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Carcinoma ex Pleomorphic Adenoma of the Parotid Gland with Metastasis to the Lung: Case Report and Review of the Literature

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Abstract

Background: Carcinoma ex pleomorphic adenoma) CA-ex-PA) is an uncommon tumor of the salivary glands that may present with metastasis. This article aimed to evaluate the response to treatment of a patient with CA-ex-PA and lung metastasis to combination chemotherapy.

Case presentation: A 21-year-old girl presented to the clinic of ENT with right-sided neck swelling, which started four months ago. The initial examination revealed three to four metastatic lesions of the lung, and the FNA raised the possibility of pleomorphic adenoma. Consequently, the pathology report of the samples collected during total parotidectomy and lymphadenectomy stated a CA-ex-PA with the lymph node invasion. The patient underwent combination chemotherapy. After the third and sixth sessions, a computerized-tomography scan of the lungs was obtained from the patient, which displayed improved lung lesions.

Conclusions: Eleven months after the end of treatment, there are no signs of recurrence in the patient. Due to the high cost of targeted therapy in the treatment of cancer in areas with low socioeconomic status, chemotherapy can still be effective.

Introduction

Background: When we talk about salivary gland tumors, we mostly indicate tumors originating from the larger glands, including the parotid, submandibular, and sublingual glands. Pleomorphic adenoma is a common type of salivary gland tumor, which usually affects the large salivary glands. This tumor shows a complex behavior and can transform into a malignant tumor with the ability to metastasize [1]. The three known clinicopathological manifestations of pleomorphic adenoma include carcinosarcoma, CA-ex-PA with metastatic potential, and high-grade salivary duct carcinoma. CA-ex-PA consists of a pleomorphic adenoma and a malignant epithelial component. CA-ex-PA is a reasonable cause for 4% of all salivary gland tumors and about 12% of salivary malignancies. The most common sites for CA-ex-PA include parotid glands 67%, small glands 18%, submandibular glands 15%, and sublingual glands 1%. Patients are usually in their 60s or 70s, and CA-ex-PA is rare in individuals younger than 30 [2]. The patient presents with a chronic mass and a recent rapid growth. Furthermore, the transformation into a malignant tumor with the ability to develop metastasis may occur in pleomorphic adenomas in 6% of cases. Although the metastasis onset is really rare, the median survival of patients with distant metastasis has been reported 4.3 to 7.3 months. The most common sites of metastasis are the

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lung and bone. Surgery [3] is considered the primary treatment; however, chemotherapy and radiotherapy [4], and sometimes target therapy have been used in some cases. In this article, we presented a case of CA-ex-PA with lung metastasis. Besides, we conducted a literature review on the etiology, behavior, optimal treatment, and prognosis of patients with metastasis.

Case presentation

A 21-year-old girl presented with right-sided neck swelling at the submandibular region starting four months ago. The mass had recently increased in size. We found swelling at the parotid and preauricular areas with upward extension to the sternocleidomastoid muscle during neck inspection. The mass had irregular edges, a firm consistency, and was fixed to the underlying tissue. Ultrasound revealed a hypoechoic oval with lobulated margin and central echogenicity (30x16 mm) in the right parotid parenchyma. Cervical and periauricular lymphadenopathy (10x9 mm and 14x2 mm, respectively) was also present. The biochemical profile was insignificant except for a slight increase in erythrocyte sediment rate (ESR). Lung CT scan showed three to four solid and round lesions in the right lung parenchyma with a maximum size of 27 x 19 mm suggestive of metastatic lesions. The initial FNA raised the possibility of pleomorphic adenoma. The patient underwent total parotidectomy and lymphadenectomy. Also, the pathologist reported a high-grade CA-ex-PA with invasion. One dissected lymph node of the parotid region, four of the nine dissected lymph nodes, and the resected tissue's deep margins were involved.

Based on the clinicopathological findings, the patient was diagnosed with CA-ex-PA with lymph node and lung metastasis. The patient was treated with combination chemotherapy as follows: Six course of 5-Fluorouracil 750 mg/m^2/day over 21 hours, Cisplatin 50 mg/m^2 infusion over 1.5 hours on the third day, one-hour infusion of Docetaxel 70 mg/m^2 on the second day, and Epirubicin 40 mg/m^2 within ten minutes on the fourth day. Drugs were administered once every 21 days. A lung CT scan was obtained from the patient after the third and sixth sessions and compared with the initial CT scan. After three treatment course, the lung CT scan revealed three 3, 12, and 8 mm nodules in the right upper lobe. A 12-mm cavitary nodule in the right lower lobe was also present. Comparison of this CT scan with the initial one showed that two out of four lung nodules were shrunk, and the remaining two nodules were up to 16 mm in size, which had a significant reduction of more than 50% in size. At the end of 6 courses, only one ground glass lesion with a maximum of 8 mm was seen on the CT scan. A PET scan was requested to rule out the possibility of remaining malignancy, which was negative. The patient is followed every three months, and the last CT scan performed three months after the PET scan showed no specific lesion. The patient is still alive ten months after the treatment, and there is no sign of disease recurrence.

Discussion

A significant percentage of CA-ex-PA tumors occur in untreated pleomorphic adenomas with malignant transformation. The epithelial component of the pleomorphic adenoma is the part that goes through a malignant transformation. CA-ex-PA is capable of distant and regional metastasis in malignant tumors. Pathologists have proposed a subclassification for CA-ex-PA, which divides them into non-invasive, minimally invasive, and invasive. The tumor is considered non-invasive if it remains inside the capsule and has an excellent prognosis with overall 5-year survival of 100%. If a small extracapsular invasion [5] is present (less than 1.5 mm), it is a minimally invasive tumor with an excellent prognosis and 5-year survival of 98%. Invasive tumors have extracapsular invasions greater than 1.5 mm and have a poor prognosis with a 5-year survival of about 25 to 65%. The degree of invasion and the histological subtype of the malignant component affect the type of treatment and prognosis for CA-ex-PA. Other important prognostic factors for patients' survival are the clinical stage, tumor size, recurrent disease, distant metastasis, malignant subtype, high-grade tumor, deep tissue invasion, perineural invasion, and lymph node metastasis [6]. Therapeutic modalities for malignant tumors include exclusive surgery performed at early stages, surgery in combination with radiotherapy for high-grade tumors, or removal of deposits at the surgical site. Moreover, postoperative radiotherapy is recommended for patients with specific histological risk factors such as positive microscopic margins and perineural invasion [7,8]. Radiotherapy alone is used in patients who cannot tolerate surgery or have non-resectable tumors. Information on the systemic management of CA-ex-PA is limited. Several studies have suggested using Trastuzumab in cases of HER2/neu positive tumors; however, its effect has not been proven in clinical trials [9]. Antiangiogenesis drugs and tyrosine kinase inhibitors are also suggested in treating CA-ex-PA [10], but their effectiveness remains controversial, and the drug was even discontinued in some trials due to toxicity. Another treatment used for X carcinoma is chemotherapy. There is still no general agreement on the role of chemotherapy, especially in advanced cases. Few studies have shown the effects of chemotherapy drugs such as Vinorelbine, Paclitaxel, Cyclophosphamide, Doxorubicin, and Cisplatin as a single or combination therapy [11,12-14]. Another study on the chemotherapy effects by [15], was performed on 22 patients with parotid CA-ex-PA; 5-year survival of 50% was reported, compared to 60% in those with no treatment. Nevertheless, in a Canadian group study, the Cyclophosphamide, Doxorubicin, and Cisplatin regimen had good responses [16].

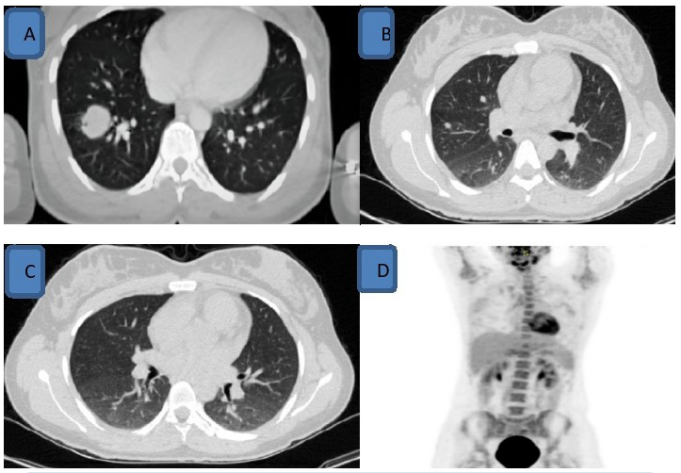

Figure 1: Axial computed tomography Scan images before combination **(A)**. After three cycles of combination chemotherapy, a singnificant reduction of more than 50% in size **(B)**. After six cycles of combination chemotherapy, the lung lesions disappeared **(C)**. A PET scan was negative fot remaining malignancy **(D)**.

Conclusion

In our study, the use of combination chemotherapy had an excellent response for our patient. The patient is currently being followed for eleven months after the treatment, and no recurrence has occurred. In countries with low economic conditions where the possibility of using targeted therapeutic drugs is low, chemotherapy can be used with significant effect.

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