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Epidemiology of Surgically Treated Foot and Ankle Bone Tumors: A Systematic Review

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Abstract

Background: Bone tumors of the foot and ankle account for 1% to 2% of all skeletal tumors. Our review aimed to determine their distribution according to histologic type, anatomical site, affected bone, patient sex, and age group.

Methods: We reviewed institutional epidemiologic studies focusing on surgically treated bone tumors of the foot and ankle, following the latest classification of the World Health Organization (2020). Our selection process adhered to the PRISMA guidelines. We provide a descriptive synthesis of the results from the included studies, along with subgroup analyses.

Results: We included 20 studies, totaling 2541 tumors: 1810 (71.2%) benign, 279 (11.0%) intermediate, and 452 (17.8%) malignant. The most frequent benign tumors were enchondroma, osteoid osteoma, and simple bone cysts. Among intermediate tumors, giant cell tumor of bone and osteoblastoma were most common, whereas chondrosarcoma, Ewing sarcoma, and osteosarcoma were the most frequent malignant tumors. The hindfoot was the most affected segment, followed by the forefoot, ankle, and midfoot. The calcaneus was the bone most affected by both benign and malignant tumors, whereas the metatarsals were the most frequent site for intermediate tumors. There was a slight predominance in men (56.8%). Benign tumors were more prevalent in individuals $<$20 years of age, whereas intermediate and malignant tumors were more common between the ages of 20 and 59 years. All tumors in individuals aged 60 years and older were malignant.

Conclusion: In surgically treated foot and ankle bone tumors, benign lesions predominated, malignancies comprised roughly 1 in 5, and the hindfoot, particularly the calcaneus, was most frequently involved. These patterns can guide differential diagnosis and operative planning, but heterogeneity and sparse demographic data (especially age) limit precision and preclude population-level estimates.

Keywords: foot, ankle, bone, bone neoplasms, epidemiology, systematic review

Introduction

Bone tumors located in the foot and ankle are uncommon,[1,4,7,13,15,20,26,27] accounting for 1% to 2% of all skeletal tumors.[27] Interestingly, their occurrence is proportionally higher compared with other anatomical segments, because the foot and ankle represent only about 3% of the total body mass.[15,20,33,34]

Bone tumors of the foot and ankle can affect people of all ages. Primary benign tumors are most often seen in individuals up to the fourth decade of life, whereas primary malignant tumors are more common in older adults. In addition, these tumors seem to occur more frequently in men,[5,16] although gender is not a strong indicator of the specific diagnosis.[27]

The thin layer of soft tissue over the foot and ankle facilitates the clinical diagnosis of bone tumors,[4,15,25-27,34] making even small tumors noticeable.[5] However, these tumors can sometimes be misdiagnosed.[25,26] This is usually because they are not considered true tumors, resulting in an underestimation of their potential severity.[15]

Orthopaedic surgeons managing patients with bone tumors of the foot and ankle must possess a comprehensive understanding of diagnostic and staging criteria,[9-12] especially in

cases where an intermediate or malignant tumor is suspected. Furthermore, the unique anatomical challenges of the ankle further complicate limb-sparing surgery, particularly in achieving sufficient surgical margins.[1,4,18,25]

Given the rarity and distinct clinical presentation of bone tumors affecting the foot and ankle, epidemiologic data on surgically treated cases remain limited. Therefore, the purpose of this systematic review was to bridge this knowledge gap by characterizing the distribution of surgically treated bone tumors of the foot and ankle according to histologic type, anatomical site (forefoot, midfoot, hindfoot, and ankle), affected bone, patient sex, and age group.

Methods

This review was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement.[19,28] The study protocol was registered with the International Prospective Register of Systematic Reviews (PROSPERO).

This systematic review was exempt from research ethics committee approval as it did not involve private information from participants or any potential violation of human rights.

Search strategy

We searched MEDLINE/PubMed, Embase, and Latin American and Caribbean Health Sciences Literature (LILACS) databases for articles published from database inception to May 31, 2025. The search strategy combined MeSH keywords, Emtree terms, and uncontrolled terms using the Boolean operators "OR" and "AND." The complete search strategy is provided in Supplemental Table 1. We also scanned the reference lists of the included studies to detect potentially relevant studies not identified by the previous search strategies.

Eligibility Criteria

Studies eligible for inclusion in this review were institutional epidemiologic studies published in English, Portuguese, or Spanish that focused on surgically treated bone tumors of the foot and ankle and enrolled patients of any age, sex, or ethnicity. Case reports and case series that focused on specific histologic types of bone tumors or did not represent the overall range of bone tumors occurring in the foot and ankle as well as studies published in languages other than English, Portuguese, or Spanish were excluded.

Screening and Data Extraction

One reviewer exported the results of the database searches into Rayyan Qatar Computing Research Institute (Rayyan QCRI) software and removed duplicates. Two reviewers independently screened titles and abstracts and then assessed the full texts of all potential studies for inclusion. Any disagreements between reviewers were resolved through discussion. If a consensus was not reached, a third independent reviewer was consulted.

To ensure unbiased data extraction, 2 reviewers independently extracted data using a standardized Excel spreadsheet (Microsoft Corporation, 2019). Data extracted included histologic type, updated according to the 2020 World Health Organization (WHO) Classification of Tumors—Soft Tissue and Bone Tumors,[35] anatomical site (forefoot, midfoot, hindfoot, and ankle), affected bone, patient sex, and age group ($<$20 years, 20-59 years, and $\geq$60 years). All tumors were reclassified according to this classification system, which allowed us to categorize tumors based on their biological potential as benign, intermediate, or malignant. This reclassification was based on the histologic terminology and diagnostic details provided in each study.

We carefully assessed potential overlapping cohorts from the same institutions by comparing author groups, study periods, centers, and patient characteristics. If multiple

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studies originated from the same center with overlapping data collection periods, we retained the study with the most comprehensive data set or the largest sample size and excluded the others to avoid double-counting. No significant overlap was detected among the included studies.

When information from potential studies was missing, insufficient, or vague, we attempted to contact the corresponding authors via email to obtain the necessary data. Ultimately, studies were excluded if the relevant data could not be obtained through this process.

Risk of Bias (Quality) Assessment

For the selected studies, 2 reviewers independently assessed the risk of bias using the tool developed by Murad et al.[22] This tool, based on the Newcastle-Ottawa Scale (NOS), was designed to assess the methodologic quality of case series and case reports and synthesize their results. It comprises 8 questions across 4 domains: selection (Q1), ascertainment (Q2 and Q3), causality (Q4-Q7), and reporting (Q8). We excluded 3 questions from this tool that were not applicable to our review: "Were other alternative causes that may explain the observation ruled out?" (Q4), "Was there a challenge/rechallenge phenomenon?" (Q5), and "Was there a dose-response effect?" (Q6).

Additionally, to more appropriately evaluate studies reporting prevalence estimates, we applied the Joanna Briggs Institute (JBI) Critical Appraisal Checklist for Studies Reporting Prevalence Data.[21] This tool consists of 9 questions across key domains of epidemiologic validity: Q1, Was the sample frame appropriate to address the target population? Q2, Were study participants sampled in an appropriate way? Q3, Was the sample size adequate? Q4, Were the study subjects and the setting described in detail? Q5, Was the data analysis conducted with sufficient coverage of the identified sample? Q6, Were valid methods used for the identification of the condition? Q7, Was the condition measured in a standard, reliable way for all participants? Q8, Was there appropriate statistical analysis? and Q9, Was the response rate adequate, and if not, was the low response rate managed appropriately? All included studies were reassessed using this checklist.

Each domain was rated as "Yes," "No," "Unclear," or "Not applicable." An unclear risk of bias was defined as a plausible bias for which insufficient information was available to permit a definitive judgement about its potential effect on the study outcomes. Discrepancies between reviewers were resolved by discussion and consensus.

Data Analysis

We provide a descriptive synthesis of the results from the included studies, expressed as absolute and relative frequencies. Additionally, we conducted structured subgroup analyses based on histologic type, anatomical site, the specific bone affected, patient sex, and age group. Because of the inability to subset for homogeneity, a meta-analysis was not feasible.

Percentages were calculated based on specific denominators defined according to the nature of each variable. For variables related to tumor location and diagnosis type, the denominator was the total number of tumors with available data in each study, which allowed for proportional calculations according to specific categories (eg, anatomical site or histologic type). For demographic variables, such as sex and age, the denominator was the total number of patients, rather than tumors, because multiple tumors could be associated with a single individual.

In data synthesis, we handled missing information based on the completeness of the data provided in the included studies. When data were complete and clearly reported, they were extracted and fully incorporated into the analysis. If only partial data were reported, partial extraction was performed to the extent that there was consistency and methodologic certainty. In cases where extraction was not possible because of a lack of clear information or an inadequate structure for analysis, the data were excluded from the corresponding pooled analysis, without attempting statistical imputation to avoid introducing bias or distortion in the calculated percentages.

All analyses were organized in Excel spreadsheets (Microsoft Corporation, 2019) and reviewed by 2 independent reviewers. This process was implemented to ensure the transparency and reproducibility of all calculations.

Results

The initial search identified 8781 records, 7035 of which remained after removing duplicates. Following title and abstract screening, 169 studies (2.4%) were considered potentially eligible and were sought for full-text retrieval. The full texts of 3 potentially eligible studies could not be retrieved, resulting in 166 studies being retrieved for full-text review. Of these, 18 (0.25%) met the inclusion criteria. Another 2 full-text articles identified through citation searching were deemed eligible, bringing the total number of studies included in this review to 20.[1-6,14,15,17,18,23-27,29-32,34] The study selection process is detailed in Figure 1.

Characteristics of Included Studies

Table 1 provides a summary of the included studies, outlining the characteristics of each study sample. All studies included in this review were retrospective, with follow-up periods ranging from 9 to 84 years, encompassing a total of 2541 bone tumors of the foot and ankle.

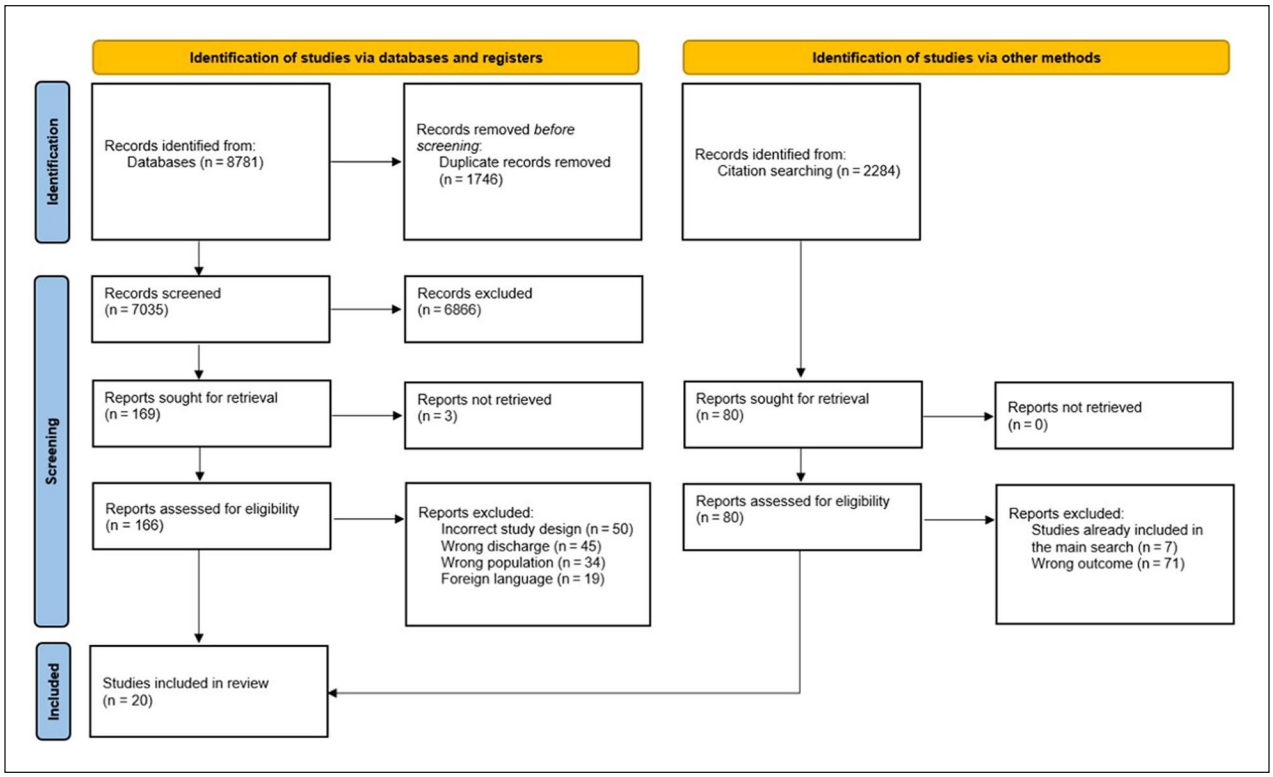

Figure 1. Flow diagram of study selection. The search was run on June 26, 2025.

Risk of Bias (Quality) Assessment

The findings from our risk of bias assessment, using the tool developed by Murad et al,[22] are shown in Figure 2. For the domain of selection (Q1), our analysis indicated an unclear risk of bias in 20% (4/20) of the included studies. For the domain of ascertainment (Q2 and Q3), we observed a low risk of bias across all the studies. In the domain of causality (Q4-Q7), questions Q4, Q5, and Q6 were not applicable to our review as they focused on adverse drug events. For the remaining question in this domain (Q7), all studies demonstrated a low risk of bias. For the domain of reporting (Q8), we identified an unclear risk of bias in 10% (2/20) of the included studies.

In addition, all studies were assessed using the JBI checklist,[21] and the findings are shown in Table 2. All studies employed an appropriate sample frame for addressing the target population and provided a detailed description of study participants and setting. Recruitment was not assessed, as all studies were retrospective and based on convenience sampling from medical records. Sample size calculations were rarely reported, introducing uncertainty regarding the precision of prevalence estimates. However, methods for identifying the condition, standardization of measurement, and statistical analysis were generally appropriate, supporting the validity and reliability of the findings. Given the retrospective design, response rate was considered not applicable, limiting the assessment of potential non-response bias.

Histological Types

Of the 2541 bone tumors, 1810 (71.2%) were classified as benign, 279 (11.0%) as intermediate, and 452 (17.8%) as malignant (Supplemental Table 2).

The most frequent benign tumors were enchondroma (n=337; 18.6%), osteoid osteoma (n=311; 17.2%), and simple bone cyst (SBC) (n=287; 15.8%). Among intermediate tumors, giant cell tumor of bone (GCTB) (n=174; 62.2%), osteoblastoma (n=91; 32.7%), desmoplastic fibroma (n=5; 1.8%), and synovial chondromatosis (n=5; 1.8 %) were most common. For primary malignant tumors, the most frequent types were chondrosarcoma (n=120; 26.5%), Ewing sarcoma (n=106; 23.5%), and osteosarcoma (n=92; 20.4%). Bone metastases accounted for 8.8% (n=40) of the malignant tumor cases.

Anatomical Sites

Regarding the 1912 tumors with described anatomical sites, the hindfoot was the most affected segment, accounting for 849 cases (44.4%), followed by forefoot in 599 cases (31.33%), midfoot in 312 cases (16.3%), and ankle in 166 cases (8.6%) (Table 3).

Table 1. General Characteristics of the Included Studies.

Study (year)	Country	Follow-up	Bone Tumors [a]				Sex		Location			
			n	Benign, n (%)	Intermediate, n (%)	Malignant, n (%)	Male	Female	Forefoot	Midfoot	Hindfoot	Ankle
Murari et al[24] (1989)	USA	16y	249	129 (51.8)	79 (31.7)	41 (16.5)	153	96	129	39	81	NR
Casadei et al[3] (1991)	Italy	84y	247	150 (60.7)	28 (11.3)	69 (28.0)	NR	NR	76	26	134	NR
Chou and Malawer[5] (1994)	USA	14y	14	5 (35.7)	1 (7.1)	8 (57.2)	12	2	2	2	2	7
Sarkar et al[30] (1996)	German	13y	23	12 (52.2)	4 (17.4)	7 (30.4)	13	10	8	3	10	1
Ozdemir et al[26] (1997)	Turkey	10y	128	112 (87.5)	10 (7.8)	6 (4.7)	NR	NR	NR	NR	NR	NR
Bakotic and Huvos[2] (2001)	USA	15y	142	59 (41.5)	16 (11.3)	67 (47.2)	70	72	NR	NR	NR	NR
Kinoshita et al[17] (2002)	Japan	26y	35	32 (91.4)	0 (0.0)	3 (8.6)	18	17	22	1	12	NR
Chou et al[4] (2009)	USA	20y	64	31 (48.4)	16 (25.0)	17 (26.6)	NR	NR	NR	NR	NR	NR
Azevedo et al[1] (2013)	Portugal	10y	9	4 (44.5)	3 (33.3)	2 (22.2)	6	3	4	1	NR	4
Ruggieri et al[29] (2014)	Italy	17y	849	640 (75.4)	68 (8.0)	141 (16.6)	NR	NR	NR	NR	NR	NR
Özer et al[27] (2017)	Turkey	30y	157	134 (85.4)	12 (7.6)	11 (7.0)	NR	NR	NR	NR	NR	NR
Delgado Cedillo et al[6] (2007)	Mexico	9y	83	75 (90.4)	7 (8.4)	1 (1.2)	NR	NR	NR	NR	NR	NR
Murahashi et al[23] (2021)	Japan	22y	23	21 (91.3)	0 (0.0)	2 (8.7)	14	11	11	1	6	7
Toepfer et al[34] (2018)	Germany and Switzerland	18y	265	212 (80.0)	18 (6.8)	35 (13.2)	163	102	53	39	104	69
Karadeniz et al[15] (2022)	Turkey	17y	55	35 (63.7)	2 (3.6)	18 (32.7)	NR	NR	NR	NR	NR	NR
Oliveira et al[25] (2022)	Brazil	30y	48	30 (62.5)	4 (8.3)	14 (29.2)	NR	NR	NR	NR	NR	48
Kokubu et al[18] (2022)	Japan	12y	47	41 (87.2)	1 (2.1)	5 (10.6)	NR	NR	NR	NR	NR	NR
Scheele et al[31] (2024)	German	12y	15	10 (66.6)	5 (33.3)	0 (0)	7	8	14	1	0	0
Scheele et al[32] (2024)	German	12y	6	5 (83.3)	1 (16.6)	0 (0)	2	4	0	0	5	1
Jenkins et al[14] (2025)	Scotland	10y	82	73 (89.0)	4 (4.8)	5 (6.0)	NR	NR	29	5	20	28

Abbreviation: NR, not reported.
[a]According to the 2020 World Health Organization (WHO) classification of bone tumors.

When examining the 1364 benign tumors, the hindfoot was also the most common site (n = 592; 43.2%), followed by forefoot (n = 477; 34.8%), midfoot (n = 197; 14.4%), and ankle (n = 117; 8.5%). Among the 227 intermediate tumors, the hindfoot was the most frequent location (n = 102; 44.9%), followed by forefoot (n = 59; 26.0%), midfoot (n = 48; 21.1%), and ankle (n = 13; 5.7%). In the 321 malignant tumors, the hindfoot was the most affected region (n = 155; 48.3%), followed by midfoot (n = 67; 20.9%), forefoot (n = 63; 19.6%), and ankle (n = 36; 11.2%).

Affected bones

Regarding the 604 tumors where the specific bone affected was identified, the calcaneus was the most frequently involved bone, accounting for 186 cases (30.7%), followed by metatarsals in 143 cases (23.7%) and phalanges in 135 cases (22.3%). The tibia and fibula were the least commonly affected, with 9 cases (1.5%) and 1 case (0.17%), respectively (Table 4).

When examining the 367 benign tumors, the calcaneus was also the most commonly affected bone (n = 125; 34.1%), followed by phalanges (n = 106; 28.9%), metatarsals (n = 64; 17.4%), talus (n = 62; 16.9%), navicular (n = 3; 0.8%), cuboid (n = 3; 0.8%), tibia (n = 2; 0.5%), cuneiform bones (n = 1; 0.3%), and fibula (n = 1; 0.3%).

Among the 128 intermediate tumors, the metatarsals were the most frequently affected bones (n = 40; 31.2%), followed by talus (n = 23; 18.0%), calcaneus (n = 21; 16.4%), phalanges (n = 21; 16.4%), cuneiform bones (n = 8; 6.2%),

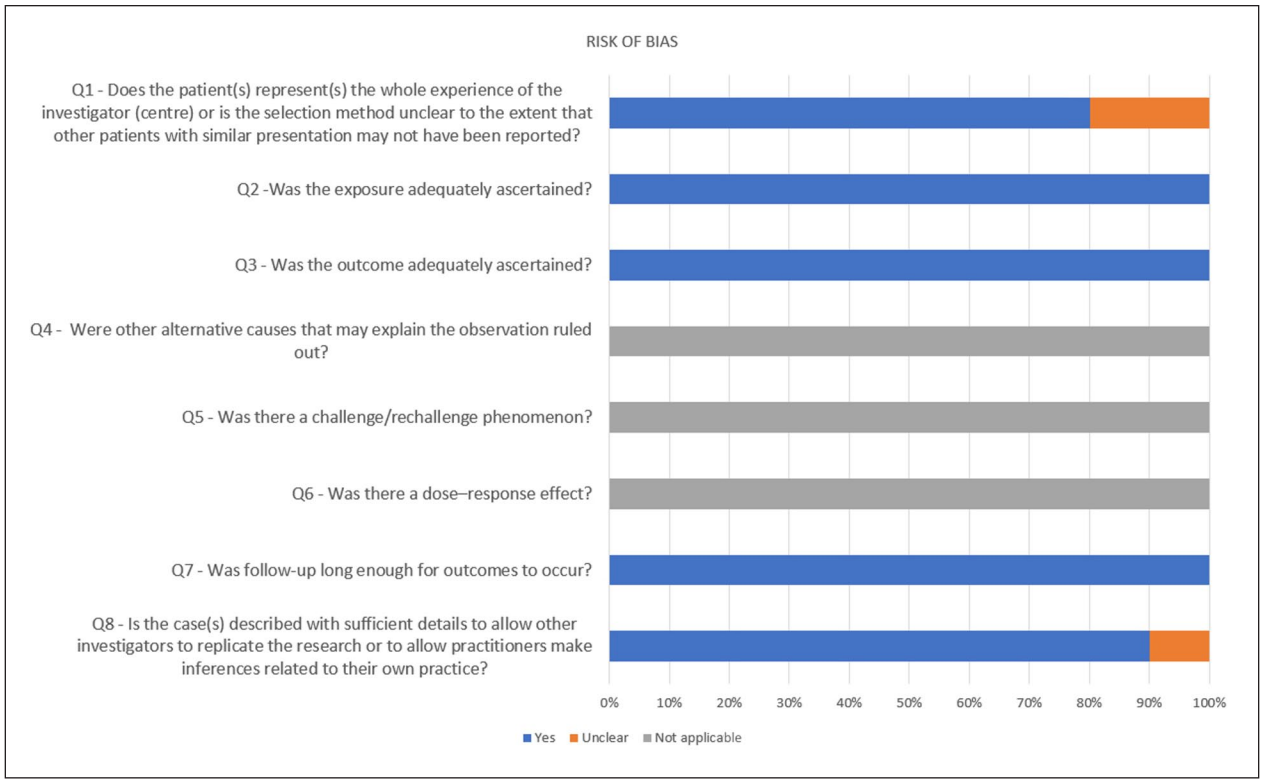

Figure 2. Assessment of risk of bias using the tool developed by Murad et al.[22]

navicular (n=6; 4.7%), and cuboid (n=5; 3.9%). We found no intermediate bone tumors in tibia or fibula.

In the 109 malignant tumors, the calcaneus was the most affected bone (n=40; 36.7%), followed by metatarsals (n=39; 35.8%), talus (n=8; 7.3%), phalanges (n=8; 7.3%), tibia (n=7; 6.4%), cuboid (n=5; 4.6%), and cuneiform bones (n=2; 1.8%). We did not identify any malignant tumors in fibula or navicular.

Patient Sex

Regarding the 989 tumors identified by sex, 558 (56.4%) were found in males and 431 (43.6 %) in females. Specifically, among the 693 benign bone tumors, 388 (56.0%) occurred in males and 305 (44.0%) in females. For the 131 intermediate bone tumors, 70 (53.4%) were found in males and 61 (46.6%) in females. Among the 165 malignant bone tumors, 104 (63.0%) affected males, and 61 (37.0%) affected females (Table 5).

Age Groups

In the selected studies where the age group was identified, a total of 113 tumors were categorized. Most benign bone tumors occurred in individuals under 20 years of age (n=36; 55.4%). Intermediate bone tumors most frequently affected individuals aged 20 to 59 years (n=11; 91.7%). Malignant bone tumors also occurred more frequently between the ages of 20 and 59 (n=22; 61.1%). All bone tumors diagnosed in individuals over 60 years of age were malignant (n=7; 100%) (Supplemental Table 3).

Discussion

In the present study, we sought to characterize representative data for all categories of surgically treated bone tumors located in the foot and ankle, recognizing that findings reflect many institutional surgical series rather than population-based incidence or prevalence. We analyzed a total of 2541 bone tumors across 34 distinct histologic types, classified according to the 2020 WHO criteria.[35] Our choice to use this classification system is justified by the standardization of terminology that facilitates communication between clinicians, oncologic surgeons, orthopaedic oncologists, pathologists, and researchers worldwide. To our knowledge, there are no published systematic reviews specifically detailing the epidemiologic characteristics of surgically treated bone tumors of the foot and ankle.

To uphold the methodologic rigor of this review and minimize selection bias, we limited our inclusion criteria to institutional epidemiologic studies encompassing all types of bone tumors of the foot and ankle. We believe that this

Table 2. Assessment of Risk of Bias Using the Joanna Briggs Institute (JBI) Critical Appraisal Checklist for Studies Reporting Prevalence Data.[a]

Study (Year)	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9
Murari et al[24] (1989)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Casadei et al[3] (1991)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Chou and Malawer[5] (1994)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Sarkar et al[30] (1996)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Ozdemir et al[26] (1997)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Bakotic and Huvos[2] (2001)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Kinoshita et al[17] (2002)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Chou et al[4] (2009)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Azevedo et al[1] (2013)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Ruggieri et al[29] (2014)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Özer et al[27] (2017)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Delgado Cedillo et al[6] (2007)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Murahashi et al[23] (2021)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Toepfer et al[34] (2018)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Karadeniz et al[15] (2022)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Oliveira et al[25] (2022)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Kokubu et al[18] (2022)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Scheele et al[31] (2024)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Scheele et al[32] (2024)	Y	N/A	U	Y	Y	Y	Y	Y	N/A
Jenkins et al[14] (2025)	Y	N/A	U	Y	Y	Y	Y	Y	N/A

Abbreviations: Q, question; Y, yes; N/A, not applicable; U, unclear.
[a]Q1: Was the sample frame appropriate to address the target population? Q2: Were study participants sampled in an appropriate way? Q3: Was the sample size adequate? Q4: Were the study subjects and the setting described in detail? Q5: Was the data analysis conducted with sufficient coverage of the identified sample? Q6: Were valid methods used for the identification of the condition? Q7: Was the condition measured in a standard, reliable way for all participants? Q8: Was there appropriate statistical analysis? Q9: Was the response rate adequate, and if not, was the low response rate managed appropriately?

strategy allows for a more accurate representation of the true distribution of these tumors. In our analysis of 2541 bone tumors, we found that 1810 cases (71.2%) were benign, 279 cases (11.0%) were intermediate, and 452 (17.8%) were malignant.

The higher incidence of benign tumors compared with malignant ones in the foot and ankle aligns with existing literature. Prior research, such as the studies by Ruggieri et al[29] and Toepfer et al,[34] reported similar benign tumor percentages (75.4% and 80.0%, respectively). Conversely, Ozdemir et al,[26] Özer et al,[27] and Delgado Cedillo et al[6] observed higher percentages (87.5%, 85.4%, and 90.4%, respectively), whereas Casadei et al[3] and Murari et al[24] found lower percentages (60.7% and 51.8%, respectively).

Notably, none of these earlier studies included a separate category for intermediate tumors, classifying them within the benign group. The concept of intermediate tumor was implemented in the third edition (2002)[8] of the WHO classification of soft tissue and bone tumors and maintained (with some changes in histologic types) in the fourth (2013) and fifth (2020-current) editions. This division can be somewhat arbitrary and subject to debate, especially regarding tumors that belong to a histologic and biological spectrum of the disease (eg, conventional central and peripheral cartilaginous bone tumors). However, our intention is to specify which tumors would be best managed by regular surveillance without surgical removal; those that need local treatment, such as curettage; and those that require wide or en bloc resection. Intermediate bone tumors have aggressive biological behavior, usually recur locally, and can be associated with an infiltrative and locally destructive growth pattern—they do not have metastatic potential but require wide excision with a margin of normal tissue or the use of adjuvants to ensure local control. Bone tumors classified as intermediate according to the 2020 WHO criteria include atypical cartilaginous tumor, synovial chondromatosis, osteoblastoma, desmoplastic fibroma of bone, epithelioid hemangioma, osteofibrous dysplasia-like adamantinoma, mesenchymoma, and Langerhans cell histiocytosis. GCTB, in addition to being characterized as a locally aggressive intermediate tumor, has well-documented evidence of occasional metastatic dissemination to the lung (<2% of cases), and for this reason, it is also classified as a rare metastatic tumor. Our study identified 11.0% as intermediate tumors. This figure is consistent with the 11.3% reported by both Casadei et al[3] and Bakotic and Huvos,[2]

Table 3. Type of Tumor vs Anatomical Site (Affected Foot Region).

Tumor Type	*k*	n	Forefoot, n (%)	Midfoot, n (%)	Hindfoot, n (%)	Ankle, n (%)
Benign bone tumors						
Enchondroma	7	294	170 (57.8)	97 (33.0)	22 (7.5)	8 (2.7)
Osteoid osteoma	9	275	37 (13.5)	23 (8.4)	201 (73.1)	14 (5.1)
Simple bone cyst	11	196	12 (6.1)	6 (3.1)	163 (83.2)	13 (6.6)
Osteochondroma	11	139	65 (46.8)	25 (18.0)	34 (24.5)	28 (20.1)
Aneurysmal bone cyst	9	105	8 (7.6)	27 (25.7)	53 (50.5)	17 (16.2)
BPOP	2	80	79 (98.8)	1 (1.2)	0 (0.0)	0 (0.0)
Chondroblastoma	8	74	3 (4.1)	7 (9.5)	62 (83.8)	2 (2.7)
Chondromyxoid fibroma	5	52	36 (69.2)	1 (1.9)	14 (26.9)	1 (1.9)
Periosteal chondroma	6	37	30 (81.1)	4 (10.8)	2 (5.4)	1 (2.7)
Lipoma	7	39	2 (5.1)	1 (2.6)	36 (92.3)	0 (0.0)
Nonossifying fibroma	3	27	0 (0.0)	0 (0.0)	0 (0.0)	27 (100.0)
Subungual exostosis	2	26	26 (100.0)	0 (0.0)	0 (0.0)	0 (0.0)
Fibrous dysplasia	5	9	0 (0.0)	4 (44.4)	1 (11.1)	4 (44.4)
Hemangioma	5	8	4 (50.0)	0 (0.0)	3 (37.5)	1 (12.5)
Osteoma	2	2	2 (100.0)	0 (0.0)	0 (0.0)	0 (0.0)
Osteofibrous dysplasia	1	1	0 (0.0)	0 (0.0)	0 (0.0)	1 (100.0)
Overall		1364	477 (34.8)	197 (14.4)	592 (43.2)	117 (8.5)
Intermediate bone tumors	*k*	n	Forefoot	Midfoot	Hindfoot	Ankle
Giant cell tumor of bone	10	138	35 (25.4)	36 (26.1)	55 (39.9)	12 (8.7)
Osteoblastoma	7	78	23 (29.5)	11 (14.1)	44 (56.4)	0 (0.0)
Synovial chondromatosis	2	5	3 (60.0)	1 (20.0)	1 (20.0)	0 (0.0)
Langerhans cell histiocytosis	3	4	1 (25.0)	1 (25.0)	1 (25.0)	1 (25.0)
Desmoplastic fibroma	2	2	0 (0.0)	0 (0.0)	2 (100.0)	0 (0.0)
Overall		227	59 (26.0)	48 (21.1)	102 (44.9)	13 (5.7)
Malignant bone tumors						
Chondrosarcoma	10	92	25 (27.2)	21 (22.8)	36 (39.1)	10 (10.9)
Ewing sarcoma	8	78	13 (16.7)	20 (25.6)	39 (50.0)	6 (7.7)
Osteosarcoma	11	62	9 (14.5)	7 (11.3)	32 (51.6)	14 (22.6)
Epithelioid hemangioendothelioma	4	32	6 (18.8)	11 (34.4)	14 (43.8)	1 (3.1)
Fibrosarcoma	5	17	2 (11.8)	2 (11.8)	12 (70.6)	1 (5.9)
Bone metastases	6	13	2 (15.4)	3 (23.1)	5 (38.5)	3 (23.1)
Lymphoma	3	11	1 (9.1)	0 (0.0)	9 (81.8)	1 (9.1)
Pleomorphic sarcoma, undifferentiated	2	5	2 (40.0)	0 (0.0)	3 (60.0)	0 (0.0)
Plasmacytoma	2	4	0 (0.0)	3 (75.0)	1 (25.0)	0 (0.0)
Giant cell tumor of bone, malignant	1	4	3 (75.0)	0 (0.0)	1 (25.0)	0 (0.0)
Leiomyosarcoma	1	2	0 (0.0)	0 (0.0)	2 (100.0)	0 (0.0)
Angiosarcoma	1	1	0 (0.0)	0 (0.0)	1 (100.0)	0 (0.0)
Overall		321	63 (19.6)	67 (20.9)	155 (48.3)	36 (11.2)

Abbreviations: *k*, number of studies in which the tumor was described; BPOP, bizarre parosteal osteochondromatous proliferation.

whereas Ozdemir et al,[26] Özer et al,[27] and Ruggieri et al[29] reported slightly lower rates (7.8%, 7.6%, and 8.0%, respectively).

Regarding malignant tumors, our rate of 17.8% is comparable to that of Murari et al[24] (16.5%) and Ruggieri et al[29] (16.6%). However, it was lower than the percentages found by Casadei et al[3] (28.0%) and Bakotic and Huvos[2] (47.2%), and higher than those reported by Ozdemir et al[26] (4.7%), Özer et al[27] (6.6%), and Delgado Cedillo et al[6] (1.2%).

In our sample of benign bone tumors, enchondroma was the most prevalent (18.6%), closely followed by osteoid osteoma (17.2%) and SBC (15.8%). It is interesting to note that, although the percentages are different, the order of frequency we observed (enchondroma > osteoid osteoma) aligns with the largest study included in our review.[29] Other studies with relevant sample sizes presented varying sequences: Toepfer et al[34] found SBC as the most frequent, followed by enchondroma and osteochondroma; Murari

Table 4. Type of Tumor vs Affected Bone.

Tumor Type	*k*	n	Tibia, n (%)	Fibula, n (%)	Talus, n (%)	Calcaneus, n (%)	Navicular, n (%)	Cuboid, n (%)	Cuneiform Bones, n (%)	Metatarsals, n (%)	Phalanges, n (%)
Benign bone tumors											
Osteoid osteoma	5	75	0 (0.0)	0 (0.0)	41 (54.7)	10 (13.3)	0 (0.0)	0 (0.0)	0 (0.0)	6 (8.0)	18 (24.0)
Simple bone cyst	5	72	0 (0.0)	0 (0.0)	4 (5.6)	61 (84.7)	0 (0.0)	0 (0.0)	0 (0.0)	7 (9.7)	0 (0.0)
Chondromyxoid fibroma	4	50	0 (0.0)	0 (0.0)	2 (4.0)	11 (22.0)	1 (2.0)	0 (0.0)	0 (0.0)	19 (38.0)	17 (34.0)
Osteochondroma	9	32	0 (0.0)	0 (0.0)	0 (0.0)	7 (21.9)	0 (0.0)	0 (0.0)	0 (0.0)	11 (34.4)	14 (43.8)
Chondroblastoma	3	29	0 (0.0)	0 (0.0)	7 (24.1)	18 (62.1)	0 (0.0)	1 (3.4)	0 (0.0)	3 (10.3)	0 (0.0)
Periosteal chondroma	4	27	0 (0.0)	0 (0.0)	1 (3.7)	0 (0.0)	1 (3.7)	0 (0.0)	0 (0.0)	6 (22.2)	19 (70.4)
Subungual exostosis	3	26	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	26 (100.0)
Aneurysmal bone cyst	5	21	2 (9.5)	1 (4.8)	6 (28.6)	6 (28.6)	0 (0.0)	1 (4.8)	0 (0.0)	4 (19.0)	1 (4.8)
Enchondroma	3	17	0 (0.0)	0 (0.0)	0 (0.0)	3 (17.6)	0 (0.0)	0 (0.0)	0 (0.0)	3 (17.6)	11 (64.7)
Lipoma	3	9	0 (0.0)	0 (0.0)	0 (0.0)	8 (88.9)	0 (0.0)	0 (0.0)	0 (0.0)	1 (11.1)	0 (0.0)
Hemangioma	3	5	0 (0.0)	0 (0.0)	0 (0.0)	1 (20.0)	0 (0.0)	0 (0.0)	1 (20.0)	3 (60.0)	0 (0.0)
Fibrous dysplasia	2	3	0 (0.0)	0 (0.0)	1 (33.3)	0 (0.0)	1 (33.3)	1 (33.3)	0 (0.0)	0 (0.0)	0 (0.0)
Osteoma	1	1	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (100.0)	0 (0.0)
Overall		367	2 (0.5)	1 (0.3)	62 (16.9)	125 (34.1)	3 (0.8)	3 (0.8)	1 (0.3)	64 (17.4)	106 (28.9)
Intermediate bone tumors											
Giant cell tumor of bone	6	67	0 (0.0)	0 (0.0)	5 (7.5)	14 (20.9)	4 (6.0)	5 (7.5)	7 (10.4)	23 (34.3)	9 (13.4)
Osteoblastoma	5	50	0 (0.0)	0 (0.0)	18 (36.0)	6 (12.0)	2 (4.0)	0 (0.0)	1 (2.0)	11 (22.0)	12 (24.0)
Synovial chondromatosis	1	4	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (25.0)	1 (25.0)	2 (50.0)
Desmoplastic fibroma	1	4	0 (0.0)	0 (0.0)	0 (0.0)	1 (25.0)	0 (0.0)	0 (0.0)	0 (0.0)	3 (75.0)	0 (0.0)
Langerhans cell histiocytosis	1	3	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	3 (100.0)	0 (0.0)
Overall		128	0 (0.0)	0 (0.0)	23 (18.0)	21 (16.4)	6 (4.7)	5 (3.9)	8 (6.2)	40 (31.2)	21 (16.4)
Malignant bone tumors											
Chondrosarcoma	4	40	2 (5.0)	0 (0.0)	2 (5.0)	13 (32.5)	0 (0.0)	4 (10.0)	1 (2.5)	15 (37.5)	3 (7.5)
Ewing sarcoma	4	24	0 (0.0)	0 (0.0)	1 (4.2)	10 (41.7)	0 (0.0)	0 (0.0)	1 (4.2)	11 (45.8)	1 (4.2)
Osteosarcoma	5	21	2 (9.5)	0 (0.0)	3 (14.3)	7 (33.3)	0 (0.0)	0 (0.0)	0 (0.0)	6 (28.6)	3 (14.3)
Fibrosarcoma	3	7	1 (14.3)	0 (0.0)	0 (0.0)	5 (71.4)	0 (0.0)	0 (0.0)	0 (0.0)	1 (14.3)	0 (0.0)
Pleomorphic sarcoma, undifferentiated	2	5	0 (0.0)	0 (0.0)	1 (20.0)	2 (40.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (20.0)	1 (20.0)
Giant cell tumor of bone, malignant	1	4	0 (0.0)	0 (0.0)	0 (0.0)	1 (25.0)	0 (0.0)	0 (0.0)	0 (0.0)	3 (75.0)	0 (0.0)
Bone metastases	2	3	2 (66.7)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (33.3)	0 (0.0)	0 (0.0)	0 (0.0)
Epithelioid hemangioendothelioma	2	2	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	2 (100.0)	0 (0.0)
Angiosarcoma	1	1	0 (0.0)	0 (0.0)	1 (100.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Plasmacytoma	1	1	0 (0.0)	0 (0.0)	0 (0.0)	1 (100.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Lymphoma	1	1	0 (0.0)	0 (0.0)	0 (0.0)	1 (100.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Overall		109	7 (6.4)	0 (0.0)	8 (7.3)	40 (36.7)	0 (0.0)	5 (4.6)	2 (1.8)	39 (35.8)	8 (7.3)

Abbreviation: *k*, number of studies in which the tumor was described.

et al[24] observed chondromyxoid fibroma as the most common, followed by SBC and chondroblastoma; and Casadei et al[3] identified osteoid osteoma as the most common, followed by SBC and enchondroma.

Regarding intermediate tumors in our sample, GCTB was the most common (62.2%), followed by osteoblastoma (32.7%). This order is consistent with the findings of Ruggieri et al,[29] Murari et al,[24] and Toepfer et al,[34] although the proportions differed. Notably, Casadei et al[3] reported an equal number of cases for these 2 tumors.

Our analysis of malignant tumors revealed chondrosarcoma (26.5%) as the most frequent, followed by Ewing sarcoma (23.5%), osteosarcoma (20.4%), and bone metastases (8.8%). These findings are supported by the study of Özer et al,[27] who also identified chondrosarcoma as the most common malignant tumor, although at a considerably higher rate (63%), with Ewing sarcoma and osteosarcoma each representing 18% of cases. In our analysis of 1912 bone tumors with reported anatomical sites, the hindfoot was the most frequently affected area, representing nearly half of the cases, followed by the forefoot, midfoot, and ankle. These findings align with the study conducted by Casadei et al,[3] who also identified the hindfoot as the primary site, followed by the forefoot and midfoot, for both benign and malignant bone tumors. However, other studies have presented different frequency sequences. Toepfer

Table 5. Type of Tumor vs Patient Sex.

Tumor Type	*k*	n	Male, n (%)	Female, n (%)
Benign bone tumors				
Simple bone cyst	8	263	156 (59.3)	107 (40.7)
Osteochondroma	8	87	45 (51.7)	42 (48.3)
Enchondroma	6	74	42 (56.8)	32 (43.2)
Chondromyxoid fibroma	5	47	25 (53.2)	22 (46.8)
Aneurysmal bone cyst	5	41	21 (51.2)	20 (48.8)
Chondroblastoma	5	38	28 (73.7)	10 (26.3)
Subungual exostosis	2	32	5 (15.6)	27 (84.4)
Lipoma	6	33	20 (60.6)	13 (39.4)
Osteoid osteoma	5	31	24 (77.4)	7 (22.6)
Non-ossifying fibroma	2	14	7 (50.0)	7 (50.0)
Periosteal chondroma	4	15	7 (46.7)	8 (53.3)
Hemangioma	5	10	4 (40.0)	6 (60.0)
Fibrous dysplasia	2	3	1 (33.3)	2 (66.7)
BPOP	1	3	3 (100.0)	0 (0.0)
Osteoma	2	2	0 (0.0)	2 (100.0)
Overall		693	388 (56.0)	305 (44.0)
Intermediate bone tumors	*k*	n	Male	Female
Giant cell tumor of bone	6	80	41 (51.2)	39 (48.8)
Osteoblastoma	5	42	22 (52.4)	20 (47.6)
Synovial chondromatosis	2	5	4 (80.0)	1 (20.0)
Desmoplastic fibroma	2	4	3 (75.0)	1 (25.0)
Overall		131	70 (53.4)	61 (46.6)
Malignant bone tumors	*k*	n	Male	Female
Chondrosarcoma	7	57	39 (68.4)	18 (31.6)
Osteosarcoma	6	31	16 (51.6)	15 (48.4)
Ewing sarcoma	4	29	20 (69.0)	9 (31.0)
Bone metastases	4	19	9 (47.4)	10 (52.6)
Epithelioid hemangioendothelioma	2	9	5 (55.6)	4 (44.4)
Fibrosarcoma	2	7	5 (71.4)	2 (28.6)
Malignant giant cell tumor of bone	1	4	3 (75.0)	1 (25.0)
Pleomorphic sarcoma, undifferentiated	3	4	3 (75.0)	1 (25.0)
Leiomyosarcoma	1	2	1 (50.0)	1 (50.0)
Plasmacytoma	1	1	1 (100.0)	0 (0.0)
Angiosarcoma	1	1	1 (100.0)	0 (0.0)
Lymphoma	1	1	1 (100.0)	0 (0.0)
Overall		165	104 (63.0)	61 (37.0)

Abbreviations: *k*, number of studies in which the tumor was described; BPOP, bizarre parosteal osteochondromatous proliferation.

et al[34] reported a sequence of hindfoot, ankle, forefoot, and midfoot, whereas Murari et al[24] observed the highest incidence in the forefoot, followed by hindfoot and midfoot.

When examining the specific bones involved in a subset of 604 tumors with this information available, the calcaneus was the most affected bone (30.7%). This predominance was consistent across both benign (34.1%) and malignant tumors (36.7%). Conversely, intermediate tumors were most often located in metatarsals (31.2%). Our observation of calcaneus being the most prevalent site is consistent with the findings of Ozdemir et al,[26] who reported this location in 33.0% of cases. It is worth mentioning, however, that Murari et al[24] found a higher incidence of tumors in metatarsals (35.2%). Regarding the sex of the affected patients in the studies where this information was available, our analysis revealed a male predominance, accounting for 56.8% of cases. This trend aligns with observations in the studies by Toepfer et al[34] (61.5% male) and Murari et al[24] (61.4% male). Within our sample, we found the following distribution across different tumor types: (1) benign tumors occurred in 56.0% of men; (2) intermediate tumors affected 53.4% of males; and (3) malignant tumors showed a considerably higher incidence in males, with 63% of cases. These findings are consistent with previous studies. Toepfer et al[34]

reported that, of 231 benign bone tumors, 59.7% occurred in men; of 35 malignant bone tumors, 57.1% affected males. Similarly, Murari et al[24] found that, among 208 benign bone tumors, 62.5% occurred in men, whereas of the 41 malignant bone tumors, 56.1% affected men.

In our review, patient age was reported for less than 5% of the tumors (113/2541). As such, these age-stratified findings are only descriptive, unreliable, and should be interpreted with extreme caution and should not be generalized to routine clinical populations. In these studies, most bone tumors were diagnosed in individuals aged 20-59 years, followed by those aged <20 years and those 60 years or older. Benign tumors were most prevalent in individuals aged <20 years (84.1% of cases) and in those aged 20-59 years (64.5% of cases). Conversely, malignant tumors showed an inverse pattern, representing 100% of tumors in the 60 years or older age group, 35.5% in the 20-59 years group, and 15.9% in the age <20 years group. Notably, Kinoshita et al[17] did not identify malignant bone tumors in patients <50 years of age, and half of the tumors in patients >50 years of age were malignant. Toepfer et al[34] also observed an increasing frequency of malignant bone tumors with advancing age. Although these trends align with previous studies, they should be viewed as exploratory because of the limited number of cases with age data.

Limitations of this systematic review include considerable heterogeneity among the included studies, particularly in the various data collection methods, varying durations of patient follow-up, and different tumor classification criteria, as well as incomplete data that restricted more comprehensive analyses. It is important to consider that our analysis focused on a subgroup of foot and ankle bone tumors that were treated surgically, with confirmed histologic diagnoses, which may limit the applicability of our findings to other contexts. Another important limitation is that our sample included only individuals who sought medical attention; therefore, it is reasonable to assume that some patients with benign bone tumors have not presented for evaluation or care, which is less likely to occur in patients with intermediate and malignant tumors. In addition to these factors, several key sources of heterogeneity must be acknowledged. The era of data collection varied widely across studies, ranging from 9 to 84 years, which encompasses significant changes in diagnostic tools, histopathologic techniques, and tumor classification systems. This temporal variability may have impacted the consistency of diagnoses. Although we reclassified histologic types according to the 2020 WHO system, many studies employed outdated or inconsistent nomenclatures. Furthermore, the majority of the included studies were retrospective, single-center case series, often lacking clear inclusion criteria or detailed reporting on data completeness, which increases the risk of selection bias.

In addition to the overall methodologic heterogeneity, another important aspect is the potential geographic bias inherent to our review. Most of the included cohorts originated from European centers, which may limit the generalizability of our findings to non-European populations. Although a subgroup analysis by region would be valuable, heterogeneity in reporting and the limited number of studies from other areas precluded a formal regional analysis. Furthermore, although the geographic location of the research centers was identifiable, information on the race, ethnicity, or nationality of the study participants was generally not reported, making it difficult to assess population-level demographic variation. Future studies should aim to include data from underrepresented regions and report participant-level demographic characteristics to enhance the external validity and applicability of the findings.

These sources of heterogeneity limited the feasibility of conducting meta-analytical synthesis and justified the choice of a descriptive approach. They also reinforce the need for future prospective studies using standardized classification systems and reporting frameworks. Another limitation is the small sample size of several included studies, which may reduce the accuracy of estimates for less common tumor subtypes and limit the robustness of subgroup analyses.

Although our review did not aim to establish clinical decision-making algorithms, the descriptive epidemiologic patterns of surgically treated foot and ankle bone tumors presented here may serve as a valuable foundation for future studies focusing on diagnostic protocols, biopsy planning, and treatment strategies for bone tumors of the foot and ankle.

Conclusion

This systematic review provides a comprehensive characterization of surgically treated bone tumors occurring in the foot and ankle. Our analysis encompasses histologic type, anatomical site, the specific bone involved, patient sex, and age group with interpretation limited by heterogeneity and incomplete demographic reporting (particularly age). While acknowledging the challenge of data heterogeneity across the included studies for absolute comparisons, the consistency of our findings provide valuable insights into the epidemiology of surgically treated bone tumors of the foot and ankle.

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Ethical Considerations

The study protocol was registered with the International Prospective Register of Systematic Reviews (PROSPERO), registration number CRD42021281687. This systematic review was exempt from research ethics committee approval as it did not involve private information from participants or any potential violation of human rights.

Consent for Publication

Not applicable.

Author Contributions

All authors declare that they participated in the research that led to the manuscript, read and approved the text, tables, and figures submitted.

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Data Availability Statement

All relevant data are within the paper.

Supplemental Material

Supplementary material is available online with this article.

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