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Chondroblastoma arising in the triradiate cartilage

Report of two cases with review of the literature

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Abstract. Chondroblastoma is a relatively rare benign bone tumor of cartilage origin. Roentgenologically it presents usually as a region of lytic destruction of bone with a thin sclerotic rim in the epiphysis of long tubular bone. Less than 9% occur in the pelvic bones but show a tendency to arise from the triradiate cartilage. We present two cases of chondroblastoma originating in the triradiate cartilage, each showing extensive lytic bony destruction and an intrapelvic soft tissue mass. A review of the literature suggests that chondroblastoma of the triradiate cartilage shows an aggressive radiological appearance.

Key words: Bone tumors – Chondroblastoma

Chondroblastoma is an uncommon benign tumor of bone, comprising less than 1% of all primary bone tumors [5, 6]. It occurs mostly in young individuals, and approximately 65% of patients are in their second decade [5, 6, 19, 26]. The tumor arises most often in the epiphyses of long tubular bones, especially the proximal humerus, proximal tibia, and distal femur. Chondroblastoma also arises in flat bones, and only 2.9% to 8.8% are reported to arise in the bony pelvis [1, 2, 5, 6, 11, 19, 26, 27, 30]. The triradiate cartilage is the most common site of origin of chondroblastomas within the pelvic bone [18, 34]. A few cases are illustrated in the literature [2, 5–7, 15, 17, 18, 21–23, 34].

Although most chondroblastomas show a benign clinical course, several reports record aggressive behavior and even malignancy [3–8, 10, 13, 15–17, 19, 20, 23–25, 28, 29, 31–33, 35, 36]. A number of these involved the pelvic bones or triradiate cartilage (Table 1).

We report two cases of chondroblastoma of triradiate cartilage origin with aggressive roentgenological appearances.

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Case reports

Case 1: A 14-year-old female was seen first in October 1981 with increasing pain in her right hip. Physical examination was normal except for pain on full flexion of the affected hip. All blood tests were normal.

Plain radiographic examination showed lytic involvement of the right acetabular region with destruction of the inner cortex. No features suggestive of calcification or ossification were observed (Fig. 1A, B). Computed tomography (CT) studies demonstrated lytic destruction of acetabulum, with involvement of subchondral bone, and an intrapelvic soft tissue mass. No calcification was observed (Fig. 2). These findings indicated a benign tumor arising in the triradiate cartilage.

Biopsy showed proliferation of small polygonal cells with eosinophilic cytoplasm and clearly-defined borders. The nuclei were round or oval with slight anaplasia. Osteoclastic giant cells were observed, especially around hemorrhagic foci (Fig. 3A). Chondroid or partly osteoid-like tissue was prominent throughout the specimen (Fig. 3B). However, the so-called "chicken-wire" calcification, said to have diagnostic value of chondroblastoma, was not observed. On the basis of these histological features, the diagnosis of chondroblastoma was made, and curettage with bone grafting was performed (November 1981). The patient is well and without recurrence 5 years after the operation.

Case 2: A 16-year-old male was first admitted to our hospital in April 1982 for left inguinal pain of 11 months duration. Plain roentgenograms showed lytic destruction of the left pubic bone with cortical expansion. No abnormal densities suggesting calcification or ossification were observed (Fig. 4). CT studies demonstrated destruction of the left pubic bone with involvement of subchondral bone. Spotty calcifications were observed within the tumor (Fig. 5). Although from all these roentgenological findings, benign bone tumors like chondroblastoma or aneurysmal bone cyst were considered, the size, cortical destruction, and intrapelvic expansion of the neoplasm did not allow exclusion of a malignant bone tumor such as osteosarcoma or Ewing sarcoma.

Table 1. Reported cases of chondroblastomas arsing in the bony pelvis

Case	Source	Age/ Sex	Location	Roentgenological appearances	Course and treatment
1	Plum and Pugh (1958) [22]	?/?	Right iliac wing	Mottled calcification with sclerotic rim	No information
2	Plum and Pugh (1958) [22]	54/F	Right triradiate cartilage	Large expansion with sharp sclerotic margin	Hindquarter amputation Died of tumor 24.5 years after: no known metastasis
3	Plum and Pugh (1958) [22]	?/?	Right ilium and ischium (? triradiate cartilage origin)	Large intra-pelvic mass	Biopsy: patient refused further surgery Died of tumor (?): sometime between 34 months and 8 years
4	Kahn et al. (1969) [15]	13/M	Right pelvic bone	Recurrent lesion showed complete destruction of right pelvis with soft tissue extension and spotty calcification	Series of curettages and resections. Died with widespread metastases 15 years after the initial treatment
5	Schajowicz and Gallardo (1970) [26]	17/F	Left iliac wing	Large heavily calcified tumor	Resection and radiation therapy. Several recurrences. Well and alive one year after resection
6	Schajowicz and Gallardo (1970) [26]	17/M	Right ischiopubic ramus (? triradiate cartilage origin)	No roentgenological information	Curettage and radiation therapy. Well and alive one year after
7	Dahlin and Ivins (1972) [6]	?/?	Ischium and pubis (? triradiate cartilage origin)	Cystic and expanded (No roentgenological illustration)	Repeated radiation therapy. Died with pulmonary metastasis 58 months after the initial treatment
8	McLeod and Beabout (1973) [18]	?/?	Left triradiate cartilage	Large expanded lesion with sclerotic rim	No information
9	McLeod and Beabout (1973) [18]	?/?	Right triradiate cartilage	Lesion with dense periosteal reaction	No information
10	Nolan and Middlemiss 41/M (1975) [21]	41/M	Left ilium (? triradiate cartilage origin)	Large osteolytic lesion with some sclerotic rim	Curettage and well for one year
11	McLaughlin et al. (1975) [17]	25/M	Left ilium, ischium and pubis (? triradiate cartilage origin)	Large tumor with dense calcification	Wide excision and well after 4 years and 5 months
12	Campanacci et al. (1977) [2]	20/?	Right iliopectineal eminence (? triradiate cartilage origin)	Large cystic lesion with expansion	Curettage and well one year after
13	Reyes and Kathuria (1979) [23]	32/M	Left ilium, ischium and pubis (? triradiate cartilage origin)	Large lytic destruction with calcification	Partial resection followed by hemipelvectomy 10 years after. Died of metastases 32 years after the initial treatment
14	Enneking (1983) [7]	8/M	Right supra-acetabular region (? triradiate cartilage origin)	Well-marginated radio-lucent lesion	Biopsy: patient refused further surgery and returned with huge intrapelvic, mass. Wide en bloc excision by hemipelvectomy with iliofemoral fusion was performed. No further information
15	Current study Matsuno et al. Case 1	14/F	Right triradiate cartilage origin	Osteolytic lesion with large soft tissue extension	Curettage and bone graft. Well 5 years after
16	Matsuno et al. Case 2	16/M	Left pubis involving subchondral bone	Osteolytic with cortical expansion	Curettage and bone graft. Well 4 years 5 months after

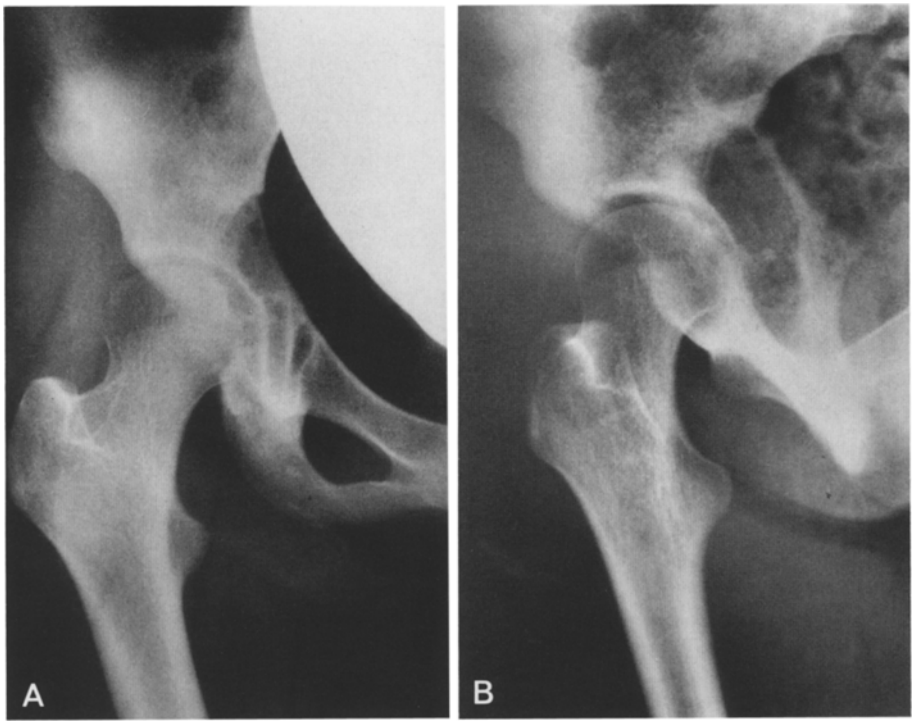


Fig. 1. A Radiograph of the right hip (Case 1), showing lytic bony destruction of the acetabular region.
B Oblique view of the pelvis showed an expanded lesion with involvement of the medial cortex

Biopsy was performed and most of the lesion showed a pattern resembling an aneurysmal bone cyst with large cystic spaces containing red blood cells. Solid tumor tissue was observed only in a small portion of the tumor (Fig. 6A). A higher power view of the solid element of the tumor revealed proliferation of tumor cells. These were round to polygonal and uniform in size with eosinophilic or granular cytoplasms. The nuclei were round or oval with no anaplasia. Osteoclastic giant cells were scattered throughout the specimen. Stromal calcification was prominent in the solid areas, and so-called "chicken wire" calcifications were prominent (Fig. 6B). A diagnosis of chondroblastoma was made, and curettage with bone grafting was performed (June 1982). At operation, the tumor was found to be almost totally cystic. A small amount of tumor tissue was obtained only from cyst wall. The histological appearance was the same as the biopsy specimen. The patient is well without recurrence 4 years and 5 months after the operation.

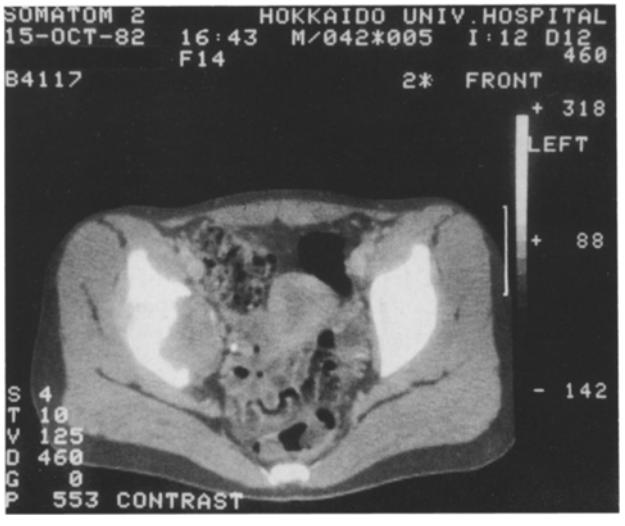


Fig. 2. CT study demonstrating lytic destruction of the acetabulum and involvement of subchondral bone

Discussion

Chondroblastoma was named and described as a distinct entity, separate from giant cell tumor of bone, by Jaffe and Lichtenstein in 1942 [14]. Since then several hundred cases have been reported. More than 60% of chondroblastomas occur before the closure of the growth plate, with a male predominance in the ratio of 2:1 to 3:1 [2, 6, 19, 26, 30]. More than 60% of chondroblastomas are located in the epiphysis of a long tubular bone; those of pelvic origin are relatively rare [1, 5, 6, 26, 30].

The most common location of chondroblastoma in the bony pelvis is the triradiate cartilage [18, 34]. Table 1 records the 16 pelvic cases reported in the literature.

In reviewing all these reported cases, many seemed to show clinical and roentgenological evidence of an aggressive nature and some even showed malignant changes. Roentgenologically, 12 out of 13 cases (Cases 1–4, 8, 10–16) showed aggressive features with extensive lytic destruction of bone, expansion of cortex, and a large intrapelvic soft tissue mass. Dense calcification was prominent in five cases (Cases 1, 2, 4, 11, 13). According to

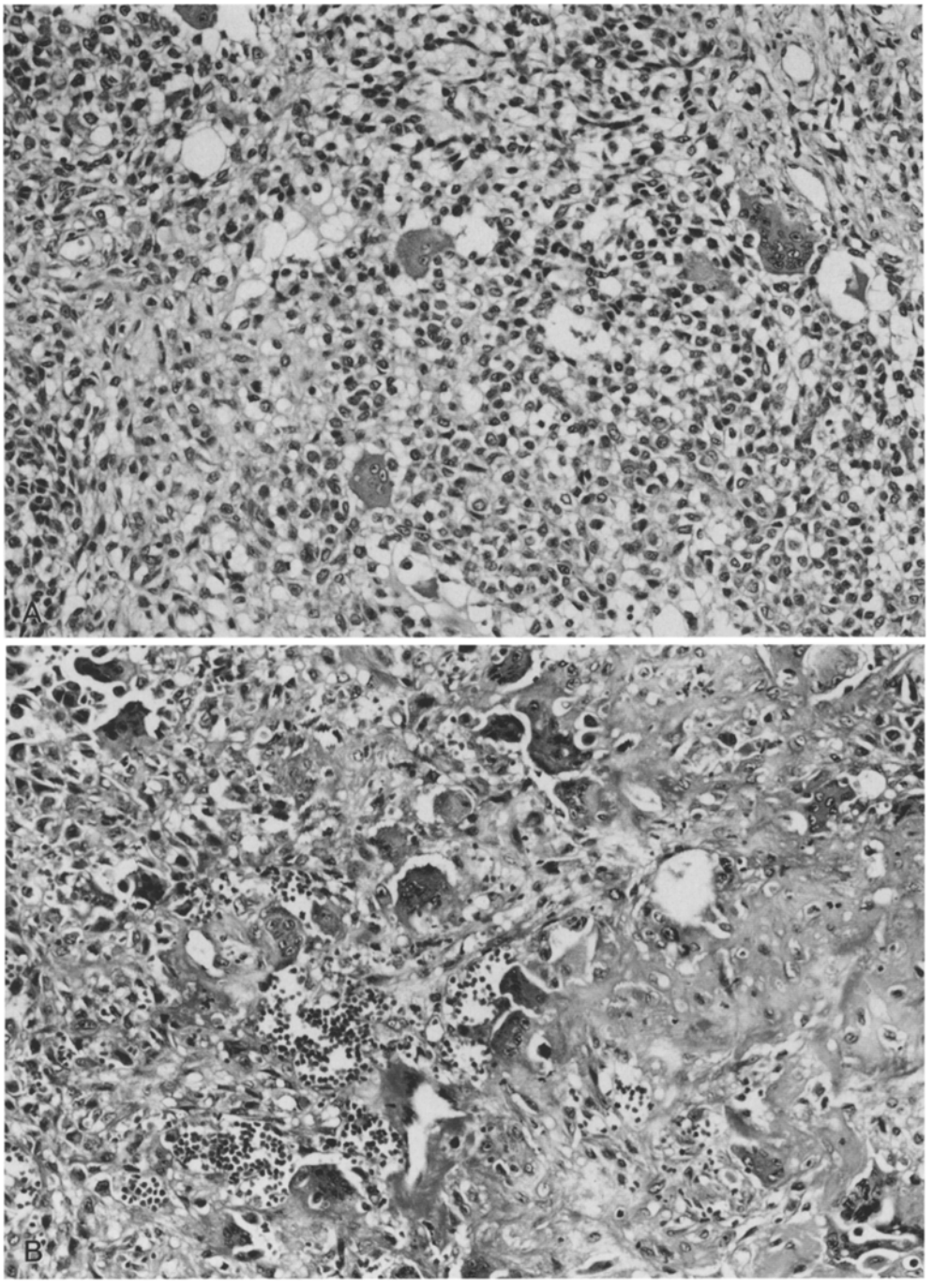
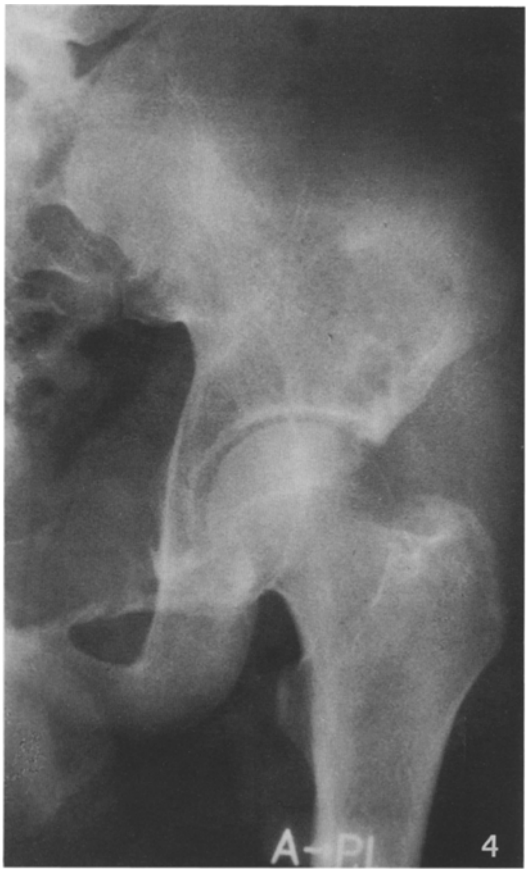


Fig. 3. A Photomicrograph (hematoxylin and eosin, ×100) showing proliferation of small polygonal cells with well-defined cell borders and sparse giant cells of osteoclastic type. **B** Photomicrograph (hematoxylin and eosin, ×100) showing chondroid tissue and some osteoid formation. Osteoclastic-type giant cells are observed near the hemorrhagic foci

Kiriakos et al. [16] who reviewed and summarized all cases of atypical, aggressive, metastasizing and malignant chondroblastomas, 3 out of 11 atypical or locally-aggressive chondroblastomas were of pelvic origin and 2 out of 10 chondroblastomas with metastases or malignant transformations also originated in the pelvis. Considering the relatively low occurrence of chondroblastomas in the pelvic bones, these figures suggest that chondroblastomas of pelvic bone, or triradiate cartilage, tend to be roentgenologically and clinically more aggressive than those of long tubular bones.

There could be several reasons to explain the aggressiveness of chondroblastomas in this location. (1) The tumor may be so deep-seated that it takes a relatively long time to cause clinical symptoms which leads to the aggressive appearances on initial roentgenograms. (2) The unusual



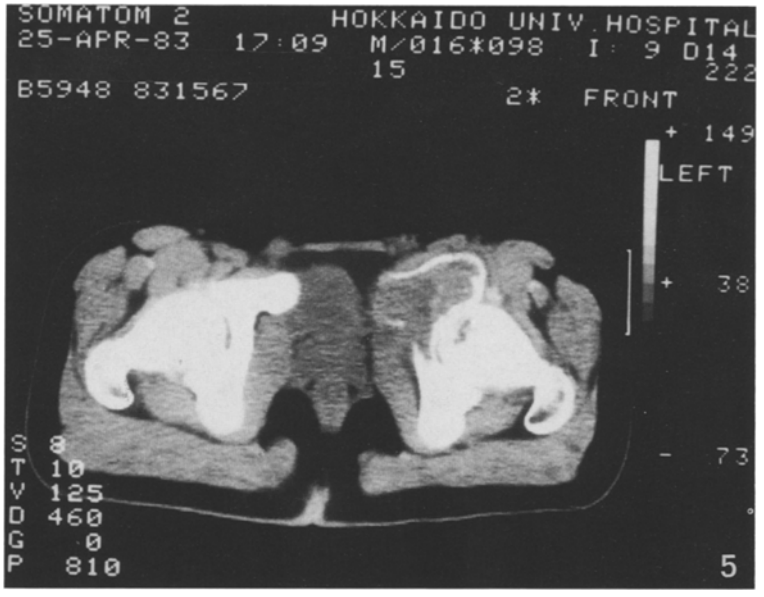


Fig. 4. Radiograph of the left hip (case 2), showing extensive destruction of the left pubic bone with cortical expansion

Fig. 5. CT study demonstrates expansion and destruction of the left pubic bone and acetabulum. Some spotty calcifications are present

location of chondroblastoma might cause problems resulting in misdiagnosis and mistreatment. (3) The location of the tumor makes complete excision or resection difficult. (4) The architecture of the pelvic bone may allow the tumor to expand the cortex and produce a large intra-pelvic mass. Bloem and Mulder [1] have already pointed out that cortical involvement and expansion occurs more often in flat bones (63%) than in long bones (31%). (5) Chondroblastoma of pelvic bone origin may be aggressive de novo.

From the histological point of view, an aneurysmal bone cyst-like feature seems to contribute to the aggressiveness of chondroblastoma and it seems to occur more frequently in pelvic lesions than those of long tubular bones [2, 5, 6, 18, 26]. Hudson and Hawkins [9] described this pattern as contributing to the aggressive appearance of chondroblastoma on roentgenograms and Huvos et al. [11–13] recorded 6 cases out of 25 chondroblastomas showing an aneurysmal bone cyst-like pattern with a recurrence rate significantly higher in the presence of this pattern.

The treatment of choice for chondroblastoma of triradiated cartilage origin is thorough curettage with bone grafting performed after wide exposure of the lesion [30]. Overdiagnosis or overtreatment which is only based on roentgenological aggressiveness should be avoided [5, 6, 30]. Resection is usually impossible without sacrificing the hip joint and should be reserved for the recurrent lesion [1, 2, 5, 30].

The prognosis of chondroblastoma is good with a cure to be expected after curettage with or without a bone graft. Its recurrence rate is generally considered to be low after adequate treatment [1, 2, 6, 20, 26, 30], although Huvos et al. [11–13] reported a high recurrence rate (35%). Although our two cases have shown no recurrence 5 years and 4 years and 5 months after curettage and bone graft, long-term follow-up is required as chondroblastomas have recurred or metastasized more than 4 years after initial treatment [5, 6, 15, 23, 29, 33, 35]. Although Schajowicz and Gallardo [26] reported five cases treated by radiation therapy with good clinical results, such therapy should be avoided because of the risk of inducing radiation sarcomas [5, 6].

In conclusion, chondroblastoma of the bony pelvis, or the triradiate cartilage may show aggres-

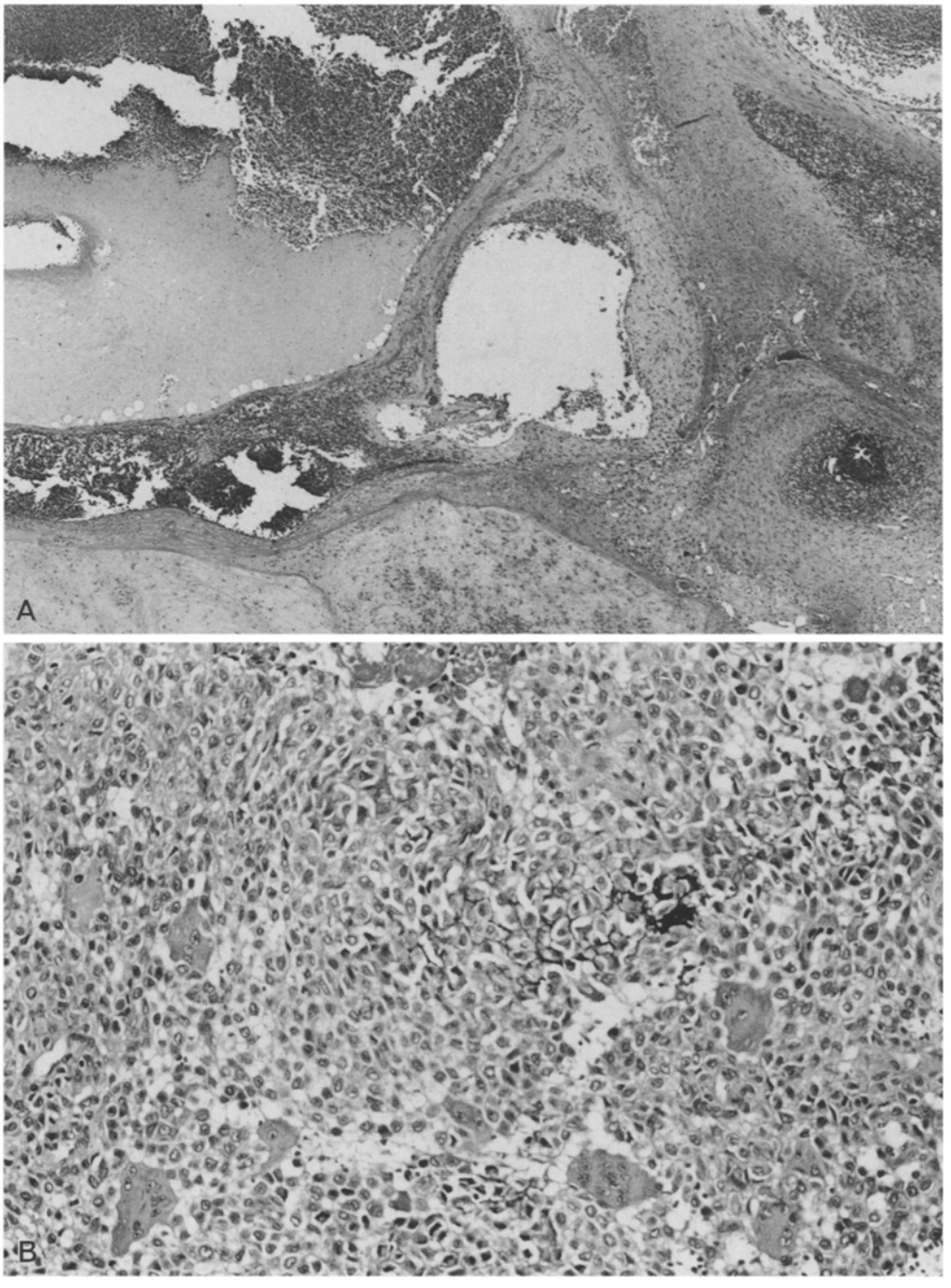


Fig. 6. A Photomicrograph (hematoxylin and eosin, ×40) showing large cystic space with loose fibrous septum. Solid tumor tissue is observed only in a small portion of the septum. **B** Photomicrograph (hematoxylin and eosin, ×100) showing proliferation of small, polygonal tumor cells. So-called "chicken-wire" calcifications are prominent in some areas, and osteoclastic-type giant cells are present

sive appearances on the roentgenogram. An adequate surgical approach is needed for thorough curettage of chondroblastoma in this location, as increased rate of recurrence at this site could be explained by inadequate curettage because of the difficulty in obtaining complete exposure of the lesion.

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