

## **ORIGINAL ARTICLE**

# Characterization of primary fibular bone tumors and post-surgical clinical and functional outcomes

Mohamed Ali Bekir, MD<sup>1</sup><sup>(b)</sup>, Coskun Ulucakoy, MD<sup>2</sup><sup>(b)</sup>, Ismail Burak Atalay, MD<sup>2</sup><sup>(b)</sup>, Aras Pervane, MD<sup>2</sup><sup>(b)</sup>, Guray Togral, MD<sup>2</sup><sup>(b)</sup>, Bedii Safak Gungor, MD<sup>2</sup><sup>(b)</sup>

<sup>1</sup>Department of Orthopaedics and Traumatology, Dr. Nafiz Korez Sincan City Hospital, Ankara, Türkiye <sup>2</sup>Department of Orthopaedics and Traumatology, Dr. Abdurrahman Yurtaslan Ankara Oncology Training and Research Hospital, Ankara, Türkiye

The fibula is a rare location for tumors, and only 2.5% of all benign and malignant bone tumors are located in the fibula.<sup>[1]</sup> Diagnosis may be delayed due to knee and ankle pain. The most common presenting complaints are pain, palpable mass, and more rarely, peroneal nerve symptoms.<sup>[2]</sup>

Treatment of benign tumors located in the fibula is primarily follow-up and conservative. Surgical treatment options are curettage or excision.<sup>[2]</sup> In malignant and borderline tumors, *en-bloc* resection is in the foreground. For the proximal fibula, Malawer described two types of *en-bloc* resection: Type I, marginal resection, includes 2 to 3 cm of the diaphysis of the proximal fibula and resection of the muscles around the fibula, while in Type II, wide extra-compartment resection, proximal fibula,

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Correspondence: Coskun Ulucakoy, MD. Dr. Abdurrahman Yurtaslan Ankara Onkoloji Eğitim ve Araştırma Hastanesi, Ortopedi ve Travmatoloji Kliniği, 06200 Yenimahalle, Ankara, Türkiye.

E-mail: coskunulucakoy@gmail.com

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#### ABSTRACT

**Objectives:** In this study, we present the characterization of tumors located in the fibula, which is a rare site for tumors, and the clinical and functional results of patients with fibular tumors that we operated in our clinic.

**Patients and methods:** Between February 2008 and December 2018, a total of 104 patients (58 males, 46 females; mean age:  $26\pm18$  years; range, 3 to 78 years) who were operated for tumor located in the fibula were retrospectively analyzed. Demographic data, histopathological diagnosis, tumor localization, surgical method and postoperative complications, metastases, follow-up time, and functional results of the patients were recorded. The fibula was divided into three groups as proximal, diaphysis and distal one-third.

**Results:** The mean follow-up was  $56\pm25.3$  (range, 24 to 108) months. Eighty (76.9%), seven (6.7%) and 17 (16.3%) patients were located proximal, diaphysis and distal to the fibula, respectively. The number of patients with malignant tumors located in the fibula was 26 (25%), the tumor was benign in 54 (51.9%) patients and 24 (23%) tumors were borderline. The most common tumor localized to the fibula was osteochondroma with 28 (26.9%) patients. The most common malignant tumors located in the fibula were Ewing sarcoma and osteosarcoma in 11 patients each. The most common surgical method was curettage in 44 (42.3%) patients. Distant organ metastases were present in 18 (17.3%) patients. The Musculoskeleteal Tumor Society score of 17 patients who underwent resection of the proximal fibula were 87%.

**Conclusion:** Fibula is a rare location for tumors and fibula tumors tend to be located more proximal. Tumors located in the fibula have good functional outcomes after surgery.

Keywords: Fibula, Malawer, outcome, resection, tumor.

5 to 6 cm diaphysis, anterior and lateral muscle compartments, anterior tibial artery and peroneal nerve should be resected. In Type I resection, the peroneal nerve is preserved.<sup>[3]</sup>

In the present study, we aimed to present the characterization of tumors located in the fibula, which is a rare localization area for tumors, and the follow-up results of patients with fibular tumors operated in our clinic.

### **PATIENTS AND METHODS**

This single-center, retrospective study was conducted at Department of Orthopaedics and Traumatology, Dr. Abdurrahman Yurtaslan Ankara Oncology Training and Research Hospital between February 2008 and December 2018. A total of 104 patients (58 males, 46 females; mean age: 26±18 years; range, 3 to 78 years) who were operated for tumor located in the fibula were included. Demographic data, histopathological diagnosis, tumor localization, surgical method, postoperative complications, metastases, follow-up time, and functional results of the patients were recorded. Tumors involving the fibula and the tibia, metastases, and tumors with a follow-up period of less than 24 months were excluded from the study. The fibula was divided into three groups as proximal, diaphysis and distal one-third.

#### Statistical analysis

Statistical analysis was performed using the IBM SPSS version 22.0 software (IBM Corp., Armonk, NY, USA). Continuous variables were expressed in mean  $\pm$  standard deviation (SD) or median (min-max), while

categorical variables were expressed in number and frequency. Descriptive analyses were presented using percentile and quantitative values.

### **RESULTS**

The mean follow-up was 56±25.3 (range, 24 to 108) months. Eighty (76.9%), seven (6.7%), and 17 (16.3%) patients were located proximal, diaphysis and distal to the fibula, respectively (Table I). While the number of patients with malignant tumors located in the fibula was 26 (25%), the tumor was benign in 54 (51.9%) patients and 24 (23%) tumors were borderline (Table II). The most common tumor localized to the fibula was osteochondroma with 28 (26.9%) patients. This was followed by giant cell tumors with 15 (14.4%) patients. The most common malignant tumors located in the fibula were Ewing sarcoma and osteosarcoma in 11 patients each. Considering the frequency of tumors by fibula location, they were proximal, diaphysis and distal osteochondroma (31%), Ewing sarcoma (42%), and giant cell tumor (29%), respectively (Table III).

Curettage was the most common surgical method for tumors located in the fibula in 44 (42%) patients. These were followed by total excision (28%), resection (23%) and amputation (5%), respectively (Table I). Resection was performed in 17 (21%) of 80 patients with tumors located proximal to the fibula. While 12 of them (70%) were Malawer Type I, five of them were Malawer Type II (Table IV). Distant organ

TABLE I										
Frequency of fibular tumors according to location, type and surgical methods										
	n	%	Mean±SD	Median	Min-Max					
Age (year)			26±18		3-78					
Follow-up period (month)				56	24-108					
Location										
Proximal	80	76.9								
Diaphysis	7	6.7								
Distal	17	16.3								
Туре										
Benign	54	52								
Borderline	18	16								
Malignant	26	24								
Surgical method										
Curettage	44	43								
Total excision	30	29								
Resection	24	23								
Amputation	6	5								
SD: Standard deviation.										

TABLE II   Frequency of fibular tumors according to location, type									
	Benign Borderline		erline	Malig	gnant	Total			
Fibula	n	%	n	%	n	%	n	%	
Proximal	46	44	15	14	19	18	80	77	
Diaphysis	2	2	1	1	4	4	7	7	
Distal	6	6	8	8	3	3	17	16	
Total	54	58	24	23	26	25	104	100	

TABLE III   Frequency of fibular tumors subtypes according to location									
	Proximal		Diaphysis		Distal		Total		
Fibula	n	%	n	%	n	%	n	%	
Osteochondroma	25	24	1	1	2	2	28	27	
Giant cell tumor	9	8	1	1	5	5	15	14	
Osteosarcoma	9	8	0	0	2	2	11	10	
Ewing sarcoma	8	8	3	3	0	0	11	11	
Non ossifying fibroma	9	8	0	0	1	1	10	9	
Aneurysmal bone cyst	6	6	0	0	3	3	9	9	
Enchondroma	7	7	0	0	0	0	7	7	
Chondrosarcoma	2	2	1	1	1	1	4	4	
Simple bone cyst	2	2	0	0	2	2	4	4	
Osteoid osteoma	1	1	0	0	1	1	2	2	
Other	2	2	1	1	0	0	3	3	
Total	80	77	7	7	17	16	104	100	

TABLE IV												
Preferred surgical methods in tumors located proximal to the fibula (%)												
Cure	ttage	Total e	al excision Amputation				Resection				Total	
						Malawer Type I		Malawer Type II				
n	%	n	%	n	%	n	%	n	%	n	%	
32	40	26	33	5	6	12	70	5	30	80	100	

metastases were present in 18 (17.3%) patients. The most common metastasis was to the lung in 10 (55.5%) patients. Of the 26 malignant tumors, 61% were older than 21 years of age. Peroneal nerve palsy was observed in five (6%) patients located proximal to the fibula.

An 18-year-old male patient was consulted to our clinic with the complaint of knee pain in his left knee lasting for four months. A cystic lesion was detected in the head of the fibula in the direct X-ray of the patient (Figure 1a, b). *En-bloc* resection (Figure 2a, b) was performed in the patient whose Tru-cut biopsy

revealed an aneurysmal bone cyst. The result of the mass sent to pathology (Figure 3a, b) was compatible with aneurysmal bone cyst. No neurological deficit was detected in the postoperative patient.

#### DISCUSSION

The literature indicates that tumors located in the fibula are frequently located proximal.<sup>[4,5]</sup> Gümüştaş et al.<sup>[6]</sup> in their epidemiological study on tumors located in the fibula, the most common sites were proximal fibula (61%), distal fibula (27%), and diaphysis (11%). Similarly, in the study of Arikan et al.,<sup>[4]</sup> the most





FIGURE 2. Postoperative X-ray image after *en-bloc* resection of the proximal fibula. (a) Anteroposterior and (b) lateral view.



FIGURE 3. Intraoperative image during *en-bloc* resection of proximal fibula. (a) Peroneal nerve, (b) excised tumor.

common localizations were proximal (67%), distal (25%) and diaphysis (8%). In our study, similar to the literature, the most common location was the proximal fibula (76.9%). This was followed by distal (16.3%) and diaphysis (6.7%), respectively.

Although tumors located in the fibula are rarely encountered, the rate of malignant tumors in the literature on the malignancy rate reaches 50%.<sup>[1]</sup> In a study including 52 patients, Guo et al.<sup>[7]</sup> reported that 15.4% of all tumors located in the fibula were malignant. In the study of Arikan et al.[4] including 264 patients, this rate was 20.8%. In our study, 24% of 104 tumors localized to the fibula were malignant and 23% were borderline. We believe that the reason for the high rates of malignant and borderline in our study is that patients with suspected malignancy are referred to us more, as we are a reference oncology hospital. In addition, as another reason, we included only the patients we operated on. Many benign tumors such as non-ossifying fibroma were excluded from the study, since they were followed conservatively.

Abdel et al.<sup>[8]</sup> had a mean age of 24.3±9.6 (range, 2 to 64) years in patients who operated for a tumor located in the fibula and followed for an average of 88 months. In the study of Guo et al.,<sup>[7]</sup> the mean age of 52 patients followed for 31 months was 26.5±5.2 (range, 4 to 72) years. Gümüştaş et al.<sup>[6]</sup> had a mean age of 25.6±17.6 years in their study, and they found that increasing patient age may be a potential risk factor for the development of malignant fibular tumor. Similarly, Arikan et al.<sup>[4]</sup> reported that fibular tumors were more common under 21 years of age and that most fibular tumors under 21 years of age were benign. In this study, the mean age of 104 patients we followed for an average of  $56\pm25.3$  months was  $26\pm18$  years, and 61%of 26 malignant tumors were older than 21 years.

In a study performed in Mayo Clinic, the most common benign tumor located in the proximal fibula was osteochondromas (38%).<sup>[8]</sup> The most common benign tumors in the diaphyseal and distal involvement were osteoid osteoma and aneurysmal bone cyst, respectively.<sup>[6]</sup> In the study of Abdel et al.,<sup>[9]</sup> the most common malignant tumor located in the proximal fibula was osteosarcoma, while in the study of Arikan et al.,<sup>[4]</sup> the most common malignant tumors were chondrosarcoma (29.1%), Ewing sarcoma (27.3%) and osteosarcoma (27.3%). Similarly, in the study of Guo et al.,<sup>[7]</sup> the most common benign tumor located in the proximal fibula was osteochondroma, and malignant tumor osteosarcoma. Atalay et al.<sup>[10]</sup> reported the results of proximal fibula tumors, and the most common malignant tumor was osteosarcoma. Similarly, in this study, the most common tumor was osteochondroma (26.9%), while the most common malignant tumors were Ewing sarcoma and osteosarcoma (10%) with 11 patients each.

Function after surgery for proximal fibular tumors varies depending on the resection performed and the tumor type. The Musculoskeleteal Tumor Society scores of benign and malignant proximal fibular tumors treated surgically are available in the literature. Dieckmann et al.[11] reported the mean MSTS scores of 90 to 97% for 57 patients who were operated for benign and malignant proximal fibular tumors. Similarly, Inatani et al.<sup>[12]</sup> reported that, in a smaller number of patients (n=12), the mean MSTS score was 96% (range, 70 and 100%) in those whose peroneal nerve could not be resected, and 65% (range, 60 and 100%) in those who were resected. 70) has been reported. Kundu et al.<sup>[13]</sup> on the other hand, in the study in which they presented the results of 46 patients, they reported MSTS as 88%. Similarly, in this study, the MSTS score of 17 patients who underwent post-tumor resection in the proximal fibula was 87% (range, 70 and 100%). The most common complication after proximal fibula resection is peroneal nerve palsy and instability in the knee. Peroneal nerve palsy is seen at a rate of 3 to 57% in the literature. 3,14 Guo et al.<sup>[7]</sup> reported peroneal nerve palsy as 13.5% in 52 patients they operated. A total of 7.7% of these persisted as permanent peroneal nerve palsy. Guo et al.<sup>[7]</sup> did not report knee instability in any of the patients they operated. While knee instability was not encountered in this study, peroneal nerve palsy was seen in nine (11%) patients located proximal to the fibula. Of these, five patients (patients who underwent Malaver type II resection) continued as permanent peroneal nerve palsy.

To the best of our knowledge, only two studies have analyzed fibular tumors as a whole so far.<sup>[4,6]</sup> In these studies, the epidemiology was emphasized. The strength of our study is that it is the first study to analyze fibular tumors as a whole and to share the oncology experience (clinical and functional outcome) of a single center with this large number of patients. However, the lack of homogeneous groups due to its retrospective nature and considering all tumors together are the main limitations.

In conclusion, the fibula is a rare location for tumors and fibula tumors tend to be located more proximal. Tumors located in the fibula have good functional outcomes after surgery.

**Ethics Committee Approval:** The study protocol was approved by the Dr. Abdurrahman Yurtaslan Ankara Oncology Training and Research Hospital Ethics Committee (number 2021-06/1151). The study was conducted in accordance with the principles of the Declaration of Helsinki.

**Patient Consent for Publication:** A written informed consent was obtained from each patient.

**Data Sharing Statement:** The data that support the findings of this study are available from the corresponding author upon reasonable request.

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#### REFERENCES

- 1. Unni KK, Inwards CY. Dahlin's bone tumors: General aspects and data on 10,165 cases. 6th ed. Philadelphia: Lippincott Williams & Wilkins 2010. p. 1-9.
- Karakoç Y, Ulucaköy C. Management and retrospective analysis of tumors and tumor-like lesions localized in the talus. Jt Dis Relat Surg 2021;32:218-23.
- 3. Malawer MM. Surgical management of aggressive and malignant tumors of the proximal fibula. Clin Orthop Relat Res 1984;(186):172-81.
- 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;26:2309499018798180.
- Zeytoonjian T, Mankin HJ, Gebhardt MC, Hornicek FJ. Distal lower extremity sarcomas: Frequency of occurrence and patient survival rate. Foot Ankle Int 2004;25:325-30.
- Gümüştaş SA, Çevik HB, Kayahan S. An epidemiological study of primary bone tumors of the fibula. Arch Bone Jt Surg 2021;9:548-53.
- 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;46:1884-92.
- 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;468:3056-62.
- 9. Abdel MP, Papagelopoulos PJ, Morrey ME, Inwards CY, Wenger DE, Rose PS, et al. Malignant proximal fibular

tumors: Surgical management of 112 cases. J Bone Joint Surg [Am] 2012;94:e165.

- Atalay İB, Yılmaz S, Korkmaz İ, Ekşioğlu MF, Güngör BŞ. Surgical management of primary malignant proximal fibular tumors: Functional and clinical outcomes of 23 patients. Eklem Hastalik Cerrahisi 2019;30:24-31.
- 11. Dieckmann R, Gebert C, Streitbürger A, Henrichs MP, Dirksen U, Rödl R, et al. Proximal fibula resection in the treatment of bone tumours. Int Orthop 2011;35:1689-94.
- Inatani H, Yamamoto N, Hayashi K, Kimura H, Takeuchi A, Miwa S, et al. Surgical management of proximal fibular tumors: A report of 12 cases. J Bone Oncol 2016;5:163-6.
- 13. Kundu ZS, Tanwar M, Rana P, Sen R. Fibulectomy for primary proximal fibular bone tumors: A functional and clinical outcome in 46 patients. Indian J Orthop 2018;52:3-9.
- 14. Erler K, Demiralp B, Ozdemir MT, Basbozkurt M. Treatment of proximal fibular tumors with en bloc resection. Knee 2004;11:489-96.