



Osteosarcoma metastatic to the thyroid gland: A rare case report

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ABSTRACT

Sarcoma is rarely seen in the thyroid gland and it is difficult to differentiate it from anaplastic thyroid carcinoma. Extrasosseus osteosarcoma, a rare malignant nonepithelial neoplasia, can be seen in the thyroid gland, breast, soft tissue, and intraabdominal organs. A 31-year-old woman with a previous history of osteosarcoma of distal femur was presented with a constantly growing mass in her neck accompanied with progressive shortness of breath and worsening dysphagia. On physical examination, a 4 x 5 cm firm mass was palpated in the left lobe of her thyroid gland. Seven years ago, during her pregnancy, she was diagnosed with osteosarcoma of the distal femur and she received chemotherapy and radiotherapy after medical abortion. A year after her initial diagnosis, she underwent surgery for osteosarcoma. Two years later, pulmonary metastasis was detected incidentally in her control computed tomography. She was operated for pulmonary metastasis since there was no remission after chemotherapy. When she presented to the clinic with a neck mass, her thyroid function tests were within the normal range. On computed tomography scan, a 2.5 cm calcified lesion in left lobe of her thyroid gland was detected. Fine-needle aspiration of the thyroid mass displayed numerous discohesive pleomorphic tumors cells identical to osteosarcoma cells seen on her previous knee biopsy specimen. On positron emission tomography, a 3 x 3 x 4.5 cm hypermetabolic lesion with pathologic 18F-FDG uptake on the left side of her neck in thyroid cartilage level was detected. A bilateral total thyroidectomy was performed. The patient was discharged without any complications.

Keywords: Osteosarcoma metastasis, thyroid gland, pathology, fine-needle aspiration, cytology

INTRODUCTION

Osteosarcoma is a high-grade malignant tumor arising from bone and producing osteoid (1). Primary osteosarcoma is most commonly encountered in the distal femur, proximal tibia, and proximal humerus (1). It is the most common primary skeletal malignancy in children and young adults (2).

In autopsy series, the incidence of metastasis to the thyroid gland ranges from 1.2% to 24% (3). But in the clinic, osteosarcoma metastasis is not very frequent. In both clinical and autopsy case series, renal cell carcinoma, breast, lung cancer are the most frequent primary tumor sites metastasizing to the thyroid gland. Although the thyroid gland is highly vascular, metastasis to this gland is infrequent (4). The rarity of metastasis to the thyroid gland is speculated to be due to high oxygen saturation and iodine content of thyroid gland and its fast blood flow (5).

CASE REPORT

A 31-year-old woman with a history of osteosarcoma of distal femur was presented with a constantly growing mass in her neck accompanied with progressive shortness of breath and worsening dysphagia in September 2019. In physical examination, a 4 x 5 cm firm mass was palpated in her left lobe of the thyroid gland.

In 2012 during her pregnancy, she was diagnosed with osteosarcoma around her right knee and had five sessions of chemotherapy and a month-long radiotherapy after medical abortion. A year after her initial diagnosis, she underwent surgery for osteosarcoma and in 2014 she had two prosthesis operations for her distal femur and proximal tibia. In 2015, pulmonary metastasis was detected incidentally on a routine computed tomography and she received chemotherapy. Failing to resolve, she underwent surgical treatment for pulmonary metastasis.

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Figure 1. Computed tomography image of thyroid mass.

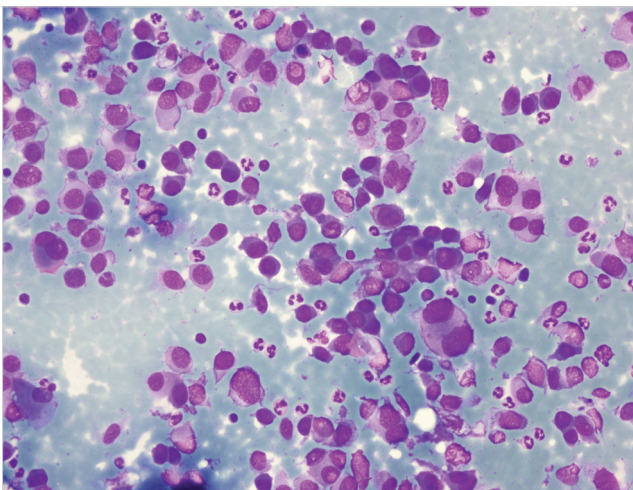


Figure 2. Fine needle aspiration biopsy hypercellular aspirates showing numerous discohesive tumor cells with intranuclear pseudoinclusions (x400, May Grünwald Giemsa).

When she presented to the clinic with a neck mass, her thyroid function tests were within the normal range. On computed tomography, a 2.5 cm calcified lesion in the left lobe of her thyroid gland was detected (Figure 1). The patient underwent ultrasound-guided fine-needle aspiration (FNA), which showed



Figure 3. Macroscopic examination, the tumor is gray-lobulated cut surface.

predominantly discohesive atypical pleomorphic tumor cells. In light of our patient’s previous history, the FNA findings were most consistent with metastasis of osteosarcoma. (Figure 2). On positron emission tomography, a 3 x 3 x 4.5 cm hypermetabolic lesion with pathologic 18F-FDG uptake on the left side of her neck in thyroid cartilage level was detected ($SUV_{max} = 12.7$). Sutureless bilateral total thyroidectomy was performed with intraoperative neuromonitoring and the patient was discharged without any complication.

Serial sections through the left lobe of the thyroid showed gray lobulated firm mass surface with areas of necrosis and calcification. Chondroid appearance in peripheral zones was seen (Figure 3). Histopathological examination of the lesion revealed thyroid parenchyma with interstitial infiltrates of high-grade malignant neoplasm composed of polygonal cells and hyperchromatic nucleus presenting high mitotic index and necrotic areas and osteoid formation and confirmed osteosarcoma metastasis to the thyroid gland (Figure 4a, 4b).

Informed consent was taken from the patient.

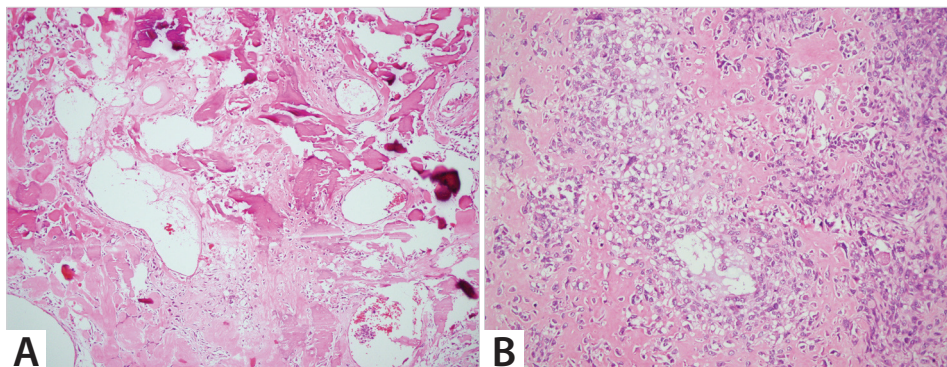


Figure 4. A. Microscopic examination: tumor with osteoid formation (x100, H/E). **B.** Microscopic examination: High grade malignant neoplasm composed of polygonal cells of intermediate size and hyperchromatic nucleus (x200, H/E).

DISCUSSION

Mesenchymal cells producing immature osteoid are the characteristic histopathologic feature of osteosarcoma. Similarly, pathologic examination of our specimen revealed interstitial infiltrates of high-grade malignant neoplasm and osteoid formation (2).

Most patients with metastasis to the thyroid gland remains asymptomatic unlike our case presenting with a growing mass accompanied by respiratory symptoms (4). In a study on 22 patients diagnosed with metastasis to the thyroid gland, 14 patients presented with palpable thyroid nodules similar to our case (6).

In a review article, 372 cases reported between 2000 and 2010 were analyzed (7). The mean age of the patients was found to be 59 years and the female to male ratio was found to be 1.4. The most frequently reported cancer metastatic to the thyroid gland was renal cell carcinoma followed by colorectal, lung, breast cancer. Only 4% of the reviewed cases were sarcomas. The interval between the initial diagnosis and metastasis was the longest with 75 months in sarcomas. For our case, this interval was almost seven years. In the same article, whether preexisting thyroid conditions such as primary thyroid neoplasm or benign thyroid conditions induce metastasis led to a controversy. Our case did not have any preexisting thyroid diseases. Also, most cases (87.6%) were euthyroid at the time of diagnosis of thyroid metastasis as our case (7).

Thyroid metastasis may be the initial evidence of diagnosis or recurrence of the primary tumor. The diagnosis was made through clinical complaints. Until proven otherwise in a patient with a history of a primary tumor, a thyroid mass should be treated as a metastatic lesion. In the review of Chung et al., it was shown that 24% of FNAs were incorrect (7). In another study, positive and negative predictive values of the fine-needle aspiration in metastasis to the thyroid gland were 89% and 93% respectively (8). A primary tumor and metastatic malignancy can be differentiated by immunohistochemical markers. Immunostaining of thyroid tumors is important in patients with a known malignancy and with appropriate immunohistochemical markers correct diagnosis can be made.

Without a history of osseus osteosarcoma, primary thyroid osteosarcoma may be kept in mind (9). It presents with an expanding lesion similar to our case. So it is clinically impossible to distinguish a primary thyroid cancer from metastatic neoplasm. FNA is a powerful tool in the diagnosis of metastatic malignancies of the thyroid. Kim et al. found patients with metastatic cancer to the thyroid diagnosed by FNA in a retrospective research (6).

In metastatic diseases, it is crucial to decide when to operate. Unnecessary surgery in patients with poor prognosis should be avoided (10). Total thyroidectomy or thyroid lobectomy is a mainstay of local treatment for metastasis of osteosarcoma (11). In a study, total thyroidectomy is favored since with lobectomy, there can be positive margins but total thyroidectomy seems to decrease recurrence (8). Total thyroidectomy both rules out the disease and treats the metastatic disease.

Survival after thyroidectomy depends on the primary tumor. Management of a metastatic patient depends on the primary tumor site, the extent of disease, metastasis to other sites, symptomatic patients. The extent of the disease and the stage of the primary tumor have a greater impact on survival (7).

Osteosarcoma metastasizes first to the lungs and it is the most common site of metastasis with more than 80% of initial metastases (11). Distant metastasis can occur in 25-30% of patients who were treated (11). The site and timing of the metastasis are crucial. While metastatic disease at presentation has a bad prognosis, patients with metastatic disease at least two years after the initial treatment have a better prognosis (1). Patients with longer disease-free survival have a better prognosis (1). Also, resectable metastatic osteosarcomas have better prognosis (1). In our case, the patient was diagnosed at a younger age, she had a disease-free period of two years till her first metastasis to lungs and four years of disease-free survival till her second metastasis to the thyroid gland. Both of the metastatic sites were resectable. Due to the site and timing of her disease, her prognosis seems to be better.

Resectable disease and the extent of the disease have an impact on survival. Treatment options should be selected carefully for each case considering metastasis site, tumor resectability, and duration between the initial diagnosis and metastasis (12).

CONCLUSION

In a patient with a prior history of malignancy presenting with a neck mass, a possible metastasis from the primary tumor site should be kept in mind.

Informed Consent: Informed consent was obtained from patient who participated in this case.

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OLGU SUNUMU-ÖZET

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Tiroid bezine osteosarkom metastazı: Nadir bir olgu

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ÖZET

Sarkom, tiroid bezinde nadir görülür ve anaplastik tiroid kanserinden ayırıcı tanısı zordur. Nadir epitelyal olmayan, malign neoplazi olan kemik dışı osteosarkom, tiroid bezinde, memede, yumuşak dokuda ve batin içi organlarda görülebilir. Daha önce distal femurda osteosarkom öyküsü olan 31 yaşındaki kadın hasta, nefes darlığı ve disfajiye neden olan boyunda büyüyen kitle ile başvurdu. Fizik muayenesinde tiroid bezinin sol lobunda 4 x 5 cm sert kütle palpe edildi. Hasta, yedi yıl önce gebelik döneminde distal femur osteosarkomu teşhisi almış, medikal abort sonrası kemoterapi ve radyoteapi almıştır. İlk tanısından bir yıl sonra, osteosarkom nedeniyle ameliyat edilmiş ve bundan iki yıl sonra çekilen kontrol tomografide insidental olarak akciğer metastazı tanısı almıştır. Kemoterapi sonrası remisyon gözlenmeyince akciğer metastazı nedeniyle ameliyat edilmiştir. Kliniğe boyunda kitleyle başvurduğunda tiroid fonksiyon testleri normaldi. Bilgisayarlı tomografide tiroid bezinin sol lobunda 2,5 cm kalsifiye lezyon izlenmiştir. Lezyonun ince iğne aspirasyonunda daha önceki diz biyopsisindeki osteosarkom hücrelerine benzeyen, çok sayıda diskoheviz pleomorfik hücreler saptanmıştır. Pozitron emisyon tomografisinde tiroid bezi sol lobunda 18F-FDG tutulumu olan, 3 x 3 x 4,5 cm hipermetabolik lezyon izlenmiştir. Yapılan bilateral total tiroidektomiye takiben hasta komplikasyonsuz olarak taburcu edilmiştir.

Anahtar Kelimeler: Osteosarkom metastazı, tiroid bezi, patoloji, ince iğne aspirasyon, sitoloji**DOI:** 10.47717/turkjsurg.2022.4860