Primary bone tumors and tumor like lesions of the ulna

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Objective: The aim of this study was to discuss the diagnosis and surgical management and their results according to stage of primary bone tumors at ulna and to share our experience on this exceptional location for bone tumors.

Methods: We have retrospectively reviewed our clinics database and identified 23 cases (14 males and 9 females, mean age was 28.9 (range 4–77)) with primary bone tumors and tumor like lesion involvement of ulna. The patients were evaluated according to complaints, type and grade of tumor, treatment, recurrence and functional status.

Results: The most common first referral complaint was constrictive pain in 52.1% of the cases, benign tumors and tumor like lesions of the bone constituted 73.9% whereas malignant bone tumors were 26.1%, 39.1% of the lesions were located in distal end of ulna and the mean follow up was 33.8 months (range 8–172 months). Local recurrence has unexpectedly occurred in 3 benign lesions (13.1%).

Conclusion: Benign bone lesions tend to involve distal and proximal ends, malign bone lesions involve diaphysis mostly. Both benign and malignant diaphyseal lesions of the ulna have better postoperative results regarding the lesions at both ends of ulna. One should also take care of recurrences even after a decade from the primary surgery.

Level of evidence: Level IV, Therapeutic study

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Lesions in postoperative long-term follow-up. Musculoskeletal Tumor Society Scoring for upper extremity (6th months and annually in malignant lesions. Working abilities of annual controls after 2 years postoperatively. X-rays are evaluated followed as; 2 weeks, 6 weeks, once in 3 months for 2 years and annual controls for 5 years postoperatively. Malignant tumors were followed by a standard follow-up protocol of tumors. Patients were monitored regarding severity of the tumors. Revealed diagnosis and Enneking grading of bone and soft tissue performed by interpretation of radiology, histopathologically classified was applied for upper extremity lesions in postoperative long-term follow-up.

### Results

There were 14 male and 9 female patients with the mean age of 28.9 (range 4–77) at admittance. Lesions were located on the right side in 13 cases (56.5%) and on the left side in 10 cases (43.5%). 17 of 23 patients had lesions on dominant side. The most common first referral complaint was constructive pain in 12 of 23 cases (52.1%) in both benign and malignant lesions followed by swelling (21.7%) and deformity (17.3%). Lesions were also encountered incidentally (8.9%). In malignant lesions of our cases pain was the leading complaint. 39.1% of the lesions were located in distal end of ulna whereas, remaining lesions were evenly located in both proximal and diaphyseal regions. Most of the swelling complaints were related with distal involvement however diaphyseal lesions referred with pain mostly. Mean follow up was 33.8 months (range 8–172 months). Patients were called for a final follow-up and evaluation. 2 of the 23 cases were deceased because of grade IIB and III osteosarcoma at 9th and 12th months respectively.

Benign tumors and tumor like lesions of the bone constituted 73.9% whereas malignant bone tumors were 26.1%. Regarding tumor like lesions of bone; 5 cases were aneurysmal bone cyst (ABC) (21.7%), 2 were unicameral bone cysts (8.6%) and a case was intraosseous ganglion cyst (4.3%). For benign bone tumors, 5 patients had osteochondroma (21.7%), 2 had enchondroma (8.6%) and 2 had giant cell tumor (GCT) of bone (8.6%). Considering malignant involvement; 3 cases had chondrosarcoma (13%), 2 cases had osteosarcoma (8.6%) and a case had Ewing sarcoma (4.3%).

According to radiologic and histopathologic assessment, Enneking classification of bone and soft tissue tumors is applied for each lesion. 2 cases were stage 1, 9 lesions were stage 2, 6 lesions were stage 3, 2 lesions were stage IA, a lesion was IB, 2 lesions were IIB and a lesion was stage III. In our series, benign lesions were seen to have slight tendency to affect distal (47.1%) and proximal (26.1%) regions rather than diaphyseal (17.8%) involvement. Except for 2 deceased patients, who had a proximal and a distal involvement of osteosarcoma respectively, all malignant lesions (3 chondrosarcomas and a Ewing sarcoma) were diaphyseally located.

Intralesional excision was the most common treatment method performed in 10 patients (43.4%) of our study, followed by marginal resections in 8 patients (34.8%), wide excision in 3 patients (13.1%) and amputation in 2 patients (8.7%). ABC was the most common indication for intralesional excision constituting 5 of 12 cases (41.7%). Marginal resection was performed for osteochondroma mostly (62.5%). The only malignant tumor that performed marginal excision was grade IB chondrosarcoma. Wide resection was performed for 2 chondrosarcomas and a Ewing sarcoma (Fig. 1). Amputations were performed exceptionally for 2 grade IIB and III osteosarcomas.

When working and household abilities are asked to the patients, they have declared the result as whether satisfactory or limited in their last follow up. Mean MSTS score was 21.5 ± 5.9. The best

### Table 1

Demographical data of the patients in this study. Abbreviations used to indicate tumor types; ABC: Aneurysmal bone cyst, GCT: Giant cell tumor of bone, UBC: Unicameral bone cyst, CS: Chondrosarcoma, OC: Osteochondroma, IOGC: Intraosseous ganglion cyst, EWS: Ewing sarcoma, EC: Enchondroma, OS: Osteosarcoma.

| Patient No. | Age | Gender | Complaint | Location | Tumor type | Enneking Stage | Treatment | Follow-up time (mo) | MSTS score | Recurrence | Management of recurrence |
|-------------|-----|--------|-----------|----------|------------|---------------|-----------|---------------------|-------------|-------------|------------------------|
| 1           | 11  | Male   | Swelling  | Proximal | ABC        | 3             | Curettage, Grafting | 14        | 8            | N/A         | N/A         |
| 2           | 27  | Female | Swelling  | Distal   | GCT of Bone| 3             | Curettage, Grafting | 172       | 9            | 14th year    | Resection   |
| 3           | 4   | Female | Swelling  | Distal   | ABC        | 2             | Curettage, Grafting | 52        | 11           | N/A         | N/A         |
| 4           | 28  | Male   | Pain      | Proximal | UBC        | 2             | Curettage, Grafting | 19        | 12           | N/A         | N/A         |
| 5           | 64  | Male   | Pain      | Mid-diaphysis | CS | IB | Curettage, Grafting | 50        | 16           | N/A         | N/A         |
| 6           | 16  | Male   | Deformity | Distal   | OC         | 3             | Resection | 30        | 17           | N/A         | N/A         |
| 7           | 40  | Female | Pain      | Mid-diaphysis | CS | IA | Curettage, Grafting | 31        | 18           | N/A         | N/A         |
| 8           | 6   | Male   | Deformity | Distal   | OC         | 3             | Resection, Vascularized Fibula | 125       | 19           | 10th year    | Resection   |
| 9           | 56  | Female | Pain      | Distal   | ABC        | 2             | Curettage, Grafting | 16        | 22           | N/A         | N/A         |
| 10          | 4   | Male   | Pain      | Proximal | ABC        | 3             | Curettage, Grafting | 36        | 23           | N/A         | N/A         |
| 11          | 24  | Male   | Swelling  | Distal   | GCT of Bone| 2             | Curettage, Grafting | 12        | 24           | N/A         | N/A         |
| 12          | 30  | Male   | Pain      | Mid-diaphysis | CS | IA | Curettage, Grafting | 32        | 25           | N/A         | N/A         |
| 13          | 42  | Male   | Incidental| Distal   | IOGC       | 1             | Curettage | 10        | 26           | N/A         | N/A         |
| 14          | 77  | Female | Incidental| Proximal | UBC        | 1             | Curettage, Grafting | 8         | 26           | N/A         | N/A         |
| 15          | 53  | Female | Deformity | Proximal | OC         | 2             | Resection | 16        | 26           | N/A         | N/A         |
| 16          | 13  | Male   | Deformity | Mid-diaphysis | OC | 2 | Resection | 16        | 27           | N/A         | N/A         |
| 17          | 22  | Male   | Deformity | Mid-diaphysis | EWS | IIB | Resection, Vascularized Fibula | 15        | 27           | N/A         | N/A         |
| 18          | 13  | Male   | Pain      | Proximal | ABC        | 3             | Curettage, Grafting | 23        | 27           | 15th month   | Curettage, Grafting |
| 19          | 18  | Female | Swelling  | Distal   | OC         | 2             | Resection | 48        | 29           | N/A         | N/A         |
| 20          | 13  | Male   | Pain      | Mid-diaphysis | EC | Curettage, Grafting | 20        | 30           | N/A         | N/A         |
| 21          | 26  | Female | Pain      | Mid-diaphysis | EC | 2 | Curettage, Grafting | 13        | 30           | N/A         | N/A         |
| 22          | 57  | Male   | Pain      | Distal   | OS         | III           | Amputation | 9         | NA           | N/A         | N/A         |
| 23          | 21  | Female | Pain      | Proximal | OS         | III           | Amputation | 12        | NA           | N/A         | N/A         |
Fig. 1. A 24-year-old case vignette from our series, who admitted with constricting pain in right forearm. A) Anteroposterior and lateral views of the case was not specifically diagnostic. B) Magnetic resonance images of the case demonstrated a diaphyseal lesion with soft tissue involvement showing low signal intensity on T1 sequences and high signal intensity on T2 sequences. C) Following standard Ewing sarcoma chemotherapy protocol, the patient was applied resection and reconstructed by vascularized fibula.
primary ulna bone malignancies indicate that most of the lesions originate from either proximal or distal ends.\textsuperscript{1,5,13} The malignant bone tumors of the ulna in our series were commonly originated from the diaphysis (4 in 6 cases, 66.7\%) unlikely.

In the recent literature aggressive benign and malignant lesions are reported mostly.\textsuperscript{5–13} Our series indicated 9 of 17 benign lesions as Enneking 2 lesions. And malignancies were mostly low grade (3 of 6 lesions were Enneking IA or IB). Curettage and allograft packing was the most common treatment method in benign tumors and tumor like lesions of ulna in accordance with the literature. However in malignancies; en-bloc resection and vascularized fibular autograft technique was the choice of treatment commonly (3 of 6 lesions). Amputation was performed in 2 high grade, late stage osteosarcoma patients who we consequently lost during follow up period because of lung metastases.

Recurrence rates depend mostly on histopathological type, grade and treatment method of lesions. The local recurrence rates of ABC were reported to be 17–37\%.\textsuperscript{10} The risk of local recurrence is relatively higher in giant cell tumors of distal ulna.\textsuperscript{10} Poor prognostic results for recurrent giant cell tumors are associated with pathologic fractures, subchondral lesions and malignant transformation.\textsuperscript{14} Higher recurrence rates are reported with curettage, cauterization and grafting or cementation when compared to resections in GCT of bone.\textsuperscript{20} Our case recurred at 14th year after the primary treatment. The local recurrence of solitary osteochondroma is most commonly associated with inadequate resection of perichondrial ring in skeletal immature patients.\textsuperscript{21}

In conclusion, primary benign or malignant bone tumors of ulna are extremely rare. Due to its close relation with major neurovascular vascular structures intraoperative care should be taken. One should also take care of recurrences even after a decade from the primary surgery. Except giant cell tumors of bone, benign aggressive tumors of the ulna can be managed by curettage and allograft packing, whereas malignant tumors require wide or en bloc resections and reconstructions or amputations regarding histopathologic grade and stage. Benign bone lesions tend to involve distal and proximal ends, whereas malignant bone lesions involve diaphysis mostly. Both benign and malignant diaphyseal lesions of the ulna have better postoperative results when compared to the lesions at both ends of ulna.

Conflict of interest

The authors of this study do confirm no conflict of interest.

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