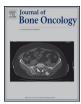
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Research Paper

Histological, epidemiological and anatomical analysis of 193 bone tumours of the scapula



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ABSTRACT

Backround: This study aimed to determine the frequency and distribution of bone tumours of the scapula as well as the histological and anatomical characteristics of these rare lesions in a large case series.

Methods: The records of all lesions of the scapula collected from 1975 to 2018 in our bone tumour registry and institute of pathology were evaluated. During these 43 years, 223 cases were identified. Analysis included assessment of age, gender, side, imaging findings, tumour location, and histological evaluation with the assignment of each lesion to one of the bone tumours according to the World Health Organization (WHO) classification of bone tumours.

Results: Bone tumours of the scapula were found in 193 cases. Mean patient age was 38.4 years (2.6–82.4). Most of the lesions were of cartilage origin (47%). 59 bone tumours were benign (30.6%), 29 were intermediate (15.0%), and 105 were malignant (54.4%). The most commonly found bone tumour was Osteochondroma (23.3%), followed by Chondrosarcoma (17.6%), Bone metastases (16.6%), Ewing sarcoma (8.8%), and Osteosarcoma (7.8%). The percentage of malignant bone tumours increased with increasing age. In patients >50 years of age, 91% had a malignant lesion of the scapula.

Conclusions: Evaluation of 193 bone tumours of the scapula revealed a high incidence of malignancy in this series, while increased patient age was identified as a potential risk factor for the development of a malignant lesion of the scapula. These findings highlight the importance of early diagnosis and treatment of suspicious lesions of the scapula to improve patient outcome.

1. Introduction

Bone tumours of the scapula are rare, although the shoulder girdle, including the proximal humerus, which is most frequently involved, the clavicle, the scapula, and the surrounding soft tissues is a relatively common site for bone and soft-tissue tumours [1-3]. In a large series of bone tumours including 566 patients, most of the lesions were found to originate from the femur (39.9%), the tibia (17.7%) and the humerus (11.8%), while the scapula was affected in 1.6% of cases only [2]. Due to the rarity of these lesions, there is a paucity of published literature [1,3,4-21]. Most of the articles are case reports or include a limited number of patients with a bone tumour of the scapula [1,3,4-21].

As tumour progression is usually associated with unspecific or delayed clinical symptoms, these lesions may be quite large at the time of diagnosis and may have extended to the surrounding tissue (rotator cuff, proximal humerus, chest wall, neurovascular structures) [10]. Along with the rarity of bone tumours of the scapula and the limited experience of most of the physicians in the management of these lesions, early diagnosis is challenging but required to improve patient outcome.

This study aimed to determine the frequency and distribution of bone tumours of the scapula as well as the histological, epidemiological, and anatomical characteristics of these lesions in a large case series. Patients who had surgery in case of suspected bone tumour of the

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scapula were included if the histological diagnosis could be definitely assigned to one of the bone tumours according to the World Health Organization (WHO) classification of bone tumours [18–20].

2. Methods

2.1. Patients characteristics and study design

The records of all lesions of the scapula collected from 1975 to 2018 in our bone tumour registry and institute of pathology were evaluated. During these 43 years, 223 bone lesions of the scapula were identified. They had been either collected by our institution or from consultation cases sent to our Bone Tumour Registry in case of suspected bone tumour of the scapula. Undecalcifed specimens were embedded in methyl-methacrylate and/or embedded in paraffin wax after EDTA-decalcification for histopathological analysis in each case. They were assessed by pathologists with special experience in orthopaedic oncology at our institution. Following data were evaluated for this study: age, gender, side, imaging findings, tumour location within the scapula, and histological analysis including dignity and type of bone tumour. Cases were included if they could be definitely assigned to one of the bone tumours according to the World Health Organization (WHO) classification of bone tumours [18-20]. Thirty cases did not meet the inclusion criteria and were excluded.

2.2. Statistical analysis

Descriptive statistics were given as mean (range) or percentage. Student *t*-tests were used to compare patient characteristics like age between the benign/intermediate and malignant tumour group. *p*-values < 0.05 denote statistical significance.

3. Results

The data of 193 patients with a bone tumour of the scapula were evaluated in this retrospective study. Mean patient age was 38.4 years (2.6–82.4). Age distribution analysis showed one age peak in the second decade of life (Fig. 1). 58% of the patients were male, and 42% were female, with a male to female ratio of 1.4:1.0. Side information was available in 89% of cases. The left scapula was involved in 53% and the right scapula in 47% of patients, respectively, yielding a left to right side ratio of 1.1:1.0. Tumour location within the scapula was recorded in 32% of cases; the acromion was most frequently involved (16.1%). Histological analysis revealed 22 different bone tumour types with 6 benign, 7 intermediate (locally aggressive and/or rarely metastasizing), and 9 malignant types, respectively, which were assigned to 10 different bone tumour type categories according to the World Health

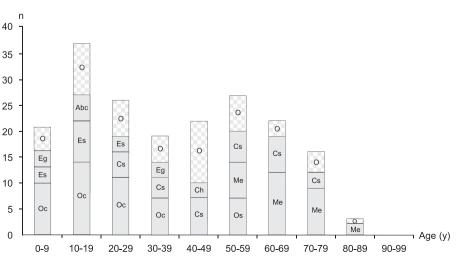


Table 1

Bone tumour type categories according to the WHO classification.

Bone tumour type category	Cases	Percent
Cartilage tumours	90	46.6
Myogenic, lipogenic, and epithelial tumours	33	17.1
Tumours of undefined neoplastic nature	24	12.4
Miscellaneous tumours	18	9.3
Osteogenic tumours	16	8.3
Fibrogenic tumours	4	2.1
Haematopoietic tumours	3	1.6
Vascular tumours	3	1.6
Osteoclastic giant cell rich tumours	1	0.5
Fibrohistiocytic tumors	1	0.5

Table 2

Benign bone tumours of the scapula.

Cases	Percent	Mean age (y)	Minimum age (y)	Maximum age (y)	Male/ female ratio
45	23.3	20.8	3.3	67.0	3.1:1.0
8	4.2	25.9	3.6	43.5	1.0:1.7
3	1.6	38.2	14.7	55.3	Male
1	0.5	6.8	6.8	6.8	Female
1	0.5	46.6	46.6	46.6	Male
1	0.5	77.9	77.9	77.9	Female
	45 8 3 1 1	45 23.3 8 4.2 3 1.6 1 0.5 1 0.5	45 23.3 20.8 8 4.2 25.9 3 1.6 38.2 1 0.5 6.8 1 0.5 46.6	45 23.3 20.8 3.3 8 4.2 25.9 3.6 3 1.6 38.2 14.7 1 0.5 6.8 6.8 1 0.5 46.6 46.6	age (y) age (y) age (y) age (y) age (y) 45 23.3 20.8 3.3 67.0 8 4.2 25.9 3.6 43.5 3 1.6 38.2 14.7 55.3 1 0.5 6.8 6.8 6.8 1 0.5 46.6 46.6 46.6

Organization (WHO) classification system (Table 1). Bone tumours were most often of cartilage origin in this series with a percentage of nearly 50% of all included cases. Metastases involving the scapula were identified in 16.6% of cases. They originated most commonly from lung cancer (8 cases), colorectal cancer (3 cases), prostate cancer (3 cases), and renal cancer (3 cases).

59 bone tumours of the scapula were benign (30.6%), 29 were intermediate (15.0%), and 105 were malignant (54.4%) (Table 2–4). The most commonly found bone tumour was Osteochondroma (23.3%) (Fig. 2), followed by Chondrosarcoma (17.6%) (Fig. 3), Bone metastases (16.6%), Ewing sarcoma (8.8%) (Fig. 4), and Osteosarcoma (7.8%) (Fig. 5).

Patient age was a mean of 24.8 years (2.6–77.9) in the benign/intermediate tumour group (male to female ratio of 1.6:1.0) as compared to 49.7 years (6.8–82.4) in the malignant tumour group (male to female ratio of 1.2:1.0). Excluding cases with bone metastases, patient age averaged 43.0 years (6.8–82.4) in the malignant tumour group (male to female ratio of 1.0:1.0). Analysis revealed that patient age was significantly higher in the malignant tumour group as compared to the

Fig. 1. *Age distribution.* Age distribution of bone tumours of the scapula including data of the commonest lesions per age group. *n*, number of patients; O, Others; Oc, Osteochondroma; Es, Ewing sarcoma; Eg, Eosinophilic granuloma; Abc, Aneurysmal bone cyst; Cs, Chondrosarcoma; Ch, Chondroma; Os, Osteosarcoma; Me, Metastases involving bone.

Table 3

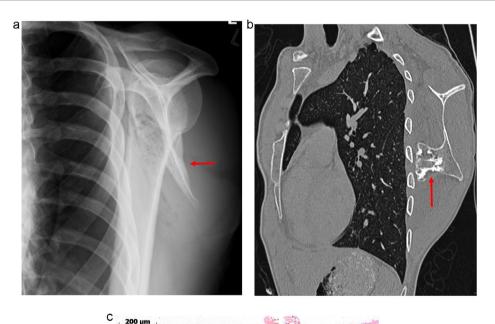
Intermediate bone tumours of the scapula.

Pathology	Cases	Percent	Mean age (y)	Minimum age (y)	Maximum age (y)	Male/female ratio
Eosinophilic granuloma	11	5.7	26.1	3.5	52.3	1.2:1.0
Aneurysmal bone cyst	9	4.7	25.1	10.9	57.0	1.0:2.0
Desmoplastic fibroma of bone	4	2.1	14.2	2.6	21.1	1.0:3.0
Chondromyxoid fibroma	2	1.0	46.7	43.4	49.9	1.0:1.0
Chondroblastoma	1	0.5	21.8	21.8	21.8	Male
Osteoblastoma	1	0.5	37.3	37.3	37.3	Male
Giant cell tumour of bone	1	0.5	69.5	69.5	69.5	Female

Table 4

Malignant bone tumours of the scapula.

Pathology	Cases	Percent	Mean age (y)	Minimum age (y)	Maximum age (y)	Male/female ratio
Chondrosarcoma	34	17.6	48.0	15.0	75.3	1.0:1.6
Bone metastases	32	16.6	65.2	5.4	80.4	1.9:1.0
Ewing sarcoma	17	8.8	20.5	6.8	52.5	3.3:1.0
Osteosarcoma	15	7.8	46.0	9.3	69.8	1.1:1.0
Angiosarcoma	2	1.0	50.7	49.6	51.7	Male
Plasma cell myeloma	2	1.0	76.1	75.0	77.2	Female
Malignant Lymphoma	1	0.5	73.2	73.2	73.2	Female
Undifferentiated high-grade pleomorphic sarcoma	1	0.5	82.4	82.4	82.4	Female
Liposarcoma of bone	1	0.5	56.2	56.2	56.2	Male



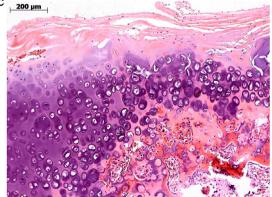


Fig. 2. Osteochondroma. Lateral radiograph of the left shoulder (Y-view) shows scapular winging in a 28 years old man (a). Computed tomography (CT) scan demonstrates a partially calcified bony lesion on the anterior surface of the scapula (b). Histological evaluation confirmed the suspected diagnosis of an Osteochondroma with the typical cartilaginous cap covered by periosteum (decalcified, stained with hematoxylin and eosin, $100 \times$ magnification) (c).

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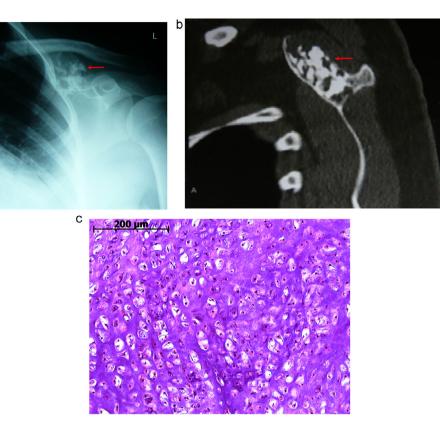


Fig. 3. Chondrosarcoma. Anteroposterior radiograph of the left shoulder shows a calcified bone tumour of the scapula in a 32 years old woman (a). CT scan demonstrates a partially calcified tumour in the supraspinous fossa (b). Histological assessment revealed Chondrosarcoma with atypical chondrocytes in a hyaline matrix (decalcified, stained with hematoxylin and eosin, $200 \times$ magnification) (c).

benign/intermediate tumour group (p < 0.001).

In young patients (0–29 years; 84 cases) Osteochondroma was the most frequently found bone tumour (41.7%), followed by Ewing sarcoma (16.7%), and Chondrosarcoma (8.3%). In old patients (50–79 years; 65 cases) Bone metastases were the commonest bone lesions (43.1%), followed by Chondrosarcoma (24.6%), and Osteosarcoma (10.8%).

4. Discussion

In one of the largest case series to date, we have reviewed 223 scapular lesions collected in our bone tumour registry and our institute of pathology over 43 years. Bone tumours of the scapula were found in 193 cases (86.5%) indicating a high incidence of these lesions in patients who had surgery in case of suspected bone tumour of the scapula. Most of the lesions were of cartilage origin, with a percentage of nearly 50% of all included cases. Osteochondroma and chondrosarcoma were the most common bone tumour types found in this study, both of which are cartilage tumours. These results indicate that the scapula, which is a flat bone, may be more prone to cartilage tumours as to other bone tumour types. It is reasonable to assume that this may be in part due to the fact that the scapula is formed by endochondral ossification with gradual replacement of cartilaginous tissue by bone tissue [13,22].

Histological analysis revealed a high incidence of malignancy (54.4%) in the scapula in our study. Four of the five most common bone tumour types were malignant. These findings are exceptional, considering that the incidence of benign/intermediate bone tumours is much higher than that of malignant bone tumours in the human skeleton [2]. Baena-Ocampo et al. [2] reported that 71.6% of the bone tumours were benign/intermediate, while 28.4% were malignant in their series of 566 patients. Previous studies focusing on bone tumours of the scapula found a high incidence of malignancy as well with a percentage of 54–67% [1,3,13]. It should be considered that benign bone tumours of the scapula may be underrepresented in these studies as well as in our series, because lesions of the scapula which were not

operated are missing.

The percentage of malignant bone tumours of the scapula increased with increasing age in our study from 23–32% in the first four decades of life (0–39 years) to 85–100% in the 6th–9th decade (50–89 years). In patients >50 years of age, 91% had a malignant lesion of the scapula (62 out of 68 tumours), while only 9% (6 out of 68 tumours) had a benign/intermediate lesion, yielding a malignant to benign/intermediate ratio of 10.3:1. These data indicate that increased age may be a risk factor for the development of a malignant bone tumour of the scapula. Khan et al. [3] found a correlation between increasing patient age and malignancy of scapular and periscapular lesions, but they included bone and soft-tissue tumours in their analysis (418 cases).

Osteochondroma was the commonest bone tumour of the scapula in our series (23% of all cases). About 51% of the benign/intermediate bone tumours were osteochondromas (45 out of 88 cases). Osteochondromas are frequently located on the anterior surface of the scapula and may lead to scapular winging, snapping sensation and pain [13]. Complete resection of symptomatic osteochondroma is recommended [13]. Khan et al. [3] found that osteochondroma was the most frequent benign bone tumour of the scapula as well with a percentage of 25% respecting all bony lesions [3].

Chondrosarcoma was the second most commonly found bone tumour in our series (17.6% of all cases), and the most prevalent malignant tumour of the scapula (32% of all malignant lesions). This is an exceptional finding, considering that Osteosarcoma is known to be the most common primary malignant bone tumour in humans [2,21]. Chondrosarcoma of the scapula is a rare entity; most of these malignant tumours were found to originate from other bones [23]. In a large case series including 194 chondrosarcomas diagnosed in Finland in 1971–1990, Söderstrom et al. [23] found that these lesions were most commonly located in the chest (21%), pelvis (21%), and femur (19%). In the upper extremity, the humerus was affected most commonly with 8% [23]. The scapula was rarely affected, with only 3% of all cases [23]. Previous studies with a limited number of patients (19 and 29 cases) reported that chondrosarcoma was the most common malignant

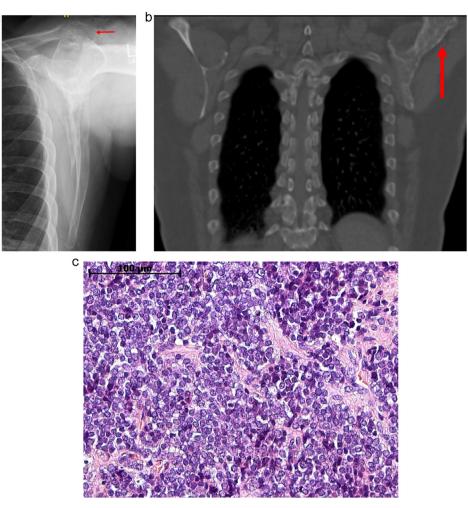


Fig. 4. *Ewing sarcoma.* Lateral radiograph of the left shoulder (Y-view) in a 52 years old man indicates permeative destruction of bone (a). CT scan confirms this type of destruction of the left scapula (b). Histological analysis revealed Ewing sarcoma with small blue round cells with finely dispersed chromatin and clear cytoplasm, arranged in a sheet-like growth pattern (decalcified, stained with hematoxylin and eosin, $400 \times$ magnification) (c).

bone tumour of the scapula as well [10,15]. Khan et al. [3] found chondrosarcoma in only 10% of all cases with a scapular lesion (including bone and soft tissue tumours), but it was the most common primary malignant bone tumour (45%) in their series [3]. Chondrosarcomas were common in the 3rd–7th decades of life in our study (85% of all chondrosarcomas). Histological analysis of all 34 chondrosarcomas of the scapula revealed 11 grade 1, 15 grade 2, and 6 grade 3 lesions with no grade documented for 2 cases in our study. The incidence of high grade chondrosarcoma (grade 3) was relatively low in our study, consistent with previous reports [14].

Bone metastases of the scapula were common (16.6% of all cases) and they were frequently found in old patients in our series. 94% of metastases involving the scapula occurred in the 6th–9th decade of life. They originated most commonly from the lung. In their analysis of scapular metastases Khan et al. [3] found lung, renal and adenocarcinoma of unknown origin to be the most common primary sites.

Ewing sarcoma was the fourth most common tumour (8.8% of all cases) and the second most common primary malignant tumour of the scapula in our study. Ewing sarcoma of the scapula is known to be a very rare entity [3,15]. 82% of Ewing sarcomas occurred in the first three decades of life with a peak in the second decade (47% of all Ewing sarcomas) in our series. 97% of ewing sarcoma were found in the first two decades of life in the study of Khan et al. [3]. Osteosarcoma was the fifth most common tumour of the scapula in our series with one age peak in the 6th decade of life (47% of all cases). Osteosarcomas usually show a bimodal distribution with peaks in the 2nd and 6th–7th decade

of life [3].

Several studies have reported on bone tumours in other flat bones, like the skull, pelvis, sternum, and ribs [24,25]. Bone metastases mainly from lung, breast, kidney and prostate cancer, as well as plasma cell myeloma are relatively common bone tumours of the ribs and sternum, while primary malignant bone tumours are rare [24,25]. The most frequent primary malignant tumour of the ribs and sternum is chondrosarcoma [24,25]. Less common primary malignant tumours are Ewing sarcoma and Osteosarcoma [24]. Other rare bone tumours of the ribs and sternum, which should be considered, include fibrous dysplasia, chondroma, aneurysmal bone cyst, and giant cell tumor [25]. In a series of 270 patients with pelvic bone tumours, who were treated by surgical resection, the authors found that chondrosarcoma was the most common bone tumour (149 cases), followed by Ewing sarcoma (40 cases), osteosarcoma (27 cases), benign bone tumours (25 cases), other malignant tumours (18 cases), and bone metastases (11 cases) [26].

The limitations of our series include the retrospective nature and preselection bias. Cases had been either collected by our institution or sent from selected pathologists and surgeons for second opinion evaluation in case of suspected bone tumour of the scapula. The strength of this series is the large number of included cases, considering the rarity of these lesions.

5. Conclusions

Evaluation of 193 bone tumours of the scapula revealed a high

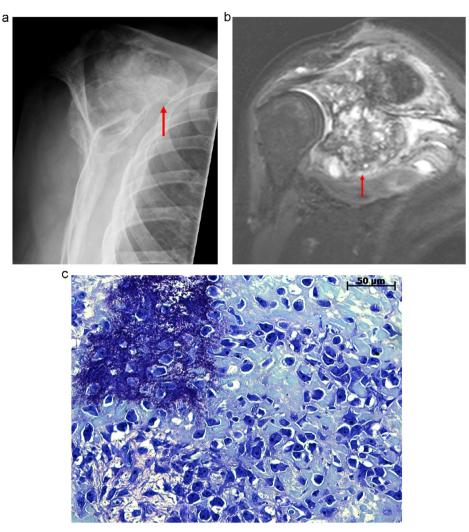


Fig. 5. *Osteosarcoma*. Lateral radiograph of the right shoulder in a 66 years old man shows a huge osteoblastic bone tumour of the scapula (a). Magnetic resonance imaging demonstrates a destructive lesion within the scapula (b). Histological analysis revealed pleomorphic epitheloid tumor cells with prominent nucleoli, directly associated with osteoid, resembling high-grade osteosarcoma (undecalcified, methyl-methyacrylate, stained with Toluidine Blue, $400 \times$ magnification) (c).

incidence of malignancy in this study, while increased patient age was identified as a potential risk factor for the development of a malignant lesion of the scapula. These findings highlight the importance of early diagnosis and treatment of bone tumours of the scapula to improve patient outcome. As tumour progression is usually associated with unspecific or delayed clinical symptoms and these lesions may not be adequately detected on plain radiographs, magnetic resonance imaging (or computed tomography) is recommended followed by biopsy in suspicious lesions of this rarely affected bone.

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CRediT authorship contribution statement

Matthias H. Priemel: Conceptualization, Data curation, Writing review & editing, Validation. Johannes M.E. Erler: Data curation, Formal analysis, Writing - review & editing, Validation. Jozef Zustin: Conceptualization, Data curation, Writing - review & editing, Validation. Andreas M. Luebke: Data curation, Writing - review & editing, Validation. Norbert Stiel: Data curation, Writing - review & editing, Validation. Alexander S. Spiro: Conceptualization, Data curation, Formal analysis, Writing - original draft, Validation.

Declaration of Competing Interest

The authors declare that there are no conflicts of interest.

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