized chemoresistance of this histological type means that radiotherapy, whether definitive or adjuvant, plays a major role in its management. We report the case of a 30-year-old patient treated in our department with definitive chemoradiotherapy.

Keywords: Adenoid cystic carcinoma; Trachea; Radiotherapy; Senegal

1. Introduction

Adenoid cystic carcinoma is a malignant tumour arising in the salivary glands. Billroth [1] first described its clinical and histopathological aspects in 1869. Formerly known as cylindroma, it accounts for 0.09% to 0.2% of respiratory tract tumors [2]. Tracheal location is rarely found in the literature. Adenoid cystic carcinoma is the second most common primary malignant tumour of the trachea [3]. Its natural history is characterized by progressive airway obstruction leading to inspiratory dyspnea [4]. The mainstay of treatment is sleeve resection of the pathological segment of the trachea (5). Post-operative radiotherapy, or exclusive radiotherapy in the event of inoperability, has been shown to be effective, unlike conventional chemotherapy [6,7]. Despite this, there are very few studies of radiotherapy for this rare site in the local and global literature. We report the case of a patient treated with concomitant chemoradiotherapy.

2. Observation

The patient was 30 years old, had taken injectable hormonal contraception for 03 years and had no history of alcohol or tobacco abuse. She had been presenting with inspiratory dyspnea for about 1 year. The initial clinical examination on 10 January 2022 revealed stage II laryngeal dyspnea, associated with a firm anterior cervical swelling that was mobile on swallowing and painless. The superficial cervical lymph nodes were free. Indirect laryngoscopy revealed no lesions. Cervical computed tomography revealed a faint tissue thickening located between the trachea and the thyroid with a maximum thickness of 14 mm. The thoracic and thyroid workups were unremarkable. Panendoscopy with multiple biopsies revealed a bulging lesion in the trachea, stenosing approximately 80% of the tracheal lumen. A subisthmic tracheotomy was performed for stage III laryngeal dyspnea.

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Given the patient's refusal to undergo surgery and the prolonged delay for radiotherapy, induction chemotherapy followed by ~~concurrent radiochemotherapy~~ concomitant chemoradiotherapy was prescribed after a multidisciplinary consultation meeting. The patient underwent multidrug therapy with 5 cycles of cisplatin, doxorubicin and cyclophosphamide. A post-treatment extension assessment was carried out, consisting of a cervico-thoraco-abdomino-pelvic CT scan, which revealed tracheal tissue thickening extending over approximately 43.8 mm, with a maximum measured thickness of 11.2 mm, without complete obstruction of the airway and with no secondary lesions, giving an estimated response of 20%.

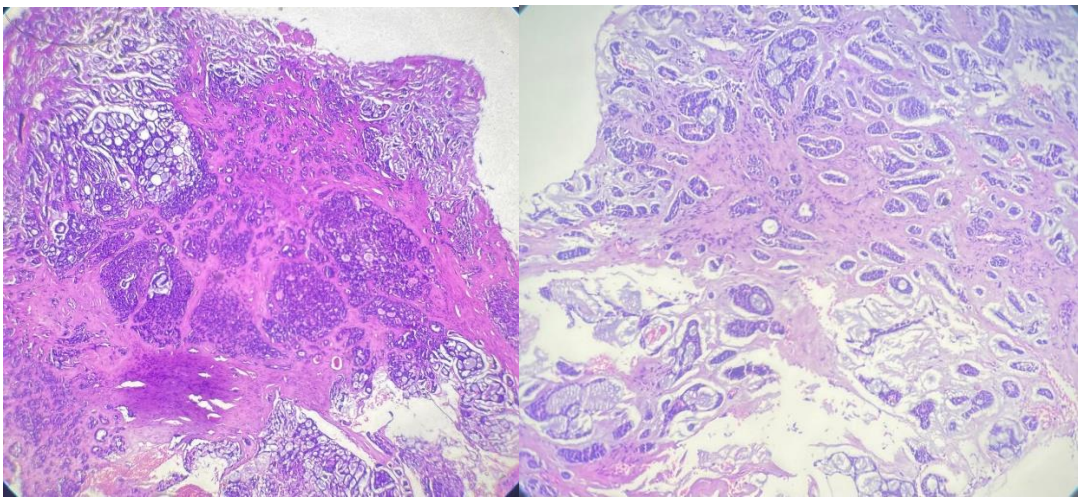

Figure 1 Haematoxylin and eosin (HE) staining revealed cystic adenoid carcinoma (low and medium magnification).

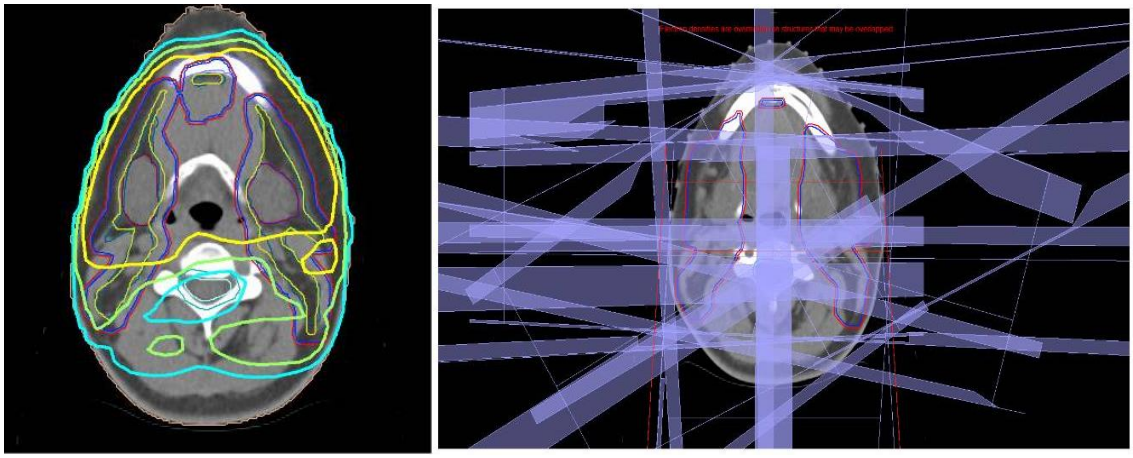

Figure 2 Ballistics, isodose curves

The patient then underwent radiotherapy over a period of 8 weeks. The technique used was 3D conformal without intensity modulation. She received 70 Gy to the primary tumour and cervical lymph nodes in 35 fractions of 2 Gy. The clinical target volume (CTV) retained included the tumour bed and lymph node areas Ia, Ib, II, III, IV and VI bilaterally with isotropic margins of 5 mm for the high-risk clinical target volume and 10 mm for the low-risk volume, while respecting anatomical barriers. The planning target volume (PTV) was delineated with 10mm margins around the CTV. Figure 3 shows the dose distribution in the treatment plan in coronal ~~and sagittal~~ sections. The patient was treated with 5 beams (anterior, posterior, right lateral, left lateral and right oblique). Radiotherapy was given concomitantly with weekly cisplatin for 5 weeks. The patient presented with grade II cervical radiodermatitis and grade II odynophagia according to CTCAE version 4.0, as well as moderate asthenia, which progressed well with medication and dietary hygiene measures. One month after radiotherapy, there was a marked regression in respiratory symptoms (inspiratory dyspnea) and disappearance of the anterior cervical swelling, but grade II radiodermatitis persisted. Local treatments were prescribed, and a post-treatment assessment requested, which the patient was unable to carry out due to lack of funds. She presented with malnutrition exacerbated by odynophagia, intolerance of the nasogastric tube and refusal of

a gastrostomy. She died ,6 months after completing treatment, of severe malnutrition complicated by multiple organ failure.

3. Discussion

Tracheal localization of adenoid cystic carcinoma is relatively rare. Tracheal tumours represent barely 0.2% of respiratory tract neoplasia in the United States and the dominant histological type is squamous cell carcinoma. Only 10% of tracheal tumours have adenoid cystic carcinoma as the histological type [8]. In Africa, the literature remains sparse, although cases have been reported in recent years in North Africa [3,9]. In Senegal, only three cases of cystic adenoid carcinoma of the head and neck have been found in the literature, and tracheal location accounted for only 0.92% of cases [10,11]. Surgical management remains the cornerstone of treatment. Radiotherapy has been shown to be effective both as an exclusive treatment and in the adjuvant setting [7,12,13].

The technique used to treat our patient is three-dimensional conformal radiotherapy, which is the most accessible in our context. It was recommended for a long time in this location until the advent of IMRT, which has proved its superiority in head and neck locations in terms of sparing organs at risk [6,14]. Today, even more advanced techniques such as tomotherapy have demonstrated their value in the treatment of adenoid cystic carcinoma of the trachea [15].

The contouring of the target volumes was based on the natural history, characterized by the propensity of this type of histology for submucosal extension and peri-nervous invasion [16]. This extension, particularly in height, led Kaminski et al to propose margins of 4-5 cm craniocaudal [17].

Adenoid cystic carcinoma is a tumour considered to be radiosensitive but difficult to radiocure; nevertheless, this histological type is a favorable prognostic factor. Le Péchoux et al. reported a 5-year overall survival of 65-85% after surgery or with the combination of surgery and post-operative radiotherapy with a dose of 45-60 Gy [16]. In patients deemed inoperable, or where surgery is refused, as in the case of our patient, radiotherapy is proposed at a dose of 66-70 Gy in standard fractionation of 1.8 - 2 Gy [17]. Complete responses have been observed in several series and appear to be linked to doses that should be higher than 60 Gy in the case of exclusive radiotherapy [18-20]. The relative rarity of adenoid cystic carcinoma in general and adenoid cystic carcinoma of the trachea in particular has limited the number of disease-specific chemotherapy trials. Responses have been observed with older agents as monotherapy, including 5-flourouracil, methotrexate, doxorubicin, cyclophosphamide, and cisplatin, although these reports are largely anecdotal [17]. Platinum-based combinations comprising cyclophosphamide, doxorubicin, and cisplatin (CAP) have also been studied [21].

For tracheal tumours, exclusive radiotherapy, whatever the histological type, results in a median survival of around 30 months and a 5-year survival of over 40%; in palliative situations, a median survival of around 10 months can be achieved [13,22]. However, as with most head and neck cancers, the prognosis may be adversely affected by factors such as poor general condition or nutritional imbalance [23]. Recommendations are clear on the need to assess the nutritional status of patients undergoing treatment for cancers of the upper aerodigestive tract, in view of the morbidity associated with the risk of complications and impairment of quality of life [24].

4. Conclusion

The complexity of this case report is due not only to the rarity of the histological type and location, particularly in our country, but also to its radiobiological characteristics and chemoresistance, which compromise response to treatment and prognosis. However, although the cornerstone of treatment remains surgery, advances in radiotherapy have clearly improved survival in these patients, whether exclusive or adjuvant.

Compliance with ethical standards

Acknowledgements

Thanks to the International Atomic Energy Agency (IAEA).

Disclosure of Conflict of interest

The authors have declared no conflicts of interest.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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