

Short communication

Metastasis of submandibular gland carcinoma to the toe bone: a case report

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Accepted 22 March 2019

Available online 8 April 2019

Abstract

Adenoid cystic carcinoma (ACC) is rare. One of its characteristics is a high rate of distant metastasis, and the most common site is the lungs, followed by the brain and liver. Bony metastases account for up to 1.96% of cases only, however, and metastasis from the blood to the bones of distal limbs is rare. We report a 62-year-old woman who presented with pain caused by bony metastases as the first symptom eight years after being diagnosed with submandibular ACC. Although such cases are rare, they indicate that when patients who have had cancer visit the hospital with other symptoms, clinicians should make sure to rule out metastasis.

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Keywords: submandibular adenoid carcinoma; pathological fracture; bone metastasis; toe; diagnosis

Case report

A 62-year-old woman had had a mass under the right submandibular gland for one and a half years with no symptoms before it was resected in August 2009. Histopathological examination showed that it was an adenoid cystic carcinoma (ACC) that had invaded the nerves and surrounding glands, and three weeks later, she had a course of radiotherapy. After 20 months, computed tomographic (CT) images showed metastases in the lungs, but she felt no discomfort, and emission CT showed no obvious bony metastases. The lesions were confirmed by histopathological

examination and were accordingly treated with radiotherapy.

Eight years after operation, she experienced pain, swelling, and ulceration in her left toe with no obvious cause (Fig. 1). The pain was not relieved by two weeks of rest, so she had treatment in our hospital. Radiographs of the feet confirmed partial destruction of the distal phalanx of the left big toe by a mass, which indicated the possibility of metastasis. Emission CT showed a mild concentration of radioactive shadowing in the maxilla, but there was no evidence of metastasis in the remaining bone (Fig. 2).

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Fig. 1. Ulceration of the left big toe.

She was followed up with CT to review the metastases in the lungs. We amputated the left big toe, and histopathological examination indicated a diagnosis of ACC (Fig. 3), and she had no symptoms during the next 12 months of follow up.

Discussion

ACC is a rare lesion that accounts for less than 1% of head and neck malignancies, and 10% of all tumours of the salivary glands.^{1,2} The survival time after bony metastasis is short, usually about 10 months.² One of the characteristics of such lesions is a high rate of distant metastases, which can be as much as 40% in the long term. They metastasise most commonly to the lungs, and then the brain and liver. Bony metastases, however, account for up to 1.96% of cases only, and metastasis from the blood to the bones of distal limbs is rare.^{3,4}

To the best of our knowledge, only two cases of metastasis to bones in the feet from submandibular adenoid cystic carcinomas have been reported.^{5,6} A 52-year-old man presented with a 2.5 cm mass in the right submandibular gland, which was resected in 2003.⁵ Pathological examination indicated that it was an ACC, which had metastasised to the big toe of his right foot 12 years later. In the other case, the patient had extensive metastases seven years after the diagnosis of submandibular ACC, and died 10 months later.⁶ A review of relevant publications indicated that metastases to the feet are rare, but we noted that in both these patients they had developed after blood transfusions. Our patient first experienced pain in her foot caused by pathological fractures, which was also the case in the previous two patients, and we think that the metastasis also developed as a result of a blood transfusion.

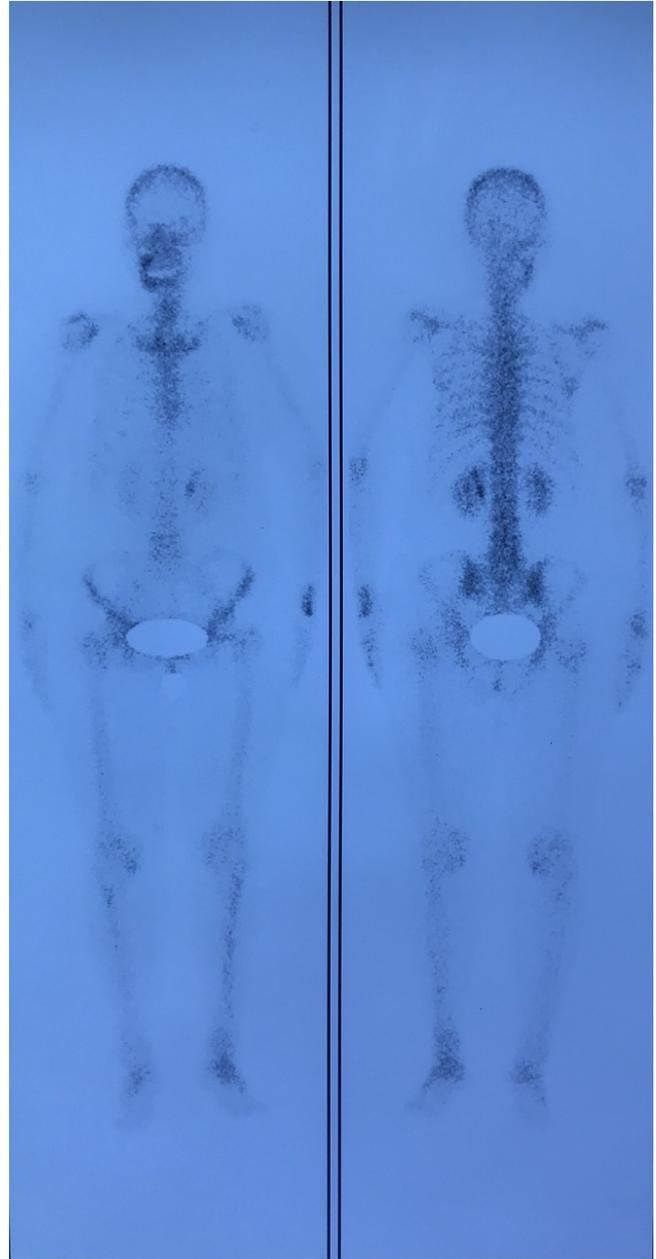


Fig. 2. Emission computed tomogram showing no evidence of bony metastasis in the feet.

The radiograph of the patient's feet had indicated the destruction of bone, but the emission CT did not confirm the presence of a lesion, which had led to a false negative result. We think, therefore, that the value of using bone emission CT alone to locate distant metastases is questionable. During diagnosis, it is better to use a combination of medical history and accurate examinations, such as CT, magnetic resonance imaging, and emission CT.

In this case, based on the pain, ulceration, and relative imaging of the toe, the symptoms were both difficult to diagnose and easy to misdiagnose. Although such cases are rare, they indicate that clinicians should pay particular attention to any history of cancer when there is no obvious cause

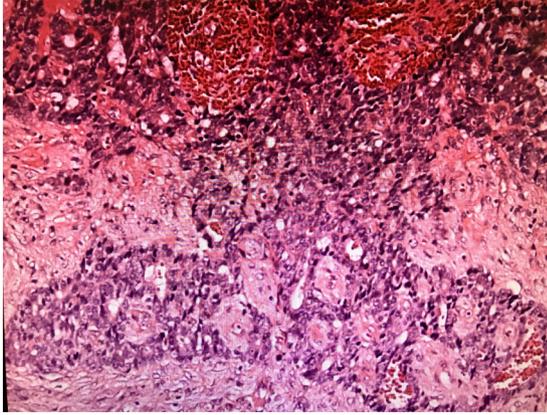


Fig. 3. Staining of the adenoid cystic carcinoma in the left big toe (haematoxylin and eosin, original magnification x 200).

for the patient's symptoms. For this reason, we think that it is necessary for them to be diagnosed and treated by a multidisciplinary team.

Conflict of interest

We have no conflicts of interest.

Ethics statement/confirmation of patient's permission

We have obtained ethics approval. We have obtained the patient's permission. This study complied with the Declaration of Helsinki and was approved by the Human Ethics and Research Ethics Committees of the Fourth Hospital of Hebei Medical University.

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