

Aneurysmal bone cyst of the hyoid

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Abstract

Aneurysmal bone cyst is a rare lesion usually of the long bones, well documented in the literature. It is a cystic, osteolytic vascular tumour, replete with giant cells and fibrous septa, yet devoid of endothelial lining. It has been reported in the larynx and maxillary sinus. This appears to be the first report of an aneurysmal bone cyst occurring in the hyoid bone.

Case report

An 18-year-old boy presented with a seven-month history of progressive mid-line suprahyoid swelling associated with odynia and dysphagia, with a past history of neck trauma. He could not protrude his tongue fully and an indirect laryngoscopy was unsuccessful.

Plain radiographs of the neck showed a diffuse circumscribed swelling of the soft tissues with scattered patches of ossification in the hyoid bone (Fig. 1). A thyroid scan was negative and the values of T_3 were 1 nmol/l, T_4 5 mcg/dl and TSH of (4 μ u/ml).

A CT scan showed 'a large mid-line rounded non-enhancing fluid dense mass, with an amorphous ring and eccentric cal-

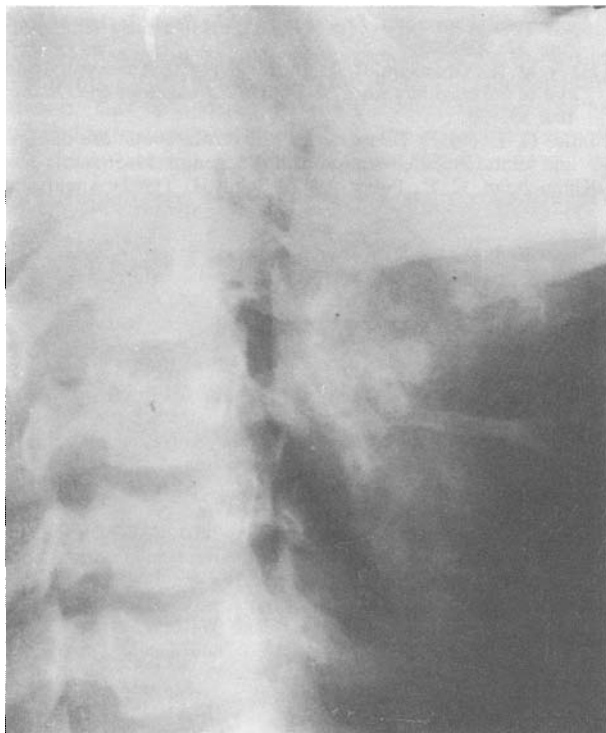


FIG. 1

X-ray lateral view of neck: Patchy calcification near a thinned out hyoid bone.

ification at the level of the hyoid bone' (Fig. 2). It extended between the surface of the tongue to the thyroid isthmus through the body of the hyoid bone.

Ultrasound examination showed a 'complex mass, solid in front of the neck with bright echogenicity and well defined walls.' Fine needle aspiration revealed frank blood even on repeated attempts. Carotid angiography did not show a vascular tumour. In view of these findings a provisional diagnosis of an infected thyroglossal cyst was considered.

Exploration of the swelling was carried out which revealed a cyst 4 × 6 cm, occupying the area from above the thyroid to the base of the tongue. The body of the hyoid bone was deficient and only the remnants of the right greater cornu could be identified. Approximately 50 ml of serosanguineous aspirate was withdrawn. The cyst was dissected away along with a patchily ossified cyst wall.

Post-operative recovery was uneventful. The initial histology report was an osteoclastoma, but on a subsequent review of the sections, keeping the age and history of trauma in mind, as well as the numerous cavernous spaces and absence of endothelial lining, a diagnosis of an aneurysmal bone cyst was confirmed.

Discussion

Aneurysmal bone cyst is classified as a tumour of blood vessels. It presents as a single cavity usually containing unclotted flowing blood. There are multiple cavernous vascular spaces that 'honeycomb' the lesions, unlined by endothelial and lack of characteristic blood vessels. The benign appearing stroma of spindle cells is sprinkled with giant cells (Jaffe, 1958).

In most cases, pre-existing pathology has been identified. Areas of resemblance to aneurysmal bone cyst occur adjacent to lesions of fibrous dysplasia, giant cell tumour, osteogenic sarcoma and benign osteoblastoma (Dabezies and Ferguson, 1982). Its exact aetiology is unknown but its association with trauma is certain. Aneurysmal bone cyst is seen to represent a reactive process in bone as a sequel to haemorrhage from primary lesion for example a fracture, leading to an osseous arteriovenous fistula. Biesecker *et al.*, (1970), Delorit and Summers (1975), and Kimmelman *et al.* (1982) in their reports of an aneurysmal bone cyst in the sphenoid sinus, noted radiological findings of bony destruction, opacification, and expansion.

The clinical diagnosis may be difficult in view of more likely possibilities such as thyroglossal cyst, haemangioma and ecto-

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Accepted for publication: 29 August 1991.

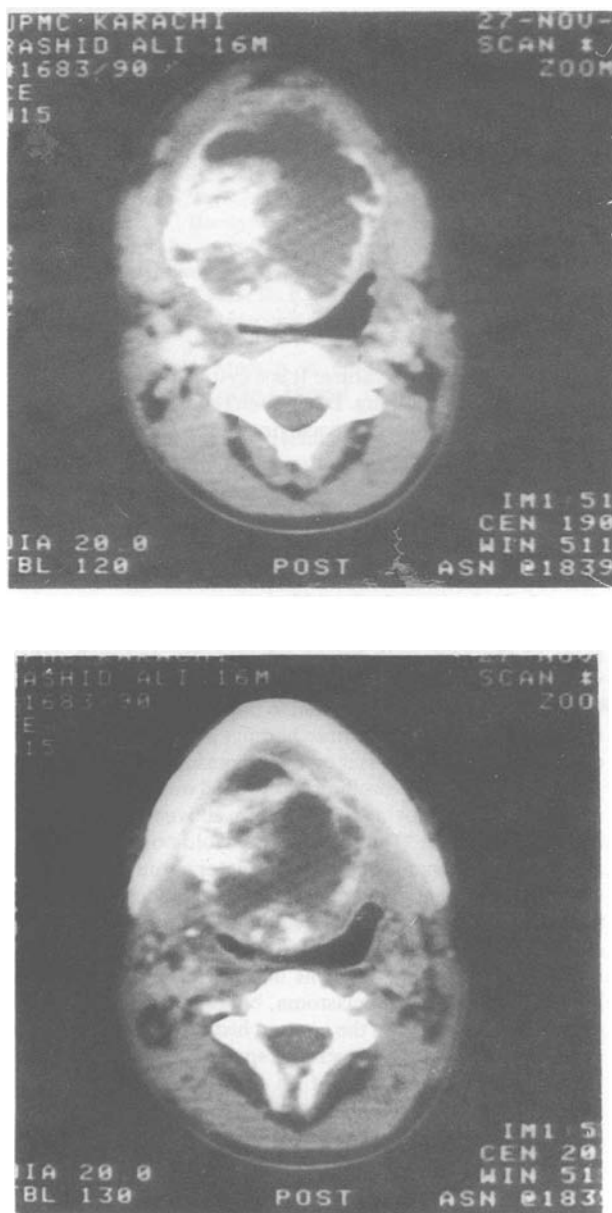


FIG. 2

CT scan: Showing a fluid dense mass, with eccentric calcification at the level of the hyoid bone. The cystic mass is obliterating the larynx and displacing the surrounding structures. The thyroid is separate from the cystic mass.

pic thyroid. Histological diagnosis is by differentiation from giant cell variants. Diagnosis should be based on a composite of multiple random sections of the tumour along with clinical and radiographic evidence (Jaffe, 1958). The lacy trabeculae provide the characteristic feel to the surgeon during curettage (Lichsenstein, 1957; Verbiest 1965; Spjut *et al.*, 1971). Thrombus admixed with the tumour is an occasional finding (Tillman *et al.*, 1968).

A definite chronological relationship between the occurrence of an aneurysmal bone cyst and an undisplaced fracture is noted by Dabezies and Ferguson (1982).

Beltran (1986) advocates the use of magnetic resonance imaging for diagnosis of an aneurysmal bone cyst, as this particular cyst has multiple fluid levels caused by intracystic haemorrhages of varying ages.

Several methods of treatment have been advocated: curet-

tage with or without grafting, cryotherapy and radiotherapy (Sherman and Soong, 1957; Godfrey and Gresham, 1958; Clough and Price, 1973). Radiotherapy in a benign condition with a tendency to spontaneous regression is unjustified (McQueen *et al.*, 1985). Radiotherapy has also been avoided on account of its potential carcinogenicity and damage to the growing ends of bone (Marks *et al.*, 1976). All these modes of treatment are associated with recurrence between 8 to 30 per cent (Donaldson, 1962; Dabska and Buraczewski, 1969).

As thrombosis and fibrolysis play a leading part in healing, therapeutic embolization has been used successfully to achieve spontaneous regression (Murphy *et al.*, 1982).

We believe total *en bloc* excision is the treatment of choice when mechanical or functional failure is likely. No recurrence follows this mode of treatment.

References

- Beltran, J. (1986) Aneurysmal bone cyst MR imaging at 1.5 T. *Radiology*, **158**: 689–690.
- Biesecker, J. L., Marcove, R. C., Huvos, A. G., Mike, V. (1970) Aneurysmal bone cysts. A clinico pathologic study of 66 cases. *Cancer*, **26**: 615–625.
- Clough, J. R., Price, C. H. G. (1973) Aneurysmal bone cyst: pathogenesis and long term results of treatment. *Clinic Orthopedics*, **97**: 52–63.
- Dabezies, E. J., Ferguson, A. B. (1982) Aneurysmal bone cyst after fracture. *Journal of Bone and Joint Surgery*, **64(A)**: 617–621.
- Dabska, M., Buraczewski, J. (1969) Aneurysmal bone cyst; pathology, clinical course and radiological appearance. *Cancer*, **23**: 371–389.
- Delorit, G. J., Summers, G. W. (1975) Aneurysmal bone cyst of the sphenoid sinus. *Transactions of the American Academy of Ophthalmology and Otolaryngology*, **80**: 438–443.
- Donaldson, W. F., Jr. (1962) Aneurysmal bone cysts. *Journal of Bone and Joint Surgery*, **44A**: 25–40.
- Godfrey, L. W., Gresham, G. A. (1958) The natural history of aneurysmal bone cyst. *Proceedings of the Royal Society of Medicine*, **52**: 900–905.
- Hady, M. R., Ghanaam, B., Hady, M. Z. (1990) Aneurysmal bone cyst of the maxillary sinus. *Journal of Laryngology and Otolaryngology*, **104**: 501–503.
- Jaffe, H. L. (1958) Tumours and tumorous conditions of bones and joints. Philadelphia Lae and Febrgen, p. 54–56.
- Kimmelman, C. P., Potsic, W. P., Schut, L. (1982) Aneurysmal bone cyst of the sphenoid in a child. *Annals of Otolaryngology and Laryngology*, **91**: 339–341.
- Lichsenstein, L. (1957) Aneurysmal bone cyst: Observations on 50 cases. *Journal of Bone and Joint Surgery*, **39A**: 873–878.
- Marks, R. D., Scurggs, H. J., Wallace, K. M., Fenn, J. O. (1976) Megavoltage therapy in patients with aneurysmal bone cyst. *Radiology*, **118**: 421–424.
- McQueen, M. M., Chalmes, J., Smith, G. O. (1985) Spontaneous healing of an aneurysmal bone cyst. *Journal of Bone and Joint Surgery*, **67(B)**: 310–312.
- Murphy, W. A., Strecker, W. D., Schoenecker, P. L. (1982) Transcatheter embolization therapy of an ischial aneurysmal bone cyst. *Journal of Bone and Joint Surgery*, **64(B)**: 166–168.
- Schilling, H. E. (1986) Aneurysmal bone cyst of the larynx. *American Journal of Otolaryngology*, **7(5)**: 370–374.
- Sherman, R. S., Soong, K. Y. (1957) Aneurysmal bone cyst. Its roentgen diagnosis. *Radiology*, **68**: 54–64.
- Spjut, J. J., Dorfman, H. D., Fechner, R. E., Ackerman, L. V. (1971) Tumours of bone and cartilage. Washington D.C. Armed Forces. *Institute of Pathology*, pp. 357–367.
- Tillman, B. P., Dahlin, D. C., Lipscomb, P. R., Stewart, J. R. (1968) Aneurysmal bone cyst: An analysis of 95 cases. *Mayo Clinic Proceedings*, **43**: 478–482.
- Verbiest, H. (1965) Giant cell tumours and aneurysmal bone cysts of the spine. *Journal of Bone and Joint Surgery*, **47(B)**: 699–704.

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Key words: Hyoid bone; Bone cyst, aneurysmal