

Osteoclastoma In An Infant

Eyichukwu G.O. MBBS FWACS, Ogugua P.C. MBBS

Department of Orthopaedic Surgery, National Orthopaedic Hospital Enugu, Enugu State, Nigeria

Abstract

Background: Conventionally, osteoclastoma is a benign but locally aggressive bone tumour with a tendency for local recurrence and rarely distant metastasis, diagnosed mainly in young adults, 15-40 years of age. Diagnosis before skeletal maturity is circumspect. We report a case of giant cell tumour of the clavicle (uncommon site) in a 2½ year old girl that was first noticed at 6 months of age.

Method: The case files of the patient as well as relevant literature were reviewed.

Results: A 2½-year-old girl presented with a 2-year history of right clavicular swelling first noticed at 6 months of age. The mass was initially painless and very slow growing involving the medial third of the clavicle. It rapidly increased in size to involve the entire clavicle with associated pain following manipulation by a traditional bone setter about 6 months after onset. She had incisional biopsy; the histological report of which was osteoclastoma. She then had excision of the mass with the involved clavicle uneventfully. She is being followed up for recurrence.

Conclusion: Osteoclastoma can develop in early childhood far before skeletal maturity, may be congenital in manifestation and can affect any bone in the body.

Key Words: Osteoclastoma, Infant, Enugu.

INTRODUCTION

Giant cell tumour is a benign but locally aggressive bone tumour exhibiting a tendency for local recurrence. It accounts for about 4% - 5% of primary bone tumours with approximately 1% of cases presenting as multiple synchronous or metachronous lesions, described as multicentric giant cell tumours.¹ At the time of involvement, about 80% of patients are from 20-40 years old.² The diagnosis before skeletal maturity is circumspect. There is a slight predilection for females.^{1,2}

About 50% of the tumour occur about the knee - the distal femur, and proximal tibia;³ other sites in decreasing order of frequency, are the distal radius, proximal humerus, proximal fibula, distal tibia, distal ulna, proximal femur proximal fibula, vertebral bodies, bones of hands and

feet. Pelvic bones, ribs and skull bones are rarely involved. No series has reported a tumour involving the clavicle, occurring in infancy to our knowledge.

Osteoclastoma presents the most difficult management problems of the benign bone tumours in part because of the difficulty in accurately predicting its natural history and in part because of its location next to the articular surface. This paper is a case report showing that this tumour can occur in early childhood as opposed to what have been reported so far in the literature and it can also occur in any site in the body.

CASE REPORT

A 2½-year-old child presented to us 12 months ago with a 2-year history of a swelling over the right clavicular region first noticed by her mother when the patient was 6 months old. The swelling was initially tiny on the medial third of the clavicle and asymptomatic, but then rapidly increased in size and became painful after massage and bandaging by a traditional bone setter. There was no history of trauma and no constitutional symptoms. Her obstetric history was unremarkable. She is the 4th of 5 children with no similar illness among her siblings.

Physical examination revealed a young girl who was in good general condition. She was not pale and was afebrile. There was an 8 by 5 by 3 cm elliptical well circumscribed mass over the right clavicle, firm and attached to the surrounding soft tissues and overlying skin. There were no palpable regional lymph nodes. Her chest was clinically clear.

A clinical diagnosis of a right clavicular tumour was made. Her haemoglobin was 12.9g/dl and total white cell count was 7200/mm³ with relative lymphocytosis. X-rays showed an expanded, rather fusiform entirely involved clavicle with thinned out cortex that were breached in areas. There was a lucent medullary cavity with areas of mottling with increased soft tissue shadows around the bone. Chest X ray was normal.

An incisional biopsy was performed and the histological diagnosis was osteoclastoma. The tumour became fungating after the procedure and she later had wide excision of the mass with the entire clavicle which was histologically reconfirmed as osteoclastoma. She is being followed up for recurrence.



DISCUSSION

Giant cell tumours are diagnosed mainly in adolescents and young adults; age ranging from 15-40 years.² There is a slight female predilection (3:2).² Diagnosis before skeletal maturity is uncommon. In a series of 218 cases by Goldenbeg et al skeletal maturity as evidenced by closure of the epiphyses was present in all the cases. This observation may explain the preponderance of females in patients less than 20 years old.³ The youngest patient with

a reported case of giant cell tumour is a girl of 11 years (she had multicentric giant cell tumour).¹ Other investigators have also noted that patients with multicentric tumours are considerably younger than those with a solitary giant cell tumour,^{4,5} though this has not been a consistent finding.⁶ Our patient was 2½ years old at diagnosis but the tumour had been noticed when she was 6 months old. This implies that this tumour could have been diagnosed at 6 months of age if she had presented promptly. It is thus our intention to bring this case to the knowledge of our colleagues so that osteoclastoma could be considered a diagnostic possibility of primary bone tumours in early childhood. Equally of importance is the fact that since this tumour was noticed at 6 months of age, it is a possibility that the process of tumour formation started in utero and that the tumour could manifest as a congenital entity.

The commonest site of occurrence is about the knee involving the distal femur and proximal tibia followed by the proximal humerus, distal radius proximal femur, distal tibia, the small bones of hands and feet and more rarely the sacrum spine and skull.^{1,3} This patient had osteoclastoma of the clavicle which is an uncommon location not yet reported in the literature. It can therefore be stated that osteoclastoma can occur in any bone in the body. In these bones, the tumour occurs at the metaphysis-epiphysis region adjacent to the joint but not invading it.

This tumour presents the most difficult management problems of the benign bone tumours in part because of the difficulty in accurately predicting its biological behaviour and partly because of its location next to the articular surface. Surgery is the mainstay of treatment and in planning the surgical approach, the risk of local recurrence and the anticipated functional result should be balanced. Radiotherapy may play some role in inaccessible lesions like the spine and post-excision residual tumour. Surgical options include intralesional curettage with or without physical adjuvants (phenol, liquid nitrogen, H₂O₂ etc), combined with bone grafting or acrylic cementing, excision and reconstruction with osteoarticular grafts or endoprosthesis and very rarely amputation.

Our patient had excision of the tumour with the entire clavicle (the bone was completely involved), the rationale being to greatly minimize local recurrence. It has been found that the adequacy of surgical removal is the most significant factor in recurrence.^{3,7,8} Tumour site; gender, Campanacci's radiological and Jaffe's

histological gradings and presence of pathological fracture have no apparent correlation to recurrence^{3, 9}. Reconstruction of the excised clavicle will be Herculean in this environment with lack of vascularized bone grafting techniques, and thus the functional outcome of the treatment may be suboptimal; however the rationale for the treatment offered has been highlighted above.

Benign giant cell tumour has been reported to metastasize to the lungs (prevalence 2-3%).¹⁰ Solitary metastases to other sites including the regional lymph nodes, the scalp and pelvis have also been reported. Complete excision of pulmonary nodules in early cases is frequently curative. Both radiotherapy and chemotherapy

have had only limited success and radiation is associated with secondary malignant change. Thus a metastatic lesion if at all possible should be resected. There was no evidence of pulmonary or other distant site metastatic involvement in our patient; however she is being followed up for recurrence or metastases.

CONCLUSION

Giant cell tumour of bone can occur in early childhood and should be considered a diagnostic possibility in children of all ages. Furthermore, it could manifest at birth and no bone is exempt from this tumour.

References

- (1). Benjamin Hoch, Carrie Inwards, Murali Sundaram and Andrew F Rosenberg: Multicentric Giant cell tumour of Bone: Clinicopathologic analysis of thirty cases. *J Bone Joint Surg. Am* Sept 1 2006 88(9): 1998-2008.
- (2). Yasko et al: Benign bone tumours in Chapman's operative Orthopaedics 3rd Ed. vol. 3 P 2523.
- (3). Goldenberg PR, Campbell CJ, Bonfiglio M: Giant cell tumour of bone; an analysis of 218 cases. *J Bone joint Surg. Am* 1970 vol. 52:619-664.
- (4). Peimer CA, Schiller AL, Mankin HJ, Smith RJ: Multicentric giant-cell tumour of bone *J Bone Joint Surg Am* 1980; vol 62: 652-656.
- (5). Sim FH, Dahlin DC, Beabout JW: Multicentric giant cell tumour of bone. *J Bone Joint Surg. Am.* 1977; Vol. 59: 1052-1060.
- (6). Bacchini P, Bertoni F, Ruggieri P, Canpanacci M: Multicentric giant cell tumour of skeleton: *Skeletal radiology* 1995; vol. 24:371-374 (Medline)
- (7). Yu Ping Su, Wei Ming Chen, Tain Hsiung Chen: Giant cell tumour of bone: An analysis of 87 cases. *International Orthopaedics* 2004; vol. 28: 239-243.
- (8). McDonald DJ, Sim FH, Macleod RA and Dahlin DC. Giant cell tumour of Bone *J Bone Joint Surg. Am.* 1986; 68:235-242.
- (9). Larsson SE, Lorentzon R, Boquist L. Giant cell tumour of bone. A demographic, Clinical and histopathological study of all cases recorded in the Swedish cancer registry for the years 1958-1968 *J Bone Joint Surg. Am.* 1975, vol. 57: 167-173.
- (10). Rock MG, Sim FH, Unni KK et al. Secondary malignant giant cell tumour. Clinicopathological assessment of 19 cases *J Bone Joint Surg. Am* 1968 vol. 68: 1073-1079.