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Sialoblastoma: A Rare Cause of Pleural Metastasis

Sialoblastoma: Plevral Metastazın Nadir Bir Sebebi

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ABSTRACT

Sialoblastoma is rare low-grade malignant tumor, which is originated from parotid or submandibular gland. Although this tumor can be successfully, treated surgically, distant metastases to different anatomical regions may be seen depending on the histopathological features of the tumor. In this article, we present a 11-year-old patient who underwent an operation due to ectopic sialoblastoma and had pleural metastasis during the follow-up. To the best of our knowledge, this is the first case report of sialoblastoma-related pleural metastasis.

Keywords: Metastasis, Pleura, Salivary glands, Sialoblastoma

ÖZET

Sialoblastoma, parotis veya submandibuler bezden köken alan, nadir görülen düşük gradeli malign bir tümördür. Bu tümör cerrahi olarak başarılı bir şekilde tedavi edilebilse de, tümörün histopatolojik özelliklerine bağlı olarak farklı anatomik bölgelere uzak metastazlar görülebilir. Bu yazıda ektopik sialoblastoma nedeniyle ameliyat edilen ve takiplerinde plevral metastaz gelişen onbir yaşında bir hasta sunuldu. Bildiğimiz kadarıyla bu, sialoblastoma ilişkili plevral metastaz ile ilgili ilk vaka sunumudur.

Anahtar Kelimeler: Metastaz, Plevra, Sialoblastoma, Tükrük bezleri

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INTRODUCTION

Salivary gland tumors are rare in children.[1] Sialoblastoma, an epithelial-originated salivary gland tumor, occurs three times more in submandibular glands than in parotid glands. It is often congenital or occurs shortly after birth. Sialoblastoma is accepted as an aggressive and low-grade malignant tumor due to its potential of recurrence, lymph node metastasis and distant metastasis. It is exceedingly rare that sialoblastoma is originated from ectopic salivary glands. Only 2 cases of ectopic sialoblastoma have been reported in literature.[1,2]

CASE REPORT

A male patient, who is now 11-year old, was admitted to the otorhinolaryngology clinic when he was 4-year old because of the swelling on his right anterior cheek. The complaint was considered to result from hemangioma. The ultrasonography (US) showed a 25x15 mm in size hypoechoic solid lesion with lobulated borders and right cervical lymph nodes with a transverse diameter of 10 mm. Magnetic resonance imaging (MRI) examination showed a 27x14 mm in size mass, isointense on T1-weighted images and slightly hyperintense on T2-weighted images to muscle, located along the anterior borders of masseter muscle The lesion revealed homogeneous enhancement after IV contrast media injection (Figure 1). Surgical resection was performed on the patient. The mass was first thought to be a hemangioma, so lymph node dissection was not performed. Histopathological specimens, obtained after the first surgical resection, revealed 3-4 mitosis, central necrotic areas and vessel invasion. The borders of resection were positive in a few small focuses. There was powerful staining with S100 protein and high-molecular-weight cytokeratin (CK). Vimentin was stained in stroma and in the periphery of solid areas. With p63 protein, focal reactivity was detected. The Ki-67 score was 80%. The patient was histopathologically diagnosed with sialoblastoma. In the first 7 months following the first surgical operation, there were two recurrences in the same location. Surgical resection was performed again. Metastatic nodules were detected in the lung parenchyma during the second recurrence (Figure 2). Radiotherapy (RT) was not performed due to the age of the patient. Instead, chemotherapy, VAC regimen includes the drugs vincristine sulfate, dactinomycin (actinomycin-D), and cyclophosphamide, was preferred. Lung parenchymal metastatic nodules disappeared. After the clinical recovery that lasted about 7 years, the patient was taken to the pediatric emergency service with complaints of dyspnea. The lung CT scan revealed nodular focal and diffuse pleural thickening with invasion to the adjacent tissues in the left hemi-thorax and also pleural effusion with compressive atelectasis (Figure 3). Tru-cut biopsy was performed from areas showing diffuse pleural thickening. Histopathological examination showed solid nests composed of basaloid cells with round to oval nuclei and relatively fine chromatin pattern. Findings were consistent with sialoblastoma (Figure 4). Chemotherapy, including ifosfamide, carboplatin, and etoposide phosphate, was given to the patient. The patient died of respiratory failure after 8th cycles of chemotherapy.

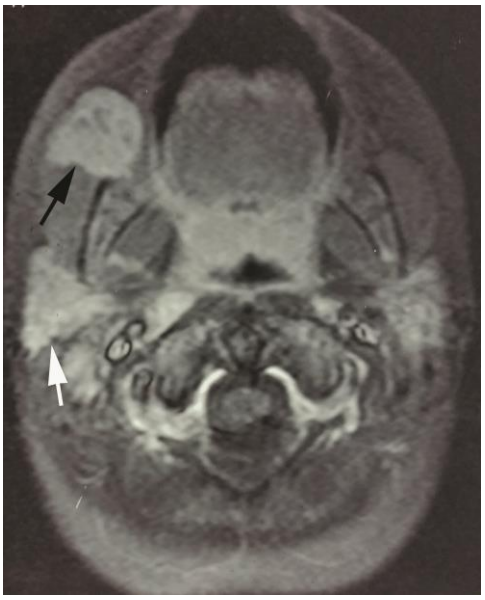

Figure 1. Axial postcontrast T1-weighted MR image shows homogeneously enhancing solid mass on right anterior cheek (black arrow). The lesion is outside the borders of parotid gland (white arrow).

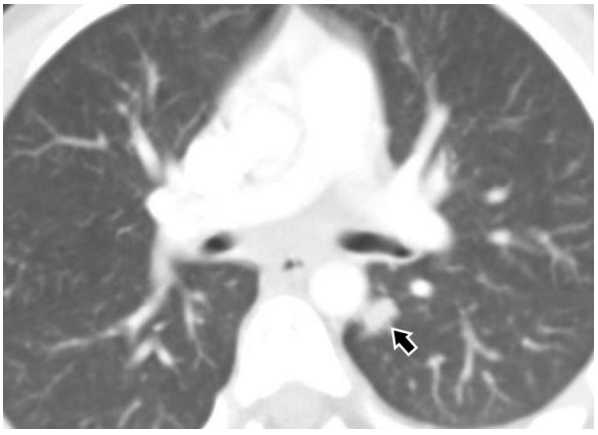

Figure 2. Axial CT shows metastatic nodule in left lung.

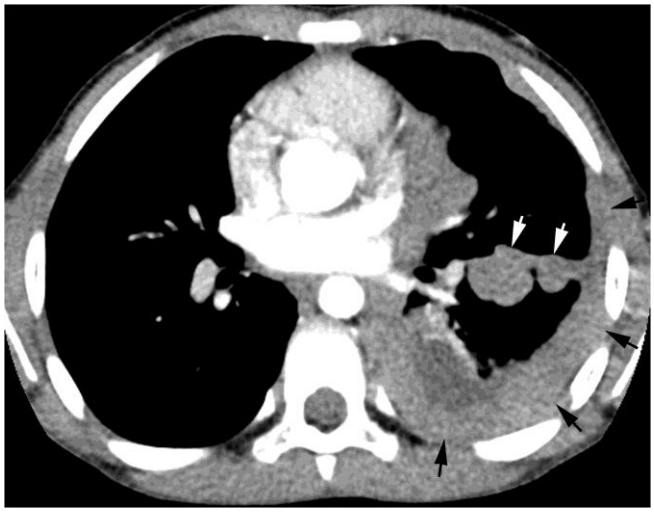

Figure 3. Axial postcontrast CT image reveals nodular (white arrows) and diffuse (black arrows) pleural thickening consistent with metastasis in the left hemi-thorax.

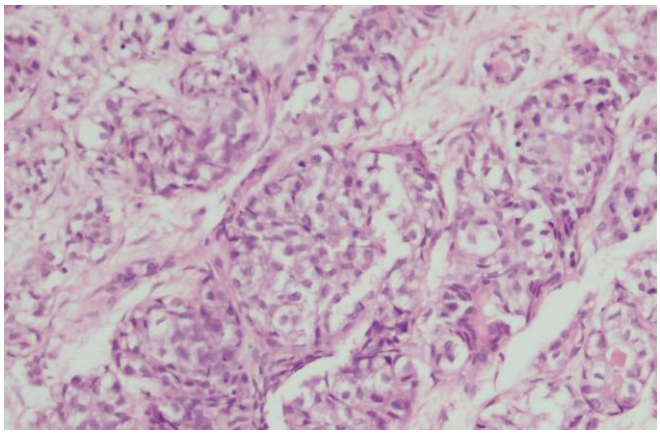

Figure 4. Microscopic features (hematoxylin-eosin x200): A neoplasm is form round and oval basaloid type epithelial cells.

DISCUSSION

OT is encountered at a rate of 1 to 5 per hundred Sialoblastoma is regarded as malignant due to its local recurrences, lymph node metastasis and distant metastasis. But some authors have classified it as favorable and unfavorable. Though histological variations can be seen in favorable sialoblastoma, solid-multinodular areas consisting of cytologically benign round or oval basaloid tumor cells divided by fine fibrosis stroma are more common.[4]

Nuclear type is not distinct. Mitosis, necrosis and apoptosis can be detected. In the follow-up of the surgical excision of favorable sialoblastoma cases, local recurrence or distant metastases have not been reported. In immunohistochemical studies, Ki-67 score which is a proliferation marker, have been reported to be between 1-20% in favorable cases and focal staining with p63 has been detected.[4]

In sialoblastoma, anaplasia may also increase in relation to the increased proliferative capacity. This can pave the way for local recurrences and even distant metastases. In our patient, local recurrence was detected twice in 7 months after the operation. During the second recurrence, metastatic nodules were found in both lungs. In unfavorable sialoblastoma, there are minimal fibro-myxoid stroma and anaplastic basaloid tumor cells. The aggressive prognostic factors are infiltrative surgical border, neuro-vascular invasion, central necrosis, and the presence of cytological anaplasia. In all cases of sialoblastoma-related lymph node metastasis, bone metastasis and lung parenchymal metastasis, there were neuro-vascular invasion and central necrosis. The Ki-67 score ranges from 40 to 80% in unfavorable cases and diffuse staining takes place with p63.[4-6] In our case, there were central necrosis areas and vascular invasion. In addition, positivity with a rate of 80% was observed with Ki-67. Sialoblastomas are seen as lobule-contoured, well-demarcated, and hypoechoic solid lesions on US. They are often seen well-demarcated solid lesions that isodense to muscle on computed tomography. Gross calcification has not been reported. The MRI signal quality changes depending on whether the tumor has necrotic and hemorrhagic area but sialoblastoma is often isointense to muscle on T1[1]. The feature of mild hyperintense signal on T2 is associated with high nucleus/cytoplasm ratio. The hemorrhagic areas on T1 have hyperintense signal quality. The cystic-necrotic areas are distinctly hyperintense on T2. The tumor reveals often poor, and homogenous or heterogeneous enhancement with contrast media.[3]

Sialoblastoma-related distant metastases have been reported in a limited number of patients.[4-8] Lung metastases generally have the feature of unilateral or bilateral solid round parenchymal nodules in upper lobes. It can be solitary or multiple. In one case, there was vertebral metastasis in the feature of lytic round lesion that could cause vertebral fracture[5]. In our case, during the second local recurrence, parenchymal and sub-pleural metastatic nodules smaller than 1 cm were detected in lungs. All metastases were seen in patients with parotid sialoblastoma.

Most of the children with sialoblastoma can be cured with surgical resection.[9] RT is not recommended for children but chemotherapy can be applied on cases which are relapsed and lymph nodes or distant metastasis are detected. Some authors have reported that

chemotherapy gives promising results in patients with local recurrence or distant metastasis.[5,6,9]

When positive surgical borders, high Ki-67 score, central necrosis and neuro-vascular invasion are evaluated in the light of literature, it is considered that all of them play an important role in the development of distant metastasis. Even though lymph nodes, lung parenchymal metastasis and bone metastasis have been reported in sialoblastoma to date, case of pleural metastasis has not been reported. Our case is the first one that has been presented sialoblastoma-related pleural metastasis.

CONCLUSION

In summary, sialoblastoma has a potential of metastasis. Depending on the histopathological features of tumor, patients should be closely monitored.

Authorship contribution statement

Consept and desing: İE, İS
Acquisition of data: İE, İS
Analysis and interpretation of data: İE, İS
Drafting of the manuscript: İE, İS
Critical revision of the manuscript for important intellectual content: İE, İS

Declaration of competing interest

None of the authors have potential conflicts of interest to be disclosed.

Availability of data and materials

All data generated or analyzed during this study are included in this published article.

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