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Larynx osteosarcoma: case report

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Summary

We present the case of a laryngeal osteosarcoma in a 69-year-old man, which generated many diagnostic and treatment difficulties. The patient was admitted to the hospital because of persisting hoarseness and a laryngeal tumor was seen on laryngoscopy. Total laryngectomy was performed and the pathological examination of the resected material showed osteosarcoma of the larynx. Postoperative radiotherapy was planned but the patient declined any treatment. He was readmitted to the hospital 5 months later with a neck tumor and enlarged lymph nodes on the right side of the neck. Chemotherapy started and during treatment metastatic lymph nodes on the left side of the neck and pulmonary metastases were detected.

Key words: larynx, osteosarcoma

Introduction

Malignant mesenchymal tumors of the larynx constitute 0.3-1% of all laryngeal cancers, and of these osteosarcoma is the rarest. Rarely, osteosarcoma of the larynx may follow radiation therapy for laryngeal squamous cell carcinoma, or the larynx may be the site of metastatic osteosarcoma [1].

Clinical diagnosis of osteosarcoma of the larynx may be difficult. To be able to reach the correct diagnosis, a high index of suspicion and due diligence in obtaining deep biopsy specimens are important [2,3].

Laryngeal osteosarcoma is a highly malignant neoplasm with early hematogenous spread. Survival rates are poor. Treatment is primarily surgical. Limited experience reveals that a combination of surgery and radiotherapy with adjuvant chemotherapy can offer palliation and sometimes cure in early-stage disease. Outcomes for this disease are generally poor, with most patients dying of the disease as a result of pulmonary metastasis [2,4].

Case presentation

A 69-year-old man was admitted to the hospital because of persisting hoarseness lasting for about 6 months. No other specific symptoms existed. Clinical examination showed no abnormal findings. Computed tomography (CT) of the thorax and abdomen were normal. Full blood count and serum biochemistry were within normal limits. Laryngoscopy showed a supraglottic mass on the right which was biopsied and the pathological diagnosis was sarcomatoid malignant tumor with fusiform cells. Total laryngectomy and right functional neck dissection was performed on May 2004. Pathological examination of the resected material showed osteoid formation by malignant infiltrating cells, while immunohistochemical examination showed high Ki67; CD68, vimentin and NSE with focal positivity; and negative pancytokeratin, EMA. The final diagnosis was osteosarcoma of the larynx (Figures 1,2). The patient was staged as T1N0M0.

Adjuvant postoperative radiotherapy was planned

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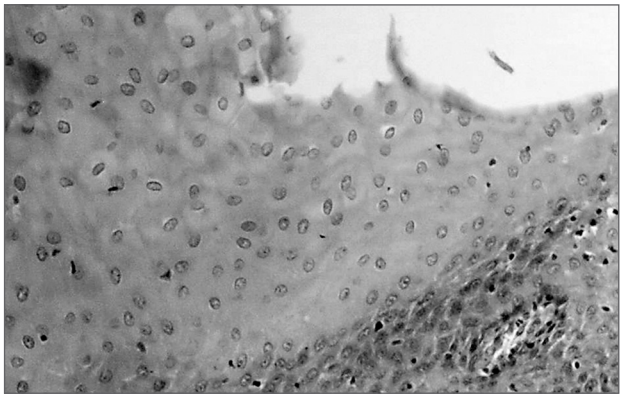

Figure 1. Malignant mesenchymal tumor showing storiform pattern under multiple layer squamous epithelium (H&E ×100).

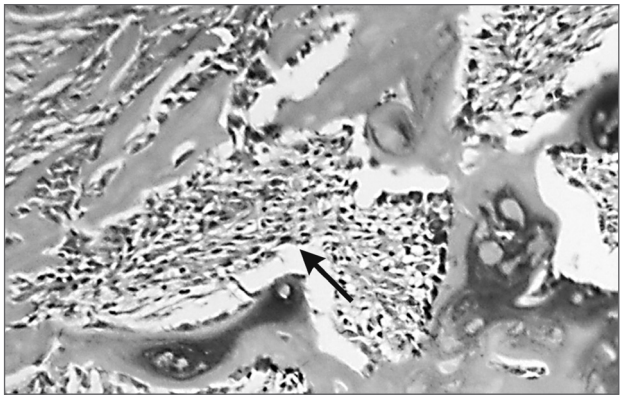

Figure 2. Osteoid regions in tumor infiltration areas (arrow, H&E ×100).

but the patient declined any treatment and was discharged.

He was readmitted to the hospital 5 months later with a neck tumor and enlarged lymph nodes on the right anterior cervical region measuring 2.5 cm in largest diameter (Figure 3). No systemic metastases were detected at that time and induction chemotherapy to be followed by radiotherapy were planned. Three cycles of cisplatin 75 mg/m^2, day 1 and doxorubicin 50 mg/m^2, day 1, every 3 weeks were administered to the patient.

On clinical and CT examination after the 3rd chemotherapy cycle the following findings were apparent: a pack of lymph nodes on the left side of the neck measuring 7 cm (Figure 4) invading the oropharynx and hypopharynx; an infiltrating ulcerated mass; and also lymphadenopathy at the right upper jugular region and the left submandibular region (4×2 cm). On chest CT scan millimeter-sized lung metastases were detected (Figure 5).

Palliative external beam radiotherapy with Co^{60},

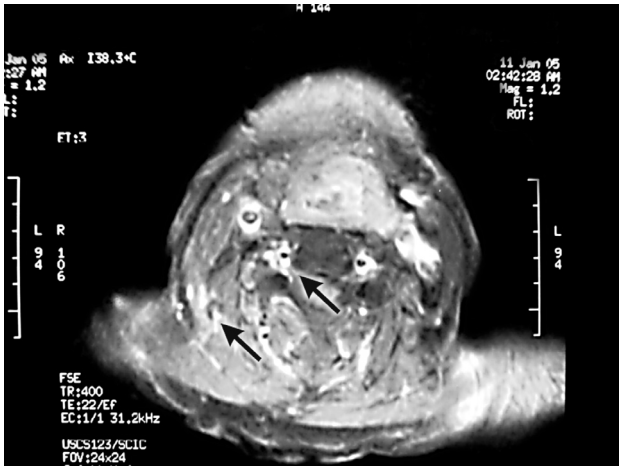

Figure 3. A 2.5 cm neck tumor on the right anterior cervical region and enlarged lymph nodes on the right side of the neck (arrows).

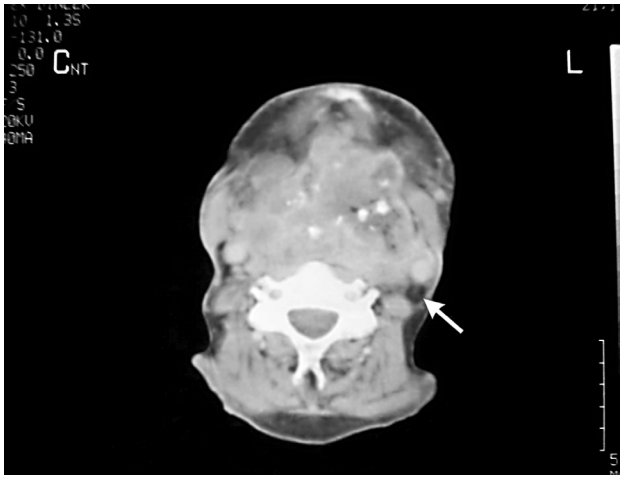

Figure 4. Metastatic lymph nodes measuring 7 cm on the left side of the neck invading the oropharynx and hypopharynx (arrow).

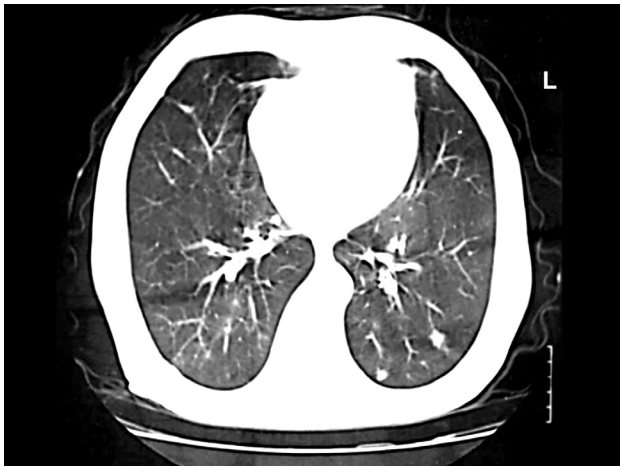

Figure 5. Bilateral millimeter-sized pulmonary metastases.

with 2 lateral fields (16 fractions/250 cGy/day, 40 Gy total dose) was delivered. Minimal disease regression and improvement in swallowing and local pain was achieved. The patient died with regional recurrence and multiple pulmonary metastases 12 months post-laryngectomy.

Discussion

Primary osteosarcoma of the larynx is a rare condition. What is known about this malignancy comes mainly from case reports. To the best of our knowledge only 15 such cases are reported in the literature till 2005 and most of them are metastatic to the larynx. In our case osteosarcoma originated from the larynx.

The osteosarcoma probably arose by dedifferentiation of a chondrosarcoma of the cricoid cartilage [5]. Rarely, laryngeal osteosarcoma may follow radiation therapy for squamous cell carcinoma in the head & neck region, or the larynx may be the site of metastatic osteosarcoma [1].

Histological diagnosis of osteosarcoma of the larynx may present difficulties [6,7]. In our case the first histological diagnosis was sarcomatoid malignant tumor of the larynx with fusiform cells. Initial full diagnostic work-up showed no systemic metastases and the patient was treated by total laryngectomy and functional neck dissection. Histological examination of the laryngectomy material put the final diagnosis of osteosarcoma of the larynx based on immunohistochemistry studies.

Treatment is primarily surgical. Limited experience reveals that a combination of surgery and radiotherapy with adjuvant chemotherapy can offer only palliation [8-12]. The overall prognosis of laryngeal osteosarcoma is poor but may be improved with early recognition and adequate surgical intervention [6,13,14].

Outcomes for this disease are generally poor, with most patients dying of the disease as a result of pulmonary metastasis [2,4,15]. Although our patient underwent radical surgical therapy the disease recurred regionally within 5 months after the operation. Combination chemotherapy with cisplatin and doxorubicin was not able to improve regional disease or to prevent the development of pulmonary metastases. Radiotherapy had minimal impact on the regional disease and the patient died within 12 months from laryngectomy.

Conclusion

Larynx osteosarcoma is a rare disease. Pathological confirmation of osteoid is required for diagnosis. The treatment of choice is aggressive surgical intervention directed at complete tumor removal. Experience with radiotherapy and chemotherapy is limited. Prognosis is poor and most patients die as a result of pulmonary metastasis.

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