Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Pathology

Radiation-Induced Leomyosacroma of the Larynx: A Case Report and Literature Review

Salma El Alaoui El Rhoul[1*], Hajar El Youbi[1], Abdellatif Oudidi[1], Nawal Hamass[2], Najib Benmansour[1], Naouar Ouattassi[1], Mohammed Ridal[1], Mohammed Noureddine El Amine El Alami[1]

[1]ORL Head & Neck Department, Hassan II University Hospital
[2]Pathology Department, Hassan II University Hospital

DOI: https://doi.org/10.36347/sjmcr.2025.v13i03.038 | **Received:** 17.02.2025 | **Accepted:** 21.03.2025 | **Published:** 26.03.2025

***Corresponding author:** Salma El Alaoui El Rhoul
ORL Head & Neck Department, Hassan II University Hospital

Abstract **Case Report**

There is a clear association between radiotherapy and an increased risk of developing secondary cancers, including sarcomas. Leiomyosarcoma is one of the rare secondary tumors reported after radiotherapy treatment. We report a rare case of laryngeal leiomyosarcoma in a 72-year-old patient with a history of radiotherapy for nasopharyngeal carcinoma treated seventeen years earlier. The patient mainly presented with progressively worsening dysphagia. The treatment consisted of surgical excision followed by adjuvant radiotherapy. The outcome was favorable. Through this case report, we aim to highlight this rare secondary tumor and its association with the effects of radiotherapy. We will also review the main therapeutic modalities for this entity.

Keywords: Leiomyosarcoma, Larynx, Post-Radiation, Total Laryngectomy, Case Report.

INTRODUCTION

Leiomyosarcoma (LMS) is a rare mesenchymal tumor that arises from smooth muscle cells. It accounts for 5 to 10% of all soft tissue sarcomas [1]. LMS of the head and neck represents 3% of all LMS cases. Sarcomas constitute a very small proportion of laryngeal malignancies, accounting for only 1% of all head and neck tumors [2]. Laryngeal localization is extremely rare (fewer than 50 reported cases) [2].

CASE REPORT

Patient Presentation:

A 72-year-old male patient, with a medical history of nasopharyngeal tumor undifferentiated carcinoma of nasopharyngeal type (UCNT) seventeen years ago, presented to the ENT outpatient clinic with four months history of dysphagia, choking and dysphonia. Nasopharyngeal UCNT was treated with concomitant chemoradiotherapy, achieving complete remission with no sign of recurrence after long-term follow-up.

Clinical Findings:

Laryngeal fibroscopy revealed a supraglottic process involving the epiglottis and extending to the right vallecula (Figure 1). Cervical examination was unremarkable.

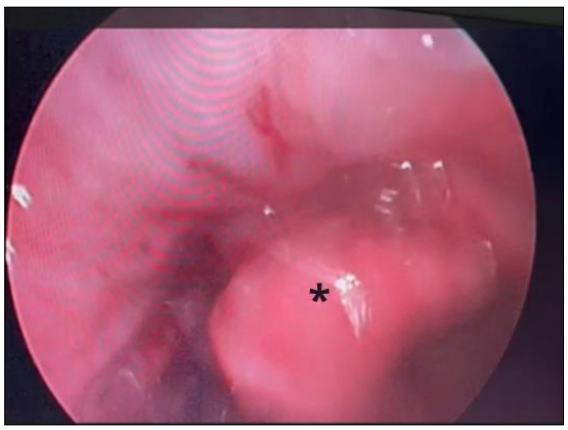

Figure 1 : Endoscopic laryngeal view showing a deformed epiglottis (asterisk) with visualization of a tumoral process invading the right vallecula.

Diagnostic Approach:

Laryngeal magnetic resonance imaging (MRI) showed a supraglottic soft tissue mass extending to the pre-epiglottic space and filling the right vallecula, measuring 27 × 20 cm, with no cervical lymphadenopathy (Figure 2).

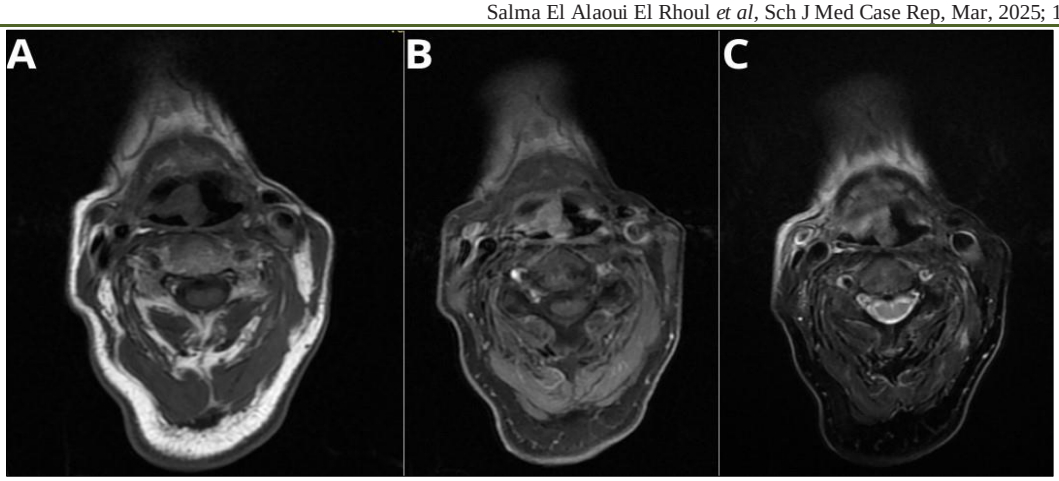

Figure 2 : Axial cervical MRI demonstrating an epiglottic tumor extending into the right vallecula. The lesion appears as an intermediate T1 signal (A), exhibits contrast enhancement following Gadolinium administration (B), and shows a heterogeneous hyperintense signal on T2-weighted imaging (C).

We performed a panendoscopy that reveals a tumor process of the epiglottis extending to the right vallecula (Figure 3). We completed by multiple tumor biopsies. Histological and immunohistochemical analysis confirmed the diagnosis of leiomyosarcoma (Figure 4). Staging workup showed no distant metastases. We therefore classify this supraglottic tumor as T2 N0 M0.

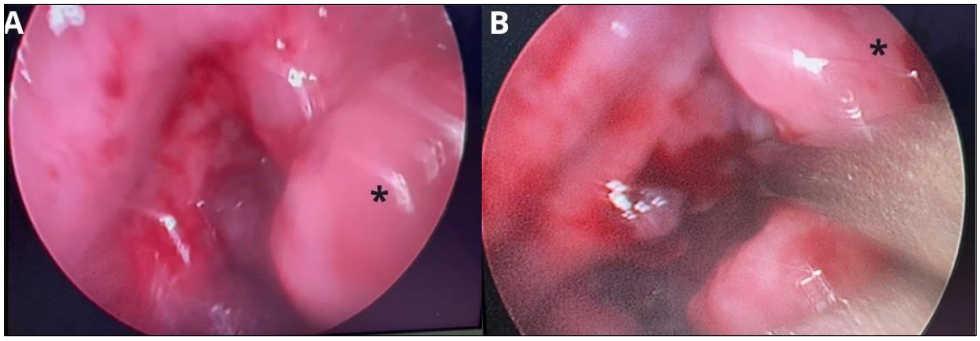

Figure 3 : Endoscopic view of the larynx showing an epiglottic tumor (asterisk) invading the right vallecula (A) with tumor biopsies being performed (B).

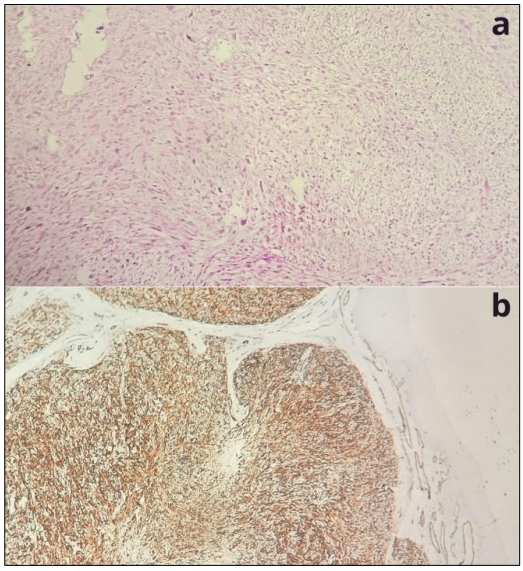

Figure 4 : Histological and immunohistochemical (IHC) features suggestive of leiomyosarcoma. (a) Laryngeal mucosa with a tumoral proliferation composed of atypical spindle cells (HES×100). (b) Immunohistochemical expression of α-SMA (alpha-smooth muscle actin).

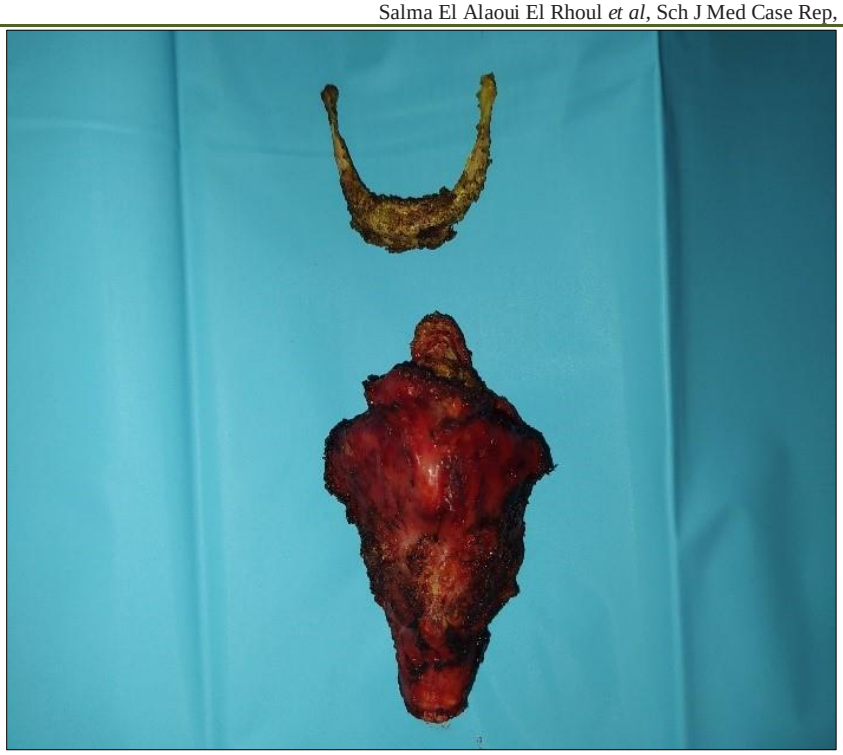

Figure 5 : Anatomical specimen of total laryngectomy with excision of the hyoid bone.

Therapeutic Intervention:

The multidisciplinary tumor board recommended surgical excision followed by adjuvant radiotherapy. The patient underwent total laryngectomy (Figure 5), confirming the diagnosis of laryngeal leiomyosarcoma with negative lymph node involvement. Postoperative recovery was uneventful, with good wound healing and resumption of oral feeding 15 days postoperatively. The patient then received postoperative radiotherapy with a dose of 60 Gray in 30 fractions to the tumor bed.

Follow-up and Outcomes:

During radiotherapy, the patient developed Grade 1 radiation dermatitis, which improved with local treatment. At one year of follow-up, the patient remained in complete remission. However, a 19 mm pulmonary nodule with secondary characteristics and two additional subcentimeter nodules were detected. The decision was made to administer stereotactic body radiotherapy (SBRT) with 56 Gray in 8 fractions for the largest nodule and 45 Gray in 3 fractions for the smaller lesions. At present, the patient has completed two years of follow-up with stable clinical and radiological findings.

DISCUSSION

Beyond its cytotoxic effect on malignant cells, radiotherapy is also a well-known mutagen capable of inducing DNA damage and chromosomal aberrations. These genetic modifications can lead to either cell death or potential malignant transformation of surviving cells [3].

To diagnose radiation-induced malignancy (RIM), the modified Cahan criteria are often used [4] :

1) The RIM must have developed within a previously irradiated field;
2) A latency period of more than four years must have elapsed between the initial irradiation and the presumed RIM;
3) The irradiated tumor and the presumed RIM must have different histology confirmed by biopsy;
4) The tissue where the presumed RIM developed must have been normal before radiation exposure.

Our case meets these criteria. Radiation-induced cancers encompass a wide variety of histological types. Carcinomas and leukemias are frequently observed in organs receiving low doses of radiation and in regions distant from the treatment site, whereas sarcomas mainly arise in tissues exposed to high radiation doses, within or near the irradiation fields. The most frequent histological types following head and neck irradiation are squamous cell carcinomas, followed by soft tissue sarcomas [3].

Radiation-induced sarcomas of the head and neck are rare [5]. However, with an increasing use of radiotherapy and improved survival rates for head and neck cancer patients, the incidence of radiation-induced sarcomas has noticeably increased. According to Wei *et al*., the incidence of radiation-induced head and neck sarcomas increased from 0.06% to 0.17% between 1960 and 2010 [5].

The combination of radiotherapy with chemotherapy in the initial treatment may contribute to the development of secondary sarcomas, especially with the growing use of 5-Fluorouracil as a radiosensitizer [6, 7]. Antimetabolites may induce improper nucleotide incorporation or other DNA repair errors following sublethal radiation-induced damage, potentially promoting sarcoma development in vulnerable smooth muscle cells [7].

Post-radiation leiomyosarcoma is an extremely rare entity. Between 2000 and 2020, only 26 cases of post-radiation leiomyosarcoma of the head and neck have been reported [6], mainly in the sinonasal localisations, oral cavity (notably the tongue), and nasopharynx. Only three cases of radiation-induced laryngeal leiomyosarcoma have been reported in the literature [8–10].

Most laryngeal leiomyosarcomas arise in the supraglottic and glottic regions. The clinical presentation is similar to other laryngeal tumors and depends on the tumor's location. Laryngeal LMS rarely presents with lymph node metastases, and distant metastases are uncommon at diagnosis. Imaging with CT and MRI provides complementary information, defining the tumor's local extent, size, and nodal status. Diagnosis is confirmed by biopsy with histological and immunohistochemical analysis.

The management of radiation-induced sarcomas remains controversial, with various treatment modalities applied, including surgery, chemotherapy, re-irradiation, and combinations of these approaches. The overall prognosis of radiation-induced sarcomas is poor, with a 5-year overall survival rate ranging from 24.2% to 38.2% [6]. Surgery is the preferred treatment for laryngeal LMS. Clear surgical margins provide a better prognosis, and lymph node dissection is not systematically performed [9].

CONCLUSION

Post-radiation leiomyosarcoma appears years after radiotherapy. The association with chemotherapy in the initial treatment, as described in our case, may contribute to the development of secondary leiomyosarcoma.

As with all radiation-induced sarcomas, secondary leiomyosarcoma has a poor prognosis, with a 5-year survival rate below 50%.

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