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Giant Cell Tumor of the Scapula Associated with Secondary Aneurysmal Bone Cyst

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Giant cell tumors are distinctive neoplasms characterized by a profusion of multinucleate giant cells scattered throughout a stroma of mononuclear cells. Most giant cell tumors are found at the epiphyses of long bones, especially around the knee joint. Flat bone involvement is rare.

However, a case of giant cell tumor with secondary aneurysmal bone cyst was encountered at the scapula of a 25-year-old man.

Since the occurrence of a giant cell tumor with secondary aneurysmal bone cyst on flat bones (i.e., the scapula) is extremely rare, the above-mentioned case is worthy of reporting.

Key Words : *Giant cell tumor, Secondary aneurysmal bone cyst, Scapula*

INTRODUCTION

Giant cell tumor, a relatively common and locally aggressive lesion, is composed of connective tissue, stromal cells, and giant cells which vary in amount and appearance. In 1940, Jaffe et al. (Jaffe et al., 1940) began a systematic analysis of those lesions that contained giant cells, emphasizing additional histological features, defining the specific characteristics of a giant cell tumor, and separating this neoplasm from other giant cell-containing tumors and tumorous conditions.

Giant cell tumors predominate in the long tubular bones (75 to 90 per cent of all cases), especially in the femur (approximately 30 per cent of all cases) and tibia (25 per cent of all cases) (Dahlin et al., 1970 ; Goldenberg et al., 1970 ; McGrath, 1972 ; Campanacci et al., 1975 ; Sung et al., 1982). The distal portions of the femur and radius and the proximal portion of the tibia are the most characteristic sites of involvement (approximately 50 per cent of all cases). The bones around the knee are affected in 50 to 65 per cent of all giant cell tumors (Dahlin and Unni, 1986 ; Resnick and Niwayama, 1988 ; Mirra et al., 1989).

The spine is occasionally involved. In the vertebral column, it is the sacrum that is the most typical site of localization ; vertebral involvement above the sacrum is uncommon and involves, in order of decreasing frequency, the thoracic, cervical, and lumbar vertebrae (Dahlin, 1977 ; Savini et al., 1983 ; Shirakuni et al., 1985).

In rare instances, giant cell tumors occur in other locations such as the ribs, the patella, the clavicle, and the sternum (Kelikian and Clayton, 1957 ; Sundaram et al., 1982). In 1974, Tuli et al. (1974) first reported scapular involvement with giant cell tumor.

This report describes the occurrence of a giant cell tumor associated with aneurysmal bone cyst of the scapula. The purpose of this report is to mark the rarity of the skeletal location of this giant

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cell tumor, especially a giant cell tumor associated with secondary aneurysmal bone cyst, as found in this male subject.

This is the first documented case in English world literature of a giant cell tumor associated with aneurysmal bone cyst of the scapula.

CASE HISTORY

The subject, a 25-year-old man, was admitted to the Department of Orthopedic Surgery for pain in the left shoulder. He had suffered intermittent pain in the left shoulder for two years and had recently noticed a growing, bulging mass. Physical examination revealed a diffuse, firm, and non-tender swelling in the left scapular region measuring about 10×15 centimeters, which extended from the axillary fold to the clavicle anteriorly and at an inferior angle to the spine of the scapula posteriorly. The surface was smooth, and the skin over it was warm and stretched but not adherent. Venous engorgement was present. Movement of the shoulder was limited by a mechanical block.

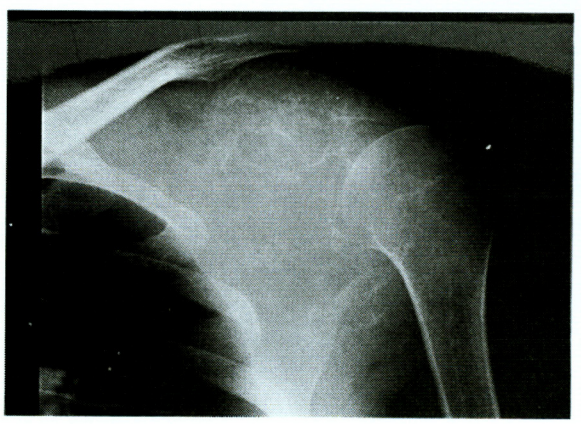

Fig. 1. Simple shoulder view showed left scapula with extensive ballooning of the bone with partly erosion of the cortex. Bony septa were crossing the lesion.

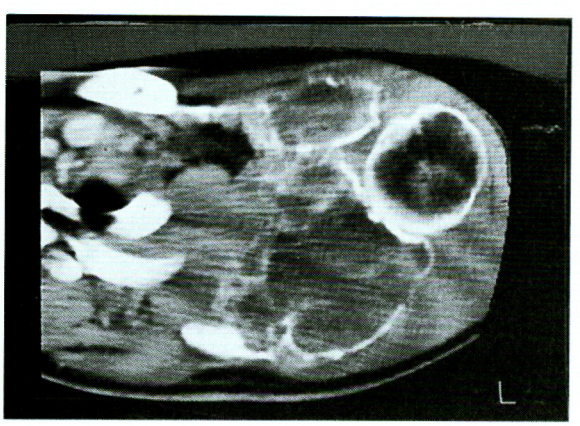

Fig. 2. Computed tomography showed an expansile mass with multiple internal septations. There were fluid levels within the multiple cysts.

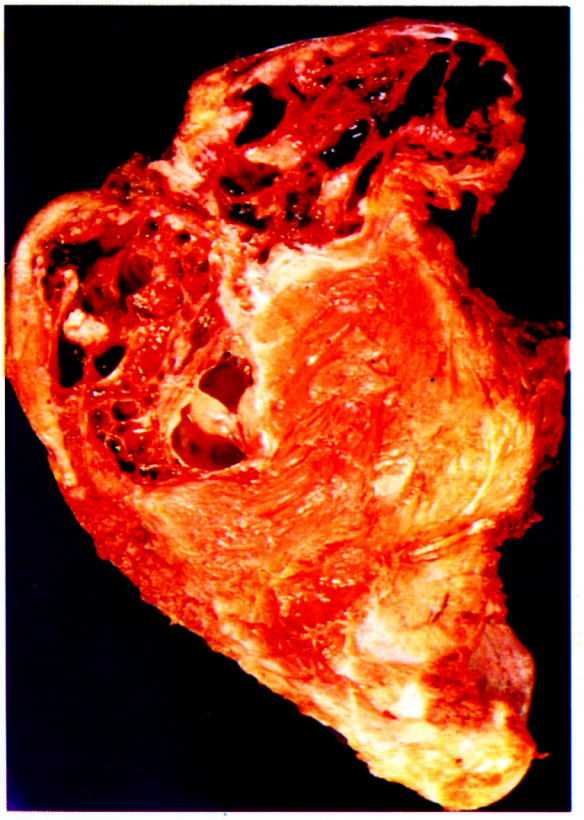

Fig. 3. The cut surface of the scapula showed multiple large cystic cavities with a few fragments of friable red tissue on their walls.

Liver function tests were unremarkable. Serum calcium and phosphorus were within normal limits. Serum parathormone level was within reference range.

A simple shoulder view and computed tomography were taken. The left scapula showed extensive ballooning of the bone, and at some points the cortex seemed to be eroded. Bony septa were crossing the lesion (Fig. 1). Computed tomography showed an expansile mass with multiple internal septations. There were fluid levels within the multiple cysts (Fig. 2).

A biopsy was done, after which the scapula was removed. The removed scapula measured 16×8.5×5.5 ㎝. The cut surface of the scapula (Fig . 3)

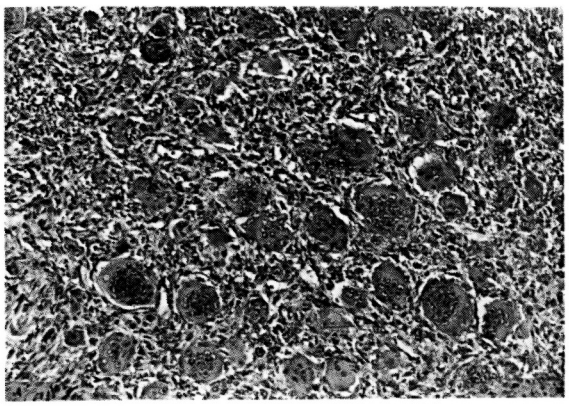

Fig. 4. Photomicrography of the giant cell lesion showed evenly dispersed giant cells having numerous giant cells (H-E, × 200).

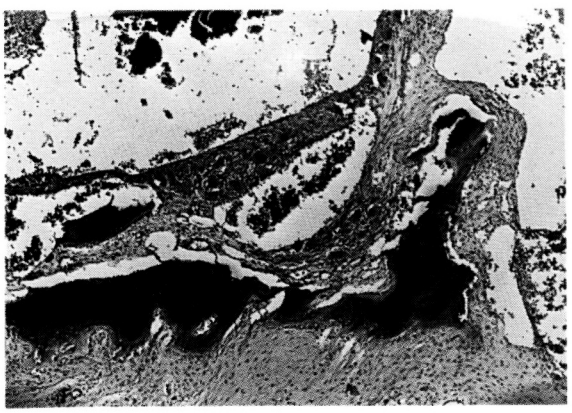

Fig. 5. The aneurysmal bone cystic lesion showed varing sized cystic lesions lined by fibroblasts and multinucleated giant cells (H-E, × 100).

showed multiple large cystic cavities with a few fragments of friable red tissued on their walls. In areas, there was a meshwork of multiple cysts varying in size. The cysts were filled with blood clots. The inferior angle of the scapula was spared.

PATHOLOGIC FINDINGS

The first biopsy and resected specimen showed identical histological features. On a low-power view, a uniform distribution of giant cells, which were huge and filled with numerous nuclei, was seen. The nuclei were round to oval, uniform in size, each separable from neighboring nuclei, and tended to collect around the center. The chromatin was blandly granular and the nucleolus was prominent. The stromal cell elements were round to oval. Each cell contained a single nucleus (Fig. 4). The walls of these cysts were formed by thin fibrous septa. Between the septa, there was solid, soft and fribale tissue. The cysts were lined by fibroblasts and multinucleated osteoclast-type giant cells. Fibrous septa were incomplete strands that had a sinusoidal shape and contained trabeculae of osteoid and woven bone and multinucleated giant cells (Fig. 5).

DISCUSSION

Giant cell tumors of the bone apparently arise from the mesenchymal cells of the bone marrow. In earlier literature, the diagnosis of giant cell tumor was frequently applied to many bone lesions with multinucleated giant cells (Jaffe et al., 1940). Therefore, there were only a few reports of a large series of giant cell tumors which fulfill currently accepted diagnostic criteria.

These giant cell tumors usually are discovered in the third and fourth decades of life (60 to 70 per cent of all tumors), and preponderantly are found in women (approximately 3 to 2 of all occurrences). Pain is the most common symptom, followed, in order of frequency, by local swelling and limitation of motion in the adjacent articulation (Dahlin and Unni, 1986 ; Resnick and Niwayama, 1988 ; Mirra et al., 1989).

Most giant cell tumors are found at the ends (epiphyses) of long bones. The distal portions of the femur and radius and the proximal portion of the tibia are the most characteristic sites of involvement (approximately 50 per cent of cases). In long tubular bones, giant cell tumors have been considered epiphyseal lesions. This consideration is supported by the fact that the vast majority of giant cell tumors occur in patients who are skeletally mature with closed epiphyseal plates (Dahlin and Unni, 1986 ; Resnick and Niwayama, 1988 ; Mirra et al., 1989). Other than long tubular bones, giant cell tumors involving the vertebra, ribs, skull, patella, clavicle and sternum were rarely reported (Kelikian and Clayton, 1957 ; Dahlin, 1977 ; Sundaram et al., 1982 ; Savini et al., 1983 ; Shirakuni et al., 1985). This rarity of involvement sites often leads to misdiagnosis, both clinically and roentgenographically.

In none of the large recent series of giant cell tumors was the scapula the site of the primary lesion (Jaffe et al., 1940 ; Dahlin et al., 1970 ;

Goldenberg et al., 1970 ; McGrath, 1972 ; Sung et al., 1982). An isolated example of giant cell tumor of the scapula was mentioned by Windeyer and Woodyatt (1949)). Samilson et al. (1968), in a series of thirty-one personally studied tumors of the scapula, reported one grade II giant cell tumor. In 1974, Tuli et al. (1974) reported a case of scapular giant cell tumor.

The lesion, such as unusual location of giant cell tumors should be differentiated from hyperparathyroidism and osteosarcoma with a large number of giant cells (MIrra et al., 1989 ; Unni, 1990). Differentiation from hyperparathyroidism is easily made in this case on the basis of the subject's serum chemistry data and parathormone level. In the case of osteosarcoma with unusual location, the subject's radiography, though aggressive appearing, show a peripheral shell suggesting a benign process, specifically aneurysmal bone cyst. We could not identify any cytological features of malignancy, however. So we could rule out osteosarcoma.

An aneurysmal bone cyst is an expansile lesion containing thin-walled, blood-filled cystic cavities. It is generally regarded as non-neoplastic in nature, resembling in some of its radiographical or histological features such reactive processes. It is also well documented that lesions resembling aneurysmal bone cysts accompany a variety of benign processes of the skeleton, including chondroblastoma, chondromyxoid fibroma, osteoblastoma, giant cell tumor and fibrous dysplasia and less frequently, some malignant tumors such as osteosarcoma, chondrosarcoma, and hemangioendothelioma (Clough and Price, 1968 ; Buraczewskin and Dabska, 1971 ; Levy et al., 1975 ; Bonakdarpour et al., 1978 ; Dahlin and McLeod, 1982 ; Diercks et al., 1986). The coexistence of aneurysmal bone cyst and a companion lesion is again consistent with the concept that a precursor tumor or event (e.g., trauma) leads to local hemodynamic changes, providing the ideal environment for the superimposition of a secondary ABC (Clough and Price, 1973). It is also possible that alterations in osseous hemodynamics resulting from the companion process give rise to rapid enlargement of an already existing aneurysmal bone cyst. In Korea, after first reported by Park et al. (Park et al., 1982), several additional cases of secondary associated aneurysmal bone cysts were found. In the 1982 Park et al. series, three of the cases of giant cell tumors were the most common primary bone lesions and two cases of fibrous dysplasia were the second lesion. In the 1975 Levy et al. series, the most common lesion was solitary bone cysts ; the second most common was giant cell tumor (Levy et al., 1975). In the 1978 Bonakdarpour et al. series, the most common tumors were giant cell tumors. (Bonakdarpour et al., 1978). This case was reviewed by Unni (1990) at the Mayo Clinic, Rochester, Minnesota. He thought this case was a giant cell tumor with abundant secondary aneurysmal bone cyst.

The purpose of this paper is to report on the rarity of the location of giant cell tumor and to emphasize association of the aneurysmal bone cyst.

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