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Bone tumors in children and adolescents



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Benign tumours of the spine in children and adolescents

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INTRODUCTION

Benign tumours of the spine are rare in children and adolescents but the overall incidence is not negligible. The amount of benign tumours of spine arises to 3,5% of all forms of benign skeletal tumours. The orthopaedic surgeon is confronted with several problems as early diagnosis, treatment and late sequelae. Because of their rarity, it is arduous to acquire a large clinical experience for an orthopaedic surgeon during his career. Also in the literature there is a lack of clinical trials whereas the majority of papers are concerned with the management of each type of benign bone tumour (1-5). The only previous retrospective studies about benign spinal tumours in children are those reported by Fraser *et al* (6) and by Weinstein and McLeod (7). Fraser analysed a series of 40 children affected by benign and malign tumours. Weinstein and McLeod reported, instead, a series of 31 patients affected by a benign tumour of the spine, but ten patients were older than 18 with three cases of lethal evolution in two giant-cell tumours (GCT) and one sacral unresectable osteoblastoma (OB). Weinstein and McLeod did not find a significant relationship between the extent of initial excision and recurrence rate. No prospective work is reported as long as we know.

MATERIAL AND METHODS

Sixty-two children aged from 2 to 16 years, affected by tumours or tumour-like lesions of the vertebral column were collected from six Italian children's hospitals and retrospectively reviewed at a clinical and radiographic follow-up ranging from 0,5 to 30 years (average 5,7). A study was made of presentation, diagnosis, treatment and

skeletal complications. We included the types listed in the Table I. Neurinomata, lipomata and angioneuromata were not considered.

The conditions at presentation were: sex, age, site, early symptoms, presenting signs, duration of symptoms.

Diagnostic tools were those of imaging techniques (plain radiographs, serial tomography, radionuclide bone scan, computed tomography, myelography, MRI, arteriography), and histology (needle biopsy, biopsy-curettage, excision-biopsy).

The treatment was non-surgical (brace, cast, intralesional corticoids injection, embolization), surgical (curettage, intralesional excision, marginal resection), and combined (embolization plus surgery).

We also considered recurrences and complications.

Clinical and radiographic outcomes were evaluated as good, fair and poor according to our fo-

TABLE I. Benign tumours of the spine (age 2-16 years; follow-up 5,7 years)

Tumour type	N. of cases	%
Eosinophilic granuloma of the bone (EGB)	18	29
Aneurysmal bone cyst (ABC)	17	27
Osteoid osteoma (OO)	14	23
Osteoblastoma (OB)	07	11
Chondroma/osteochondroma (Ch)	3	5
Giant cell tumour (GCT)	1	2
Others (undetermined)	2	3
Total	62	100

Following criteria: **Good** (healing of the lesion, no pain, no deviation or functional deficit); **Fair** (healing of the lesion, no pain, scoliosis $\leq 10^\circ$, changing in sagittal curves $< 25\%$, no functional deficit); **Poor** (one or more of the following conditions: lack of or incomplete healing of the lesion, pain, scoliosis $> 10^\circ$, changing in sagittal curves $> 25\%$, functional deficit involving > 2 vertebrae, neurologic deficit).

RESULTS

Sex: The global ratio male/female was 1.4/1. A strong prevalence in males was observed in eosinophilic granuloma of bone (EGB) with a ratio 5/1 and a slight prevalence of females was found in aneurysmal bone cyst (ABC) with a ratio 0.7/1.

Age: The average age of the patients at the onset was 8.7 ranging from 2 years in EGB to 16 years in GCT. The 82% of them were older than 7 years.

The most frequent involved site was lumbar level (Table II).

In Table III the relative incidence of the presenting symptoms and signs is reported: 80% of the patients complained back or neck pain and almost 2/3 was having paravertebral contracture. A palpable mass was evident in ten cases (16%). Neurologic deficits were presents in 14% of the patients but only 3% had sphincter disturbances.

The mean duration of symptoms before diagnosis was seven months with a minimum/maximum delay of 2/24 months.

In the preoperative assessment plain radiographs were employed in all cases and in 20 cases the plain radiographs were the only diagnostic procedure. Technetium bone scan was used in 37% of cases and CT in 40%; MRI was employed in 12% of all cases and in the 23% of the patient firstly seen after 1987 (Figures 1 and 4). Arteriography was employed in five patients (8%) with selective embolization in four cases of ABC (6%).

Non-surgical treatment was performed in 12 cases (66%) of EGB and in two cases (12%) of ABC. In six cases (33%) of EGB surgical treatment consisted in curettage (four cases) and curettage-bone grafting (two cases). In 15 cases (88%) of ABC surgical treatment consisted in curettage (ten cases), curettage-bone grafting (one case), laminectomy and curettage (one case), partial sacrectomy (one case), emiarthrectomy (one case), resection of spinous process (one case). In four cases of ABC a selective arterial embolization was performed before surgical excision. Bone tumours sensu stricto

TABLE II. Site involvement

Spine level	N. of cases (%)	N. of vertebrae (%)
Lumbar	22 (35)	22 (32)
Thoracic	18 (29)	19 (28)
Sacral	12 (19)	15 (22)
Cervical	11 (17)	12 (18)

One case of triple combined localisation (ccl). Two cases of triple localisation in cervical and sacral level. Two cases of double localisation in thoracic and sacral level.

TABLE III. Incidence of presenting symptoms and signs

	Patients (n.)	Incidence (%)
Presenting symptoms		
Back or neck pain	50	80
Weakness	5	8
Sphincter disturbance	2	3
Presenting signs		
Scoliosis or torticollis	22	35
Mass	10	16
Muscle weakness	6	10
Paravertebral muscle spasm	40	64
Limp	7	11
Radicular impairment	16	25
Neurologic deficit	9	14

tu were all treated surgically. Six osteoblastoma (OB) were resected and only one curettage was performed in a sacral localization. Except one excised by curettage, OOb were always *en bloc* resected (Figure 2): laminectomy or emilaminectomy, arthrectomy, excision of spinous process.

The diagnosis was histologically confirmed in all surgical cases. However, no patients underwent incisional biopsy or needle biopsy or needle aspiration.

Two patients had major complications: the first with ABC presented an intraoperative bleeding, and later on underwent arterial embolization and mass resection. The second patient with EGB of D11 developed few months later a severe cyphosis that required a circumferential arthrodesis.

Recurrence was observed in three cases: two patients with ABC, who underwent arterial embolization

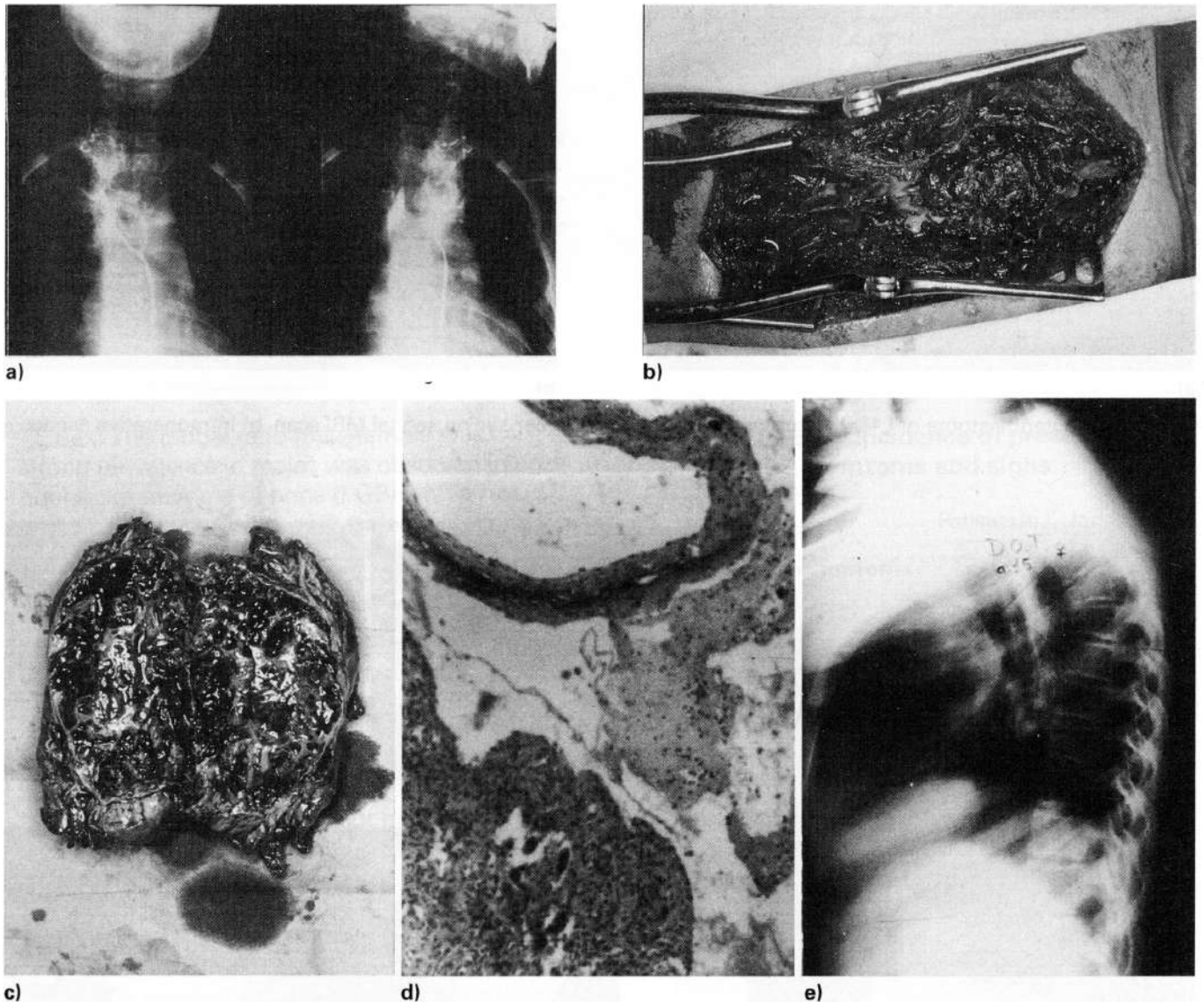


Figure 3. Aneurysmal bone cyst of T4. **a)** Arteriography and selective arterial embolization. **b)** Intraoperative aspect by posterior approach: mild bleeding. **c)** Cutting surface of the mass. **d)** Histologic view. **e)** Moderate cyphosis at eight years follow-up due to partial wedging of vertebral body.

TABLE IV. Results

Tumours	Good	Fair	Poor
EGB	15 (24%)	2 (3%)	1 (2%)
ABC	16 (25%)	1 (2%)	0 (0%)
OO	13 (21%)	0 (0%)	1 (2%)
OB	07 (11%)	0 (0%)	0 (0%)
CH/OC	03 (5%)	0 (0%)	0 (0%)
GCT	00 (00%)	0 (0%)	1 (2%)
Others	02 (3%)	0 (0%)	0 (0%)
Total	56 (89%)	3 (5%)	3 (6%)

third one with ABC of T4 developed ipercyphosis at eight years follow-up (Figure 3). Poor results were represented by one child with EGB surgically treated with severe cyphosis, the second one with OO presented persistent pain, in the third patient with GCT, the radicular deficit of S1 did not recover.

DISCUSSION

Benign tumours of the spine are very rare in children younger than 7. They occur in both ma-

les and females, but EGB is five times more frequent in boys and ABC demonstrates a slight prevalence for girls. Posterior arch of lumbar and thoracic level are the most frequent involved sites (1, 4, 6, 7) but ABC occurs often in the sacrum. Clinical features at presentation are non-specific and proper investigations are set-up tardively. Delayed diagnosis is also due to difficulties in the interpretation of the radiographic pictures of the spine in children. Only in a minority of cases, i.e. in vertebra plana by histiocytosis X and in OO when the typical nidus is recognized, conventional plain radiographs could be employed as the only imaging diagnostic procedure. Actually, conservative treatment is indicated in vertebra plana (3) whereas a surgical approach is recommended in all cases of OO (2, 4). In other cases further investigations are necessary: bone cortex or posterior wall are appreciated by CT, MRI is indicated in case of neurological involvement or in case of wide extension of the mass (Figures 1 and 4). When ABC is suspected, arteriography and selective arterial embolization are indicated as diagnostic and therapeutic procedure (8, 9) (Figure 3).

Even though we agree with the effectiveness of the needle aspiration/biopsy in the preliminary assessment of osteolytic bone lesion (10) and even it is now proved that in very rare cases vertebra plana may be caused by a malignant tumor (11, 12), our patient never underwent this diagnostic procedures. We can explain this observations as follows:

— Precise location of the lesion is too difficult in children's spine, even with the help of fluoroscopy.

— Needle aspiration or needle biopsy is unsafe about cervical spine.

— If an ABC is examined the specimen could be poor or insufficient for the diagnosis of certitude;

— Finally in all cases of our series laboratories data and radionuclide bone scan findings allow to exclude systemic disease or aggressive growth.

Furthermore when a very expansive, osteolytic lesion is found, ABC must be suspected. In these cases arteriographic picturing is pathognomonic and selective arterial embolization is demonstrated to be successful (8, 9). However definitive diagnosis was never mistaken in our patients.

Recovery of vertebral height is achieved in the majority of cases of vertebra plana and conserva-

tive treatment by a brace or plaster cast is recommended. In destructive forms surgical approach must be performed in order to excise the lesion, make histological diagnosis, decompress spinal cord. According to the literature (1, 2, 4), in our series there was a very low incidence of recurrences in OO and OB after block resection. We point out that wide resection is recommended, because it is difficult to localize the nidus of the tumor intraoperatively.

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