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Occult Papillary Carcinoma Thyroid with Solitary Appendicular Bone Metastasis in Proximal Humerus – A Case Report

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Learning Point of the Article:

Papillary thyroid carcinoma can present clinically with metastasis as the initial manifestation, including rare occurrences in the appendicular skeleton. Comprehensive whole-body evaluation is essential when assessing tumorous bone lesions in the elderly to differentiate primary bone tumors from metastatic disease.

Abstract

Introduction: An occult presentation of primary thyroid malignancy refers to a clinically silent primary tumor that initially manifests through metastasis or secondary paraneoplastic phenomena. Papillary carcinoma is a well-recognized thyroid malignancy associated with this pattern of presentation, but bone metastases are uncommon in occult papillary thyroid carcinoma (OPTC), with reported cases typically involving the axial skeleton.

Case Report: A woman in her 70s presented with swelling and pain in her right arm. Examination revealed an oval swelling on the proximal humerus. Radiographs and magnetic resonance images suggested a lytic lesion in the proximal humerus. Upon metastatic workup, biopsy, and immunohistochemistry, the diagnosis turned out to be metastasis from occult papillary thyroid cancer.

Conclusion: Bone metastases are uncommon in OPTC, with reported cases typically involving the axial skeleton. Metastasis to the appendicular skeleton as the initial presentation is exceptionally rare.

Keywords: Occult papillary cancer; bone metastasis; malignancy; geriatrics.

Introduction

The bone ranks as the third most common site for metastasis in various solid tumors, encompassing lung, breast, prostate, colorectal, thyroid, gynecologic, and melanoma. Unfortunately, once cancer spreads to the bone, a cure is rarely achieved, leading to a spectrum of morbidities such as pain, an increased risk of fractures, and hypercalcemia [1, 2]. An occult presentation of primary thyroid malignancy is defined as an unknown primary malignancy that is symptomless, which first manifests itself as metastasis or secondary paraneoplastic phenomena, and papillary carcinoma is known for this kind of presentation [3]. Bone metastases are infrequently observed in cases of occult papillary thyroid carcinoma (OPTC), with metastases to the appendicular skeleton being exceedingly rare.

Case Report

A woman in her 70s presented to our tertiary specialist unit with swelling and pain in her right proximal arm for the past 5 months. The onset of swelling was insidious and had rapidly progressed in

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Author's Photo Gallery


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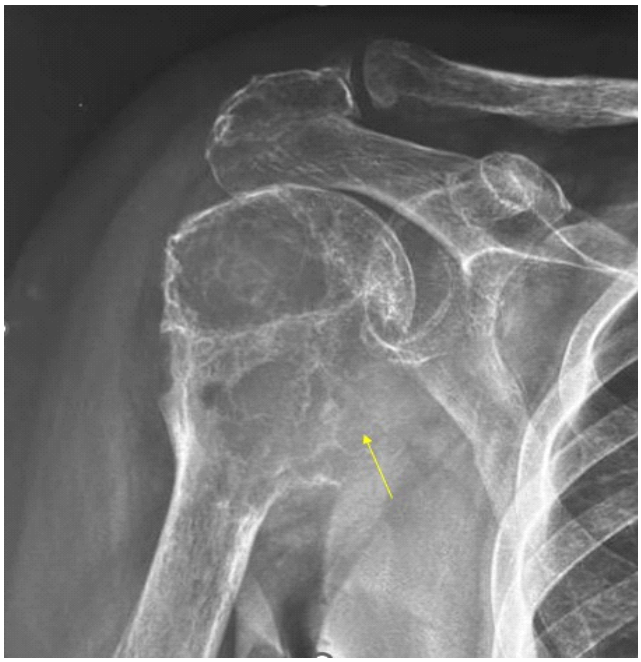

Figure 1: Plain radiograph – anteroposterior projection shows an expansile, lytic lesion with a moth-eaten appearance involving the proximal part of the humerus with the cortical breach and soft-tissue component (marked with yellow arrow).

size, accompanied by a history of weight loss and loss of appetite. The pain, which was also insidious in onset, had a dull, aching nature and gradually progressed, but its intensity rapidly increased over the past month. There were no associated histories of trauma or fever.

Upon examination of the local site, an ovular swelling over the proximal humerus, measuring 8 × 5 × 5 cm, was observed. The swelling was firm in consistency, with irregular surfaces, and was tender upon palpation. The overlying skin was free from the swelling, but it was fixed to the bone. Shoulder movements were restricted, although distal neurovascular structures remained intact.

Plain radiographs revealed an expansile, lytic lesion involving the proximal part of the humerus, displaying a moth-eaten appearance, endosteal scalloping, and periosteal reaction. The lesion exhibited a narrow zone of transition, cortical breaches at multiple locations, and the presence of a soft-tissue component (Fig. 1).

Contrast-enhanced computed tomography (CT) scan showed a lytic mass with a soft-tissue component in the right proximal humerus involving the head and upper shaft with cortical destruction and extension to the adjacent soft-tissue planes (Fig. 2).

Magnetic resonance imaging of the proximal humerus showed a lesion involving the proximal humerus with a cortical breach and soft-tissue component, which is hyperintense in T2 images and mild-to-intermediate in T1 images (Fig. 3).

A metastatic workup was conducted, including contrast-enhanced CT scans of the chest and abdomen. The scan revealed a heterogeneously enhancing lesion measuring 4.5 × 3.3 cm in the right lobe of the thyroid, along with a small nodule in the left lobe of the thyroid.

The fluorodeoxyglucose (FDG) positron emission tomography scan revealed a small metabolically active lesion in the right lobe of the thyroid gland, indicating a primary malignant site. In addition, an FDG avid lytic expansile lesion with a soft-tissue component was observed in the right proximal humerus (Fig. 4).

Fine needle aspiration cytology of the thyroid gland revealed papillary carcinoma of the thyroid.

An incisional biopsy of the proximal humerus was performed to confirm the diagnosis, revealing tumor cells predominantly

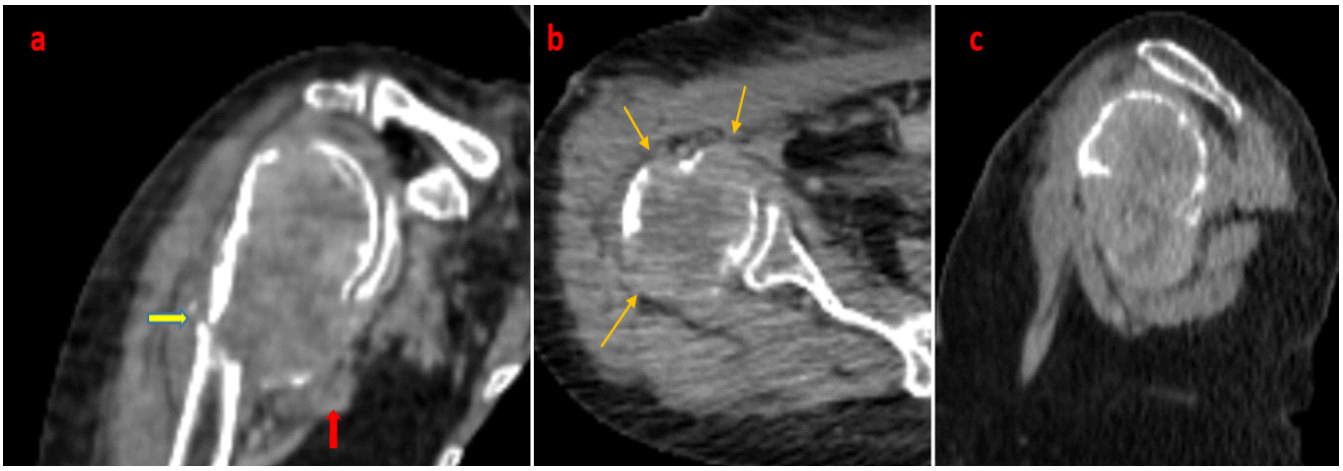

Figure 2: Computed tomography images show a lytic mass with cortical destruction extending into adjacent soft-tissue planes with a pathological fracture at the head–neck junction. (a) Coronal image (cortical breach marked with red arrow and soft-tissue extension marked by red arrow), (b) Axial image (lesion marked with yellow arrow), (c) Sagittal image.

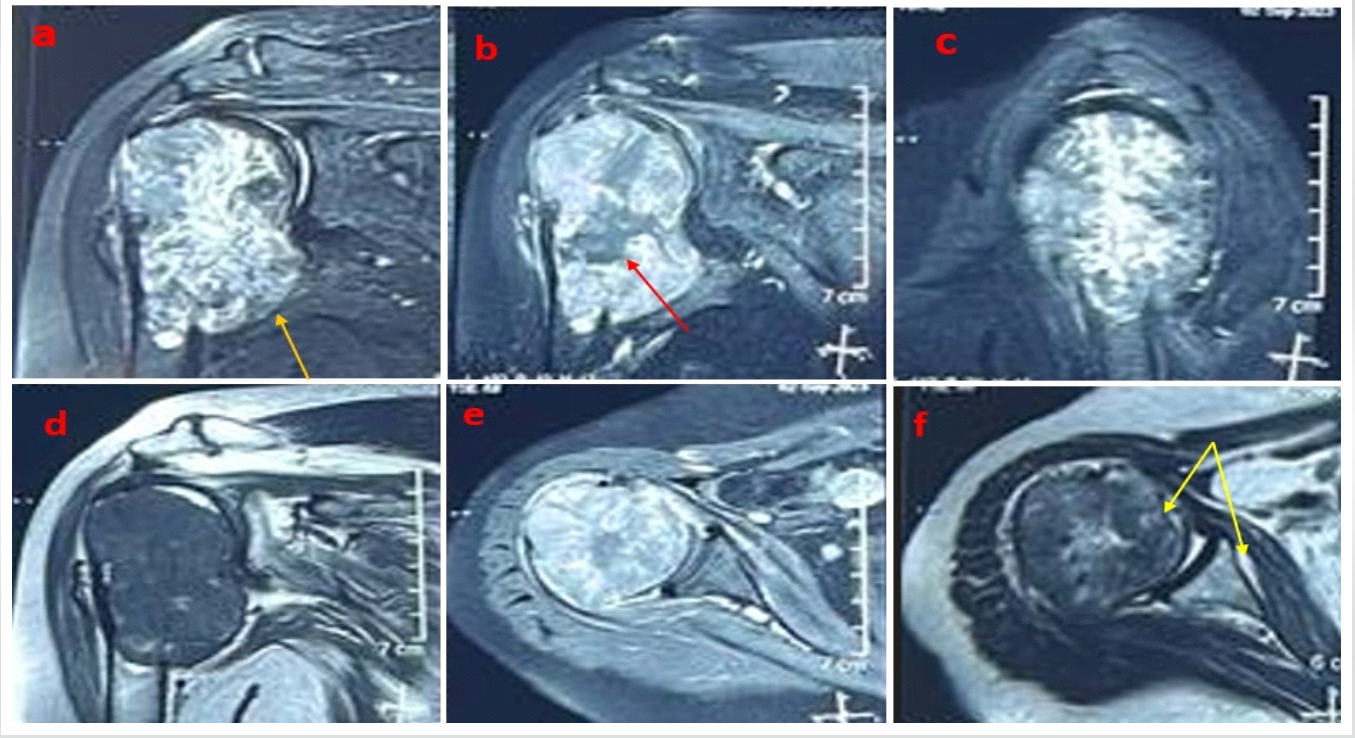

Figure 3: Magnetic resonant images show hyperintense lesions in T2 images and mild-moderate intensity lesions in T1 images. (a and b) Coronal T1 Images, (c) Sagittal T2 image, (d) Coronal T1 image, (e and f) Axial T2 and T1, respectively.

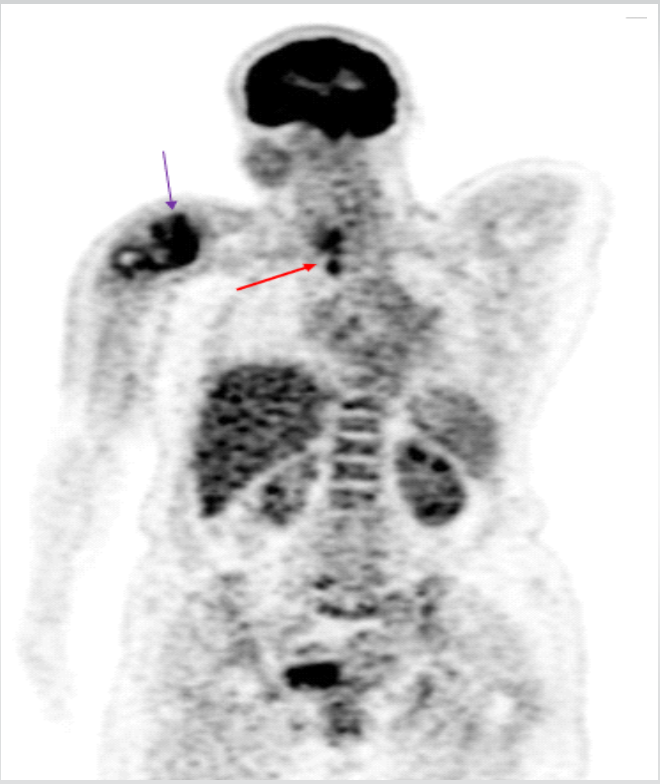

Figure 4: Positron emission tomography scan image shows a metabolically active lesion in the right lobe of the thyroid (red arrow) and a fluorodeoxyglucose avid lytic expansile lesion with a soft-tissue component in the right proximal humerus (blue arrow).

arranged in a back-to-back complex papillary pattern with some follicles. Tumor cells displayed pleomorphism, round to oval nuclei, fine chromatin, inconspicuous nucleoli, and a mild-to-moderate amount of cytoplasm. The tumor cells exhibited positive for PAX8, CK7, TTF-1, focal positivity for thyroglobulin, and negativity for GATA3, WT-1, and Vimentin. The morphological features, along with immunohistochemistry, confirmed the diagnosis of metastatic papillary thyroid carcinoma (PTC) (Fig. 5).

Following the histopathological confirmation of the diagnosis, the multidisciplinary team at our hospital's Tumor Board, comprising experts from the Departments of Orthopaedics, Pathology, and Oncology, collaborated to determine the treatment strategy. Considering the stage of the disease, the patient's age, and multiple comorbidities, including hypertension and dilated cardiomyopathy, decided to start palliative radiation therapy.

During 1 year follow-up, the patient symptomatically improved with the union at the proximal humerus pathological fracture and improved quality of life.

Discussion

Bony metastasis is common in the elderly age group, and we cannot conclusively identify any tumor as the primary source without confirmation through histopathological examination

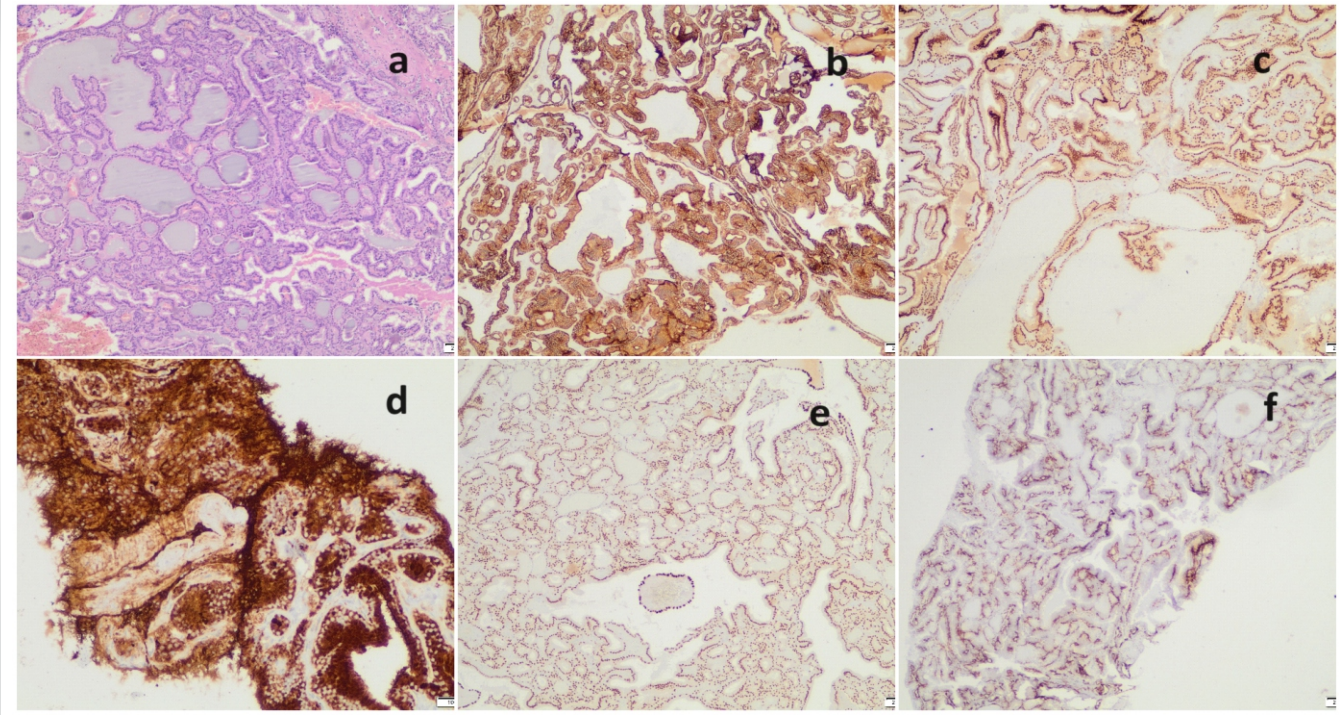

Figure 5: (a) Hematoxylin and eosin, ×40: Tumor comprising of thyroid follicles and papillary configuration, (b) Tumor cells are immune-reactive for CK7, (c) Tumor cells are immune reactive for PAX8, (d) Tumor cells are immune reactive for thyroglobulin. (e) Tumor cells are immune reactive for TTF, (f) Vimentin expression is noted in fibrovascular cores of the papillary region.

and whole-body evaluation for metastasis or a primary lesion [4,5]. This case represents a rare occurrence: A lytic lesion in the proximal humerus, noted over a period of 5 months, was found to be the metastasis from an occult papillary thyroid malignancy.

The McGraw-Hill Concise Dictionary of Modern Medicine (2002) defines "occult primary malignancy" as an unknown primary malignancy that is asymptomatic and initially reveals itself through metastases or secondary paraneoplastic phenomena [3,6]. While bony metastasis occurs in approximately 2–13% of individuals with thyroid malignancy, it is exceedingly rare in occult papillary cancers [7-9]. Only a handful of reported cases exist where OPTC has manifested as skeletal metastasis, with these instances primarily involving axial bones [10-13]. Jouhar et al. 2014 reported a case of OPTC with metastasis to the skull and sacrum, whereas Ikejiri et al. reported a case of metastasis to the vertebrae and pelvis in 1997 [6,14]. To date, there have been no reported cases of appendicular skeletal metastasis from OPTC, making this case unique. In this instance, the papillary carcinoma followed a silent asymptomatic course, with its initial presentation occurring in the appendicular bone, specifically the proximal humerus. All other reported cases of occult thyroid cancer presenting as bone metastasis are of the follicular variant [2,3,13-16].

Histopathological examination is crucial for distinguishing primary lesions from metastasis. The fundamental morphological characteristics of classical PTC encompass the presence of papillae and nuclear alterations. While immunohistochemistry generally offers limited diagnostic utility for PTC, it has a role in cases of metastatic disease. Neoplastic cells typically display strong and diffuse immunoreactivity to markers including keratin, CK7, thyroglobulin, TTF1, and PAX8, while the outcomes of other markers such as HBME-1, Galectin-3, S100 protein, CITED1, and CK19 can be variable. Standard treatments for bony metastases from papillary thyroid cancer include radioactive iodine treatment, surgical removal of bone metastases, beam radiation, arterial embolization, and chemotherapy [13,15,17-20].

This case underscores the diagnostic challenges posed by secondary lesions from papillary carcinoma of the thyroid mimicking primary lytic lesions in the proximal humerus. The case emphasizes the need for a comprehensive diagnostic approach involving histopathological examination and imaging modalities to accurately differentiate between primary and metastatic lesions.

Conclusion

Bone metastases are uncommon in OPTC, with reported cases typically involving the axial skeleton. Metastasis to the appendicular skeleton as the initial presentation is exceptionally rare. Our case highlights this exceptional presentation, emphasizing the importance of a thorough whole-body evaluation when investigating tumorous bone lesions in the elderly to accurately distinguish between primary bone tumors and metastatic disease.

Clinical Message

Papillary thyroid carcinoma can initially manifest with metastasis, including rare cases in the appendicular skeleton, necessitating a comprehensive whole-body evaluation in elderly patients to distinguish primary bone tumours from metastatic disease.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

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