ANEURYSMAL BONE CYST OF THE BASIOCCIPUT AS AN INTRACRANIAL SPACE OCCUPYING LESION

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SUMMARY :

An aneurysmal bone cyst of the basiocciput as an intracranial space occupying lesion is reported. The localization in the base of the skull is extremely are. Clinical and neuroradiological findings are described in detail and the pertinent literature is reviewed.

KEY WORDS :

Aneurysmal bone cyst-basiocciput-space occupying lesion.

INTRODUCTION

An aneurysmal bone cyst is a benign, usually solitary expansile lesion of bone most often occurring in the metaphysis of long bones and in vertebrae and flat bones. Localization of aneurysmal bone cyst in the bones of the skull has rarely been mentioned although all bones have been reported as the site. From a collected series of 193 cases the distribution of the lesion was 18 % upper limb, 34 % lower limb, 27 % vertebrae, 9 % thorax, 7 % pelvis and 6 % skull and mandibula (16). Keuskamp et al. were able to collect only 43 cases of aneurysmal bone cysts of the skull reported in the world literature from 1942 to 1980 (12). We were able to find 11 more cases which were reported since 1980 (2, 3, 5, 6, 10, 15, 17) and are now reporting an additional case.

Since the initial recognition as a distinct entity by Jaffe and Lichtenstein in 1942, the aneurysmal bone cyst has continued to be a controversial lesion. Ewing described it ats aneurysmal or benign form of giant cell tumor (9) and this entity is still felt by some to be a giant cell variant. However, both Lichtenstein and Dahlin considered aneurysmal bone cyst as a distinct clinical and pathological entity (8, 14). The differential diagnosis between aneurysmal bone cyst and giant cell tumor is important because giant cell tumors show a malignant evolution in 10 % of cases, whereas aneurysmal bone cysts are benign lesions and recurrence is only seen after incomplete removel.

CASE REPORT

This 22-year old right-handed woman was admitted to Selçuk University Hospital with a two-month history of diffuse headache, nausea, vomiting and dizziness.

Examination: The patient was seen during her 7th month of pregnancy and physical examination showed a well-developed woman. On neurological examination bilateral horizontal nystagmus, broad-based gait, mild incoordination of the extremities and neck stiffness were observed. Radiological evaluation revealed an irregular lytic area within the left suboccipital region with a blow-out appearance. Computerized tomography demonstrated a multiloculated lesion originating in the diploe of the left part of the suboccipital bone and expanding intracranially (Fig.1). There was irregular destruction at the left suboccipital bone, posterior rim of the foramen magnum and petrous apex. The brain stem was compressed and the fourth ventricle was displaced anteriorly and contralaterally. Minimal enlargement of the third and

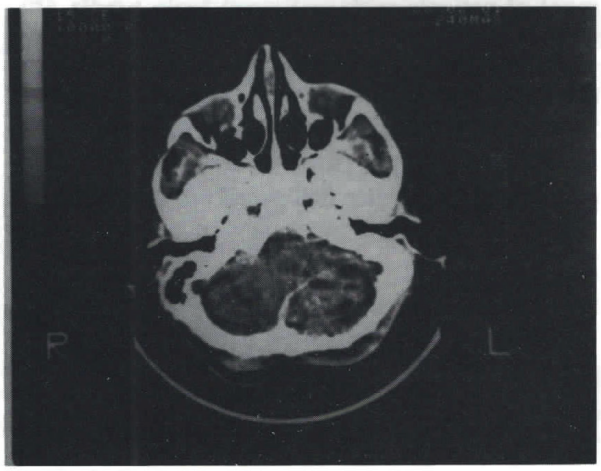

Fig.1 : The lesion, originationg in the diploe and expands intracranially

lateral ventricles was present. The contents of the cavities within the lesion had different densities. After injection of contrast medium some of the cavities enhanced strongly but some remained unchanged (Fig.2). Because of her pregnancy we decided to perform the operation with minimal radiological investigation and postponed the arteriographic studies.

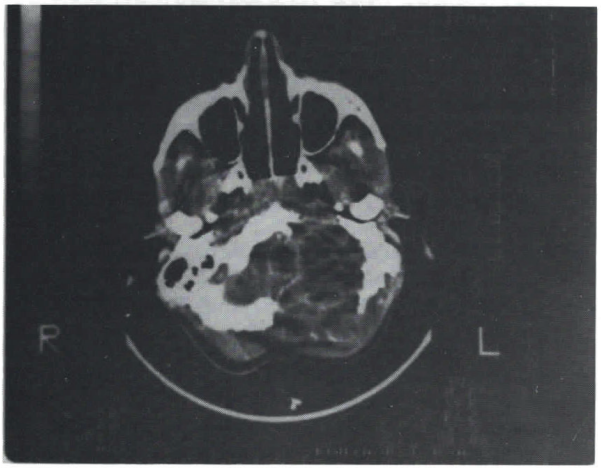

Fig.2 : The aneurysmal bone cyst presents areas with different densities separated by bony septa.

Operation : A left-sided suboccipital craniectomy was performed with a paramedian vertical incision. When the muscles were split a massive hemorrhage developed. After all the muscles had been completely split and the suboccipital bone was exposed a large bony defect was seen at the suboccipital region and a partly necrotic, moderately firm, reddish neoplastic mass protruded through the bony defect and the dura mater was displaced upwards and medially. Because of the massive hemorrhage we aspirated the contents of the mass and curetted some of the cavities in order to remove the mass subtotally. After careful hemostasis, the scalp wound was closed. The postoperative course was uneventful except for minimal left peripherial facial palsy.

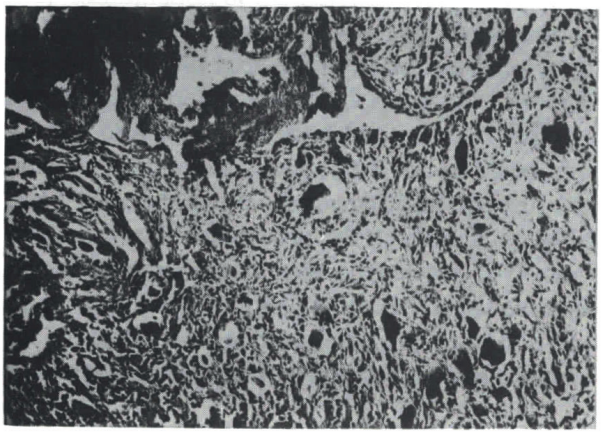

Fig.3 : Photomicrography of lesion showing islands of bony tissue, fusiform cells and multinucleated osteoclastic giant cells.

Microscopic findings: Microscopic examination of the lesion disclosed islands of bony tissue and blood-filled channels of different sizes. The lesion contained fusiform cells and among them multinucleated osteoclastic giant cells (Fig.3).

DISCUSSION

Aneurysmal bone cyst was first described by Jaffe and Lichtenstein in 1942 as an aneurysmal cyst of bone (11). The lesion was subsequently established as a pathologicallentity by Lichtenstein in 1950 (14). Although further studies of this lesion by Lichtenstein, Tillman, Sherman and others have added greatly to our knowledge concerning its diagnosis and course, aneurysmal bone cyst is not a fully understood illness because of the insufficient number of cases.

The pathogenesis of this lesion remains obscure. Among hypotheses concerning the pathogenesis of aneurysmal bone cysts, Lichtenstein's concerning a local hemodynamic disturbance was supported by other authors for a long time. Lichtensteinpostulated a circulatory abnormality such as distal venous occlusion or arteriovenous malformation with resulting increased venous pressure causing a dilated, engorged capillary bed in the involved bone. Subsequent reports have confirmed the association of aneurysmal bone cysts with osteoclastoma, osteosarcoma, non-osteogenic fibroma, osteoblastoma, hemangioendothelioma hemangioma of bone and fibrous dysplasia (1, 3, 4, 13). These reports support the view that a secondary form of aneurysmal bone cyst may occur. Alteration of other pre existing lesions such as fibro-osseous tumors has become an increasingly popular concept. Levy et al. reported 57 cases of aneurysmal bone cyst associated with multiple underlying abnormalities (13) but Tillman et al. found no preexisting lesion in their 95 cases (18) and moreover Ikeda and Niizuma treated a nonaneurysmal bone cyst successfully by using estrogen for the first time in 1982, which is one of the chemical embolizing agents, and supported the view of local hemodynamic disturbance as initial cause of the lesion (10).

An aneurysmal bone cyst as an intracranial space-occupying lesion was first reported by Costantini and Iraci in 1966. These authors portulated that whatever the precipitating agent or incident, it affects the periosteal and bony tissue at the site of implantation of the cyst and leads to erosion of the dura and added that there may then have been proliferation of the internal periosteum throught this erosion with the formation of an incracranial, subdural aneurysmal bone cyst following (7).

The existence of aneurymal bone cyst arising primarily in the base of the skull is generally regarded as exceptional. Calliauw was able to collect only three cases reported in the literature and in 1985 (5) added two more cases which were located at the temporal fossa. As far as we know, except the special case of Verbiets (19), in which the tumor was situated in the right and medial portion of the atlas and extended into the right posterior cranial fossa with destruction of the right atlanto-occipital joint, this is the first case of an aneurysmal bone cyst which was located in the basiocciput as an intracranial space-occupying lesion.

Although many authors agree that aneurysmal bone cysts in the skull are readily recognizable lesions and their radical surgical removal is easily possible, there are special difficulties in the diagnosis and treatment of cases with localization in the base of the skull. Complete removal of these cysts is impossible and we think chemical embolization will not be effective in aneurysmal bone cysts as an intracranial space-occupying lesion.

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