Giant Cell Tumor with Multi-Focal Pulmonary, Scapular and Rib Metastases in A 65-Year Old Hypertensive Woman

Anas Ismail1, Kabiru Isyaku1, Adekunle Y Abdulkadir2

ABSTRACT

Giant cell tumors is a primary benign bone tumors but with a tendency for significant local bone destruction, recurrence, and occasionally, distant metastasis. Most metastases are to the lungs but rarely metastases to the regional lymph nodes, the scalp, and the pelvis have been reported. Giant cell tumor in the elderly are very rare; and to the best of our knowledge, there is no prior report on concomitant pulmonary, scapular and rib metastases from giant cell tumor in the elderly in the literature at the moment. Hence, a case in a 65-year-old woman is presented.

Key words: Giant cell tumor, metastasis, scapular, rib, old woman

INTRODUCTION

Giant cell tumors (GCTs) constitute about 5% of all bone tumors and about 21.5% of all primary benign bone tumors; with slight female predominance (female-to-male ratio of 1.40:1.28) (1-4). Majority of cases present between 20-50 years, with peak incidence in the third decade of life (2). Giant cell tumors are observed predominantly at the ends of long bones, most commonly located in and around the knee, elbow and wrist (1-3). Pain is the most common presenting symptom. Swelling and deformity are associated with larger lesions.2 GCTs are benign, but with a tendency for significant bone destruction, local recurrence, and occasionally, distant metastasis (2). The frequency of metastases is between 2 to 3%. Most metastases are to the lungs but rarely metastases to the regional lymph nodes, the scalp, and the pelvis have been reported (2,3). GCTs in the elderly are very rare; and to the best of the knowledge of this author, there is no prior report on concomitant pulmonary, scapular and rib metastases from GCT in the elderly in the literature at the moment. Hence, a case in a 65-year-old woman is presented.

CASE

A sixty-five-year-old woman was referred to the medical outpatient clinic of Aminu Kano teaching hospital on account of two-month history of cough, difficulty with breathing and swelling on the right shoulder region. There was no history of orthopnea, paroxysmal nocturnal dyspnea or fever. She had a bone curettage and grafting of giant cell tumor of the proximal right tibia five years prior to presentation, which was incidentally diagnosed following one year history of knee pain and deformity. She presented with a two-month history of cough, difficulty with breathing and swelling of her right shoulder region. On examination, there was a soft tissue swelling on the right shoulder region with some deformity of the right shoulder. There was no history of orthopnea, paroxysmal nocturnal dyspnea or fever. She was a known case of hypertension for 10 years and was on regular medication. She had no history of smoking or alcohol intake. She had no history of family members with similar complaints.

Correspondence: Anas Ismail, Department Of Radiology, Aminu Kano Teaching Hospital, Pmb, 3452, Kano, Nigeria. Tel: +2348039275786 E-mail: ibnmaliknas@yahoo.co.uk

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pain, swelling and pathologic fracture. The only available frontal pre-operative right knee radiograph (Figure 1) showed the primary GCT as a sub-articular, eccentric, expansile radiolucent lesion that shows a narrow zone of transition and classic loculated soap-bubble appearance without sclerotic margin. A focal cortical discontinuity was noted but there is no periosteal reaction or soft tissue extension. There was no genito-urinary, nervous system or gastrointestinal symptoms. She was diagnosed hypertensive twenty years prior to presentation with blood pressure being controlled with nifedipine and alpha-methyl dopa tablets. She is not a known diabetic.

Physical examination revealed an elderly woman, who is not pale, anicteric, acyanotic with no obvious respiratory distress. She had no peripheral lymphadenopathy. The respiratory rate was 15 cycles per minute. The chest was resonant to percussion and auscultation revealed vesicular breath sounds bilaterally with no added sound. The pulse rate was 81 beats per minute, regular with full volume. Blood pressure was 150/90 mmHg. The apex beat was located at the fifth left intercostal space and not heaving. The heart sounds were first and second with loud A2 only. The abdominal and nervous system

Figure 1. Pre-operative frontal right knee radiograph, showing a subarticular, eccentric, expansile radiolucent lesion with ‘soap bubble’ appearance.

Figure 2. Scanogram of the chest CT scan, showing a fairly roundish right hilar opacity of soft tissue density. Similar but smaller lesion is also noted overlying the left costo-phrenic angle.

Figure 3. Axial contrast enhanced CT scan at the level of lung apices shows an osteolytic destructive heterogenous mass lesion involving the right scapular spine with faint contrast enhancement.

Figure 4. A coned view of coronal reformatted chest CT scan (mediastinal window), showing lytic destruction of the spine and acromium of the right scapula by a slightly hyperdense fairly roundish mass lesion with patchy contrast enhancement.
examinations were essentially within normal limits. Musculo-skeletal review was essentially normal; except for the mildly tender hard immobile swelling over the right scapular region. Healed surgical scar was noted over the right tibia, corresponding to the site of the previous curettage and grafting. Frontal chest radiograph (Figure 2) shows a right hilar roundish irregular opacity of soft tissue density. Another similar but smaller and oval-shaped mass lesion is also noted overlying the left costo-phrenic angle. The remaining lung fields, heart and remaining outlined bones appear normal; except for the noted aortic unfolding. The scanogram of the high resolution chest computed tomography (CT) scan (Figure 3) confirmed the above chest findings. The axial and reformatted coronal images show a lytic destruction of the spine and acromion of the right scapula by a slightly hyperdense fairly roundish mass lesion (measuring 50 x 39 mm in size) that demonstrates heterogeneous patchy contrast enhancement (Figure 3 and 4). There are multiple fairly roundish isodense masses in the posterior aspect of the right hilum and left costo-phrenic angle, measuring 45 x 38 mm and 43 x 24 mm in their respective sizes. They both show patchy contrast enhancement. The latter also shows adjacent rib destruction (Figure 5 and 6). The remaining lung fields, the heart and remaining mediastinal structures are intact. The remaining bony thorax and overlying soft tissues are within normal limits. Abdominal ultrasound scan, complete blood count, liver and renal function tests as well as the serum levels of calcium and phosphate show no abnormality.

Assessment of multiple chest wall and pulmonary masses with systemic hypertension was made. Four weeks later, she had a wide local excision biopsy of the scapular and left costal lesions, which were confirmed to be metastatic deposits of giant cell tumor origin at histology. She was maintained on her antihypertensives and subsequently referred to cardio-thoracic surgeon at the neighboring teaching hospital, where she was booked for intrapulmonary tumor excision via thoracotomy. Thereafter, she was lost to follow up.

**DISCUSSION**

Giant cell tumor is a primary bony lesion of variable severity and unclear histogenesis (5). The peak age incidence of GCT is the third decade of life, with slight female preponderance (2,6) The presented case is also female but was seen in the seventh decade. About 50% of GCTs are located around the knee; being more common in distal femur, the proximal tibia (shown in the presented case). Other sites include: the proximal humerus and distal radius, appearing as subarticular, eccentric, expansile radiolucent lesion with classic loculated soap-bubble appearance without sclerotic margin, periosteal reaction or soft tissue extension (2,4,6). However, it can affect the flat bones and the small bones of the hands and feet (2).

An important differential of GCT is osteoclastoma of hyperparathyroidism, (7) which in this case, was excluded by normal renal function test, serum levels of calcium, phosphate and alkaline phosphatase. The biological mechanism of metastasis of benign giant-cell tumor is uncertain, though; microvascular trauma resulting in
Giant cell tumor embolization at the time of curettage was implicated in some cases. Other biological factors, postulated in these mechanisms include immune surveillance and the intrinsic biological characteristics of the GCT (3). However, it is difficult to determine the process of multifocal metastasis from GCT in the presented patient due to limited facilities. GCT metastases occur about 3-5 years after diagnosis of the primary lesion and sometimes, may not be detected for 10 years or longer; (2) as seen in this case, with features of metastasis five years after removal of primary tumor. This implies that patients with confirmed GCT must be followed up as long as possible to detect possible metastases early. Most GCT metastases show preponderance to the lung (1), as partly seen in the presented case. Further, Eyesan et al. documented a case with lung, hepatic and splenic metastases of GCT. Other possible sites include scalp and pelvis (3,9). A review of literature showed no concomitant pulmonary, scapular and costal metastasis of GCT in the elderly as seen in the patient; except for the report of Alacacioglu et al. (10) on concomitant pulmonary and breast metastases have been reported for the first time in Turkey.

The natural history of these metastases is unpredictable. Pulmonary metastases may spontaneously regress, remain stable, continuously grow slowly, or rapidly progress have been reported (1). However, the presented case was not followed long enough to actually monitor these outcomes. Treatment of metastatic GCT has been mainly surgical. Metastasectomy may result in long-term survival (1,3). As offered to the presented patient. Steroid chemotherapy has been used but with variable results but it was found to facilitate resection by reducing the size of the tumor (3).

A case of sixty-five-year old patient with scapular, pulmonary and chest wall metastases (giant cell tumor of the right tibial primary) was presented. The CT diagnosis was confirmed by histology on the surgically resected chest wall lesions. She was referred to cardiothoracic surgeon for pulmonary metastasectomy. The relevant literature was also reviewed.

REFERENCES