

RESEARCH PAPER

An Epidemiological Study of Primary Bone Tumors of the Fibula

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Background: Relatively low incidence has led to an under-rating of fibula tumors. This study aimed to evaluate fibula tumors as a whole and to give detailed information based on histological types according to the anatomic location of the tumors in the fibula.

Methods: Evaluation was made of all the primary bone tumors of the fibula recorded in our bone tumor registry and institute of pathology from 2007 to 2018. Of these, 62 cases were identified. Analysis included assessment of age, gender, tumor localization, the presenting symptoms, the duration of symptoms, and treatment methods.

Results: There were 48 (77.4%) benign and 14 (22.6%) malignant tumors. The most commonly found benign tumors were non-ossifying fibroma (12/48; 25%) and aneurysmal bone cyst (12/48; 25%), and the malignant tumors were chondrosarcoma (3/14; 21.4%) and chondroblastic osteosarcoma (3/14; 21.4%). The most common location for both benign and malignant tumors (58.3%, 71.4%) are the proximal fibula, followed by the distal fibula (27.1%, 28.6%) and the diaphysis (14.6%, 0%). Six (9.7%) patients presented with pathological fibula fractures.

Conclusion: Fibular tumors are rarely encountered in clinical practice but are mostly benign, with malignancy determined in approximately a quarter of patients. However, as most benign tumors are asymptomatic, and therefore remain undetected, the actual proportion of malignant tumors will be much lower.

Level of evidence: IV

Keywords: Benign, Distribution, Fibula, Malignant, Primary bone tumors, Tumors

Introduction

Primary fibular bone tumors are uncommon, constituting only 2.5% of all primary bone tumors (1). Although, most primary bone tumors of the fibula are benign, considerable morbidity and mortality are caused by local aggressive, or malignant tumors (1). The most common benign tumors in this region are osteochondroma, enchondroma and aneurysmal bone cyst (ABC), while the most common malignant tumors are chondrosarcoma, osteosarcoma and Ewing's sarcoma (2). Proximal tumors are seen more often than diaphyseal and distal tumors (3).

Previous epidemiological studies have provided information on primary bone tumors including those of the scapula, clavicle, and patella (4-6). However, there have been few studies providing detailed information on

primary bone tumors of the fibula (2, 7, 8). In the field of orthopedic oncology, most studies of the fibula have been related to the use of the fibula as an autograft (9-11). Besides the great value as an autograft, the fibula is important for the knee and ankle kinematics (12, 13). The aim of this study was to evaluate fibula tumors as a whole and to give information according to the anatomic location of the tumors in the fibula.

Materials and Methods

The protocol for this retrospective study was approved by the Local Ethics Committee. The Department of Pathology database was examined to detect all cases involving the fibula between January 2007 and December 2018. After excluding soft tissue tumors, metastases, tumor-like

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lesions, patients with previously known neoplasms and had another possible primary site outside the fibula, 62 primary bone tumors of the fibula were included in this study. While the patients were not specifically invited for the study, the hospital charts, radiographs, and pathology records of each patient were analyzed.

The data of the demographic features, including age, gender, tumor localization, the presenting symptoms, the duration of symptoms, and treatment methods were evaluated. All the radiodiagnostic images of the patients were carefully re-evaluated and fibula tumors were examined in three localizations as proximal third, middle third (diaphysis), and distal third of the fibula. All analyses were descriptive. Patient data were summarized as mean±standard deviation (SD), minimum, maximum values, number (n) and percentage (%).

Results

The 62 patients comprised 29 (46.8%) females and 33 (53.2%) males with a median age at diagnosis of 25.6 ± 17.6 years (range, 8–73 years). Benign tumors were determined in 48 (77.4%) cases and malignancies in 14 (22.6%) [Table 1]. There was a slight predominance of

males in the study sample. Localization was right-side in 26 (41.9%) patients and left-side in 36 (58.1%).

The distribution of primary bone tumors of the fibula according to age groups and localizations is given in Table 1. Almost a quarter of all the lesions were malignant. The mean age was determined as 23.2 ± 15.7 years in patients with benign lesions, and 34 ± 21.5 years in those with malignant tumors. Both pain and swelling were the most common presenting symptoms.

Of the 62 patients, 6 (9.7%) patients had pathological fractures, 5 of which were non-ossifying fibroma and the other was ABC. Details of the pathological fractures are given in Table 2.

Of the benign tumors, 61.3% were followed up conservatively, 33.9% were surgically treated, and 4.8% were treated with percutaneous radiofrequency ablation. Of the intermediate tumors (ABC and giant cell tumor), all of them were surgically treated including intralesional resection was applied to 12 (80%) patients and partial fibulectomy to 3 (20%). All the malignant tumors were treated with partial fibulectomy.

Table 1. Distribution of benign and malignant primary bone tumors of fibula according to histologic types, locations and age groups

Diagnosis	Patients (n)											
	Proximal				Diaphyseal				Distal			
Location												
Age	12>	12-21	21-51	51<	12>	12-21	21-51	51<	12>	12-21	21-51	51<
Benign (n=48)	7	8	12	1	1	3	2	1	4	6	1	2
Non-ossifying fibroma		7					2			3		
Aneurysmal bone cyst		7					1			4		
Osteochondroma		4					1					
Intraosseous ganglion		4								1		
Fibrous dysplasia		1					1			2		
Osteoid osteoma		1					2					
Giant cell tumor		3										
Enchondroma		1								1		
Osteoblastoma										1		
Simple bone cyst										1		
Malignant (n=14)		4	4	2						2	1	1
Chondroblastic osteosarcoma		3										
Chondrosarcoma		3										
Ewing sarcoma										2		
Osteoblastic osteosarcoma		1										
Clear cell chondrosarcoma		1										
Fibroblastic osteosarcoma		1										
Undifferentiated pleomorphic sarcoma		1										
Adamantinoma										1		
Primary bone lymphoma										1		
Total (n=60)	7	12	16	3	1	3	2	1	4	8	2	3
	38					7			17			

Table 2. Features of pathological fractures due to primary bone tumors of fibula

	Age	Gender	Localization	Diagnosis	Management
1	9	F	Distal	Non-ossifying fibroma	Conservatively followed-up
2	11	M	Diaphyseal	Non-ossifying fibroma	Conservatively followed-up
3	11	F	Distal	Non-ossifying fibroma	Conservatively followed-up
4	13	M	Proximal	Non-ossifying fibroma	Conservatively followed-up
5	13	F	Proximal	Non-ossifying fibroma	Conservatively followed-up
6	17	M	Diaphyseal	Aneurysmal bone cyst	Partial fibulectomy

Discussion

The leading findings of this study were that the most common localization of fibula tumors is the proximal third, and that 26.3% of proximal tumors and 23.5% of distal tumors were malignant. The most prevalent bone tumors of the fibula were non-ossifying fibroma, ABC, osteochondroma, and intraosseous ganglion in the benign group, and chondrosarcoma and chondroblastic osteosarcoma in the malignant group. These results were similar to the findings of some previous reports (14-16).

Abdel et al reported that benign proximal fibula tumors are rare (14). In contrast, benign tumors were more common than malignant tumors in the current series. Like many other studies in the literature, the patients included were only those who had undergone bone biopsy from the fibula or had a definite diagnosis from radiodiagnostic images. It has been reported that benign primary bone tumors of the fibula are reported less than malignant tumors, which could be attributed to the fact that that benign lesions do not require bone

biopsy for diagnosis and are followed-up conservatively, so no pathology sampling is performed (14, 17, 18). In the current study, the higher number of benign tumors was due to the high number of biopsies, pathological fractures and radiological diagnoses. The reason for the higher number of biopsies was that the benign lesions appeared more aggressive because of the gracile nature of the fibula (15).

In previous studies, both male and female predominance has been reported (2, 8). In the current study, there was a slight male predominance in the benign tumor group and no gender predominance in the malignant tumor group. However, there was male predominance in intermediate primary fibula tumors.

In the present study, there were no malignant tumors in patients aged <12 years, while 42.8% of the tumors in patients aged >51 years were malignant, and malignant tumors comprised approximately 25% of the tumors in patients between the ages of 12 and 51 years. In

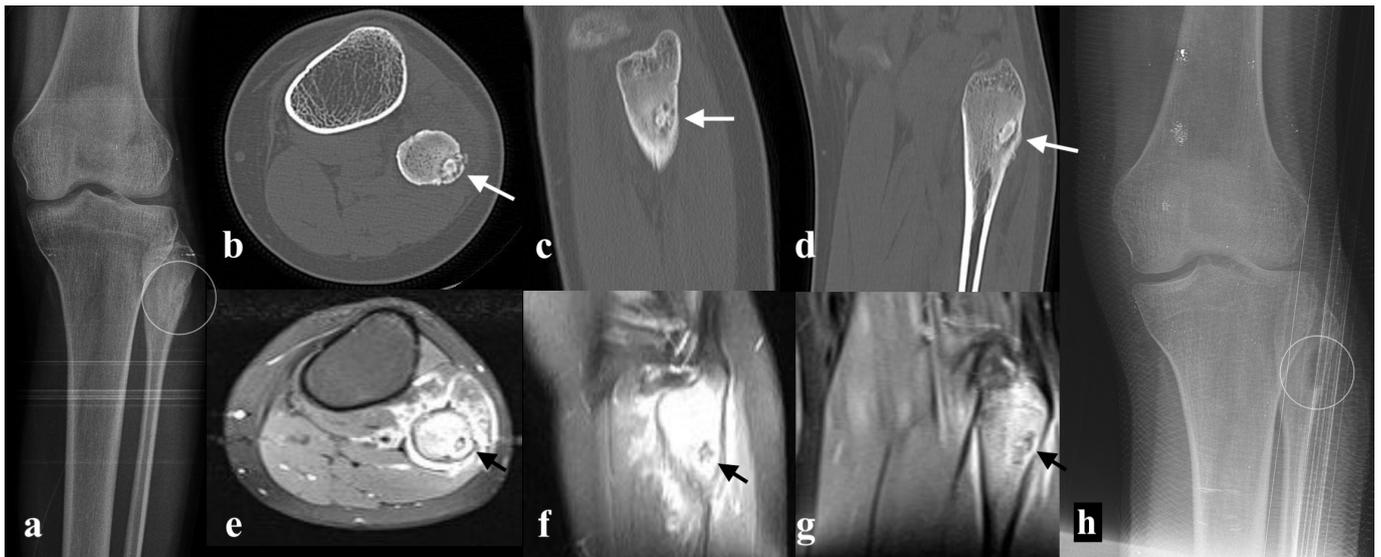


Figure 1. A 18-year-old female patient with osteoid osteoma of the proximal fibula. (a) Preoperative X-ray findings show focal sclerosis and periosteal reaction (the lesion inside the circle). (b, c, d) Axial, sagittal and coronal CT images showing an ovoid medullary-based radiolucent lesion of the proximal fibula measuring 14×9 mm in diameter (white arrow). (e, f, g) Axial, sagittal and coronal MRI images showed that the lesion has central calcifications and is associated with moderate peripheral osteosclerosis and marked periosteal reaction (black arrow). (h) Postoperative X-ray findings 2 years later (the lesion inside the circle).

Table 3. Features of pathological fractures due to primary bone tumors of fibula

	Age	Gender	Localization	Diagnosis	Management
1	9	F	Distal	Histiocytic fibroma	Conservatively followed-up
2	11	M	Diaphyseal	Histiocytic fibroma	Conservatively followed-up
3	11	F	Distal	Histiocytic fibroma	Conservatively followed-up
4	13	M	Proximal	Histiocytic fibroma	Conservatively followed-up
5	13	F	Proximal	Histiocytic fibroma	Conservatively followed-up
6	17	M	Diaphyseal	Aneurysmal bone cyst	Partial fibulectomy

contrast, a previous study in literature examining fibula tumors as a whole reported malignant tumors at the rate of 6.5% under the age of 12 years, 24.5% between the ages of 12 and 51 years, and 19.2% over 51 years (2). Increased patient age might be a potential risk factor for the development of a malignant tumor of the fibula.

The localization of tumors in the fibula may cause different presentations (8, 14, 19). Patients may present with swelling, pain, peroneal nerve symptoms, or pathological fractures. In the present study, the most common symptoms were pain and swelling, as in previous studies. (2, 19) Pathological fractures in the current study were seen in benign tumors and in young-active patients. Although pathological fractures diagnosed with non-ossifying fibroma were detected at different localizations in the fibula, these patients were followed up conservatively without any intervention or immobilization. Since the lesions of the patients with ABC were located in the diaphysis, and the resection of this tumor in the diaphysis might cause minimal morbidity, this made the decision for resection easier.

Previous studies and this present study showed that the proximal fibula is by far the most common localization for bone tumors of the fibula (2, 8). Furthermore, 58.3% of the benign, and 71.4% of the malignant tumors involved the proximal fibula [Figure 1]. In order of the most common benign tumors of the proximal fibula were non-ossifying fibroma, ABC, osteochondroma, and intraosseous ganglion. The most common malignancies were chondrosarcoma and chondroblastic osteosarcoma.

Involvement of the distal fibula was seen in 28.9% of the benign, and 28.6% of the malignant tumors [Figure 2]. The most common benign tumors of the distal fibula were ABC and non-ossifying fibroma.

The least common location was the diaphysis with 15.5% of the benign tumors. Osteoid osteoma, and non-ossifying fibroma were the most common tumors of the diaphysis. There were no malignant tumors in this location.

To the best of our knowledge, there has been only one study to date that has analyzed fibula tumors as a whole (2). From a scan of the literature, it can be seen that there have been examination of proximal fibula tumors, and case reports of distal fibula tumors (17-21). Arikan et al reported that the most common benign and malignant tumors were osteochondroma and chondrosarcoma, respectively (2). However, in the current study, the most common benign tumor was non-

ossifying fibroma and ABC, and malignant tumors were chondrosarcoma and chondroblastic osteosarcoma. The reason for this variety in the incidence might be associated with the relatively low number of patients in the current study.

Fibulectomy may be required especially in malignant tumors (16, 22). Local aggressive benign tumors such as ABC and GCT may often require intralesional resection, or occasionally partial fibulectomy (21, 23, 24). However, almost all benign tumors can be followed-up without surgical treatment(18), even if they have pathological fractures. In addition, osteoid osteoma and osteblastoma require percutaneous or open surgical treatment.

The present study is a relatively small epidemiological series about fibula tumors. The main limitation of this study was the lack of clinical and functional results. However, as fibula tumors are rare, this case series can be considered to contribute to the literature.

In conclusion, fibular tumors are rarely encountered in clinical practice, and they are mostly benign tumors.



Figure 2. A 24-year-old male patient with an Ewing sarcoma of the distal fibula. A partial fibulectomy of the distal fibula was necessary. (a) Preoperative X-ray findings show osteolytic lesion with sunburst appearance. (b) Postoperative X-ray findings 2 years later.

Approximately a quarter of the patients have malignant tumors. As most benign tumors are asymptomatic, they can remain undetected and the exact number of benign tumors is undoubtedly more than the number of cases in clinical practice. The vast majority of fibula tumors are seen in the proximal third. In the current study, the most common benign tumors were non-ossifying fibroma and ABC, and the most common malignant tumors were chondroblastic osteosarcoma and chondrosarcoma.

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References

- Unni KK, Inwards CY. Dahlin's bone tumors: general aspects and data on 10,165 cases: Lippincott Williams & Wilkins; 2010.
- Arikan Y, Misir A, Ozer D, Kizkapan TB, Yildiz KI, Saygili MS, et al. The incidence and distribution of primary fibula tumors and tumor-like lesions: A 35-year experience. J Orthop Surg (Hong Kong). 2018;26(3):2309499018798180.
- Zeytoonjian T, Mankin HJ, Gebhardt MC, Hornicek FJ. Distal lower extremity sarcomas: frequency of occurrence and patient survival rate. Foot Ankle Int. 2004;25(5):325-30.
- Song M, Zhang Z, Wu Y, Ma K, Lu M. Primary tumors of the patella. World J Surg Oncol. 2015;13:163.
- Kaiser CL, Yeung CM, Raskin K, Gebhardt MC, Anderson ME, Lozano-Calderon SA. Tumors of the scapula: A retrospective analysis identifying predictors of malignancy. Surg Oncol. 2019;32:18-22.
- Ren K, Wu S, Shi X, Zhao J, Liu X. Primary clavicle tumors and tumorous lesions: a review of 206 cases in East Asia. Arch Orthop Trauma Surg. 2012;132(6):883-9.
- Franchi A. Epidemiology and classification of bone tumors. Clin Cases Miner Bone Metab. 2012;9(2):92-5.
- Picci P, Manfrini M, Fabbri N, Gambarotti M, Vanel D. Atlas of musculoskeletal tumors and tumorlike lesions: the Rizzoli case archive: Springer Science & Business Media; 2014.
- Luo S, Jiang T, Yang X, Yang Y, Zhao J. Treatment of tumor-like lesions in the femoral neck using free nonvascularized fibular autografts in pediatric patients before epiphyseal closure. J Int Med Res. 2019;47(2):823-35.
- Vicenti G, Maruccia M, Carrozzo M, Elia R, Giudice G, Moretti B. Free vascularized osteoseptocutaneous fibular flap for radius shaft nonunion: The final solution when the iliac crest autograft fails. A case report. Injury. 2018;49 Suppl 4:S63-s70.
- Stevenson JD, Doxey R, Abudu A, Parry M, Evans S, Peart F, et al. Vascularized fibular epiphyseal transfer for proximal humeral reconstruction in children with a primary sarcoma of bone. Bone Joint J. 2018;100-b(4):535-41.
- Bozkurt M, Yavuzer G, Tonuk E, Kentel B. Dynamic function of the fibula. Gait analysis evaluation of three different parts of the shank after fibulectomy: proximal, middle and distal. Arch Orthop Trauma Surg. 2005;125(10):713-20.
- Akdoğan M, Ateş Y. Ayak bileği ve distal tibia anatomisi. TOTBİD Dergisi. 2016(15):158-65.
- Abdel MP, Papagelopoulos PJ, Morrey ME, Wenger DE, Rose PS, Sim FH. Surgical management of 121 benign proximal fibula tumors. Clin Orthop Relat Res. 2010;468(11):3056-62.
- Hansford BG, Smith ZC, Stacy GS. Imaging of Benign Fibular Tumours and Their Mimics. Can Assoc Radiol J. 2018;69(3):293-302.
- Perisano C, Marzetti E, Spinelli MS, Graci C, Fabbri C, Maffulli N, et al. Clinical management and surgical treatment of distal fibular tumours: a case series and review of the literature. Int Orthop. 2012;36(9):1907-13.
- Guo C, Zhang X, Gao F, Wang L, Sun T. Surgical management of proximal fibular tumors: risk factors for recurrence and complications. J Int Med Res. 2018;46(5):1884-92.
- Sun T, Wang L, Guo C, Zhang G, Hu W. Symptoms and signs associated with benign and malignant proximal fibular tumors: a clinicopathological analysis of 52 cases. World J Surg Oncol. 2017;15(1):92-.
- Jamshidi K, Mazhar FN, Masdari Z. Reconstruction of distal fibula with osteoarticular allograft after tumor resection. Foot Ankle Surg. 2013;19(1):31-5.
- Arikan Y, Misir A, Gur V, Kizkapan TB, Dincel YM, Akman YE. Clinical and radiologic outcomes following resection of primary proximal fibula tumors: Proximal fibula resection outcomes. J Orthop Surg (Hong Kong). 2019;27(2):2309499019837411.
- Bhowmick K, Boopalan P. Saving the ankle in distal fibular giant cell tumour - A case report. J Clin Orthop Trauma. 2019;10(6):1054-8.
- Monson DK, Vojdani S, Dean TJ, Louis-Ugbo J. Lateral

- ankle stabilization after distal fibular resection using a novel approach: a surgical technique. Clin Orthop Relat Res. 2014;472(4):1262-70.
23. Natarajan M, Paraskumar M, Rajkumar G, Sivaseelam A, Natarajan S. Limb salvage in aggressive and malignant

- tumours of the fibula. Int Orthop. 2004;28(5):307-10.
24. Erler K, Demiralp B, Ozdemir MT, Basbozkurt M. Treatment of proximal fibular tumors with en bloc resection. Knee. 2004;11(6):489-96.