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EPIDEMIOLOGICAL PROFILE AND EVOLUTION OF ANKLE MUSCULOSKELETAL TUMORS

PERFIL EPIDEMIOLÓGICO E EVOLUÇÃO NOS TUMORES MUSCULOESQUELÉTICOS AO NÍVEL DO TORNOZELO

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ABSTRACT

Objective: Characterizing ankle tumors, presenting the epidemiological profile of these lesions. Methods: Retrospective observational case series study to evaluate the results of clinical and/or surgical treatments of patients with ankle tumors whose first visit occurred from 1990 to 2020. The dependent variables were: benign bone tumor, malignant bone tumor, benign soft tissue tumor, malignant soft tissue tumor, and infection. The independent variables were: sex, age; presence of symptoms (pain/local volume increase/fracture), duration of symptoms until treatment, diagnosis, treatment, and recurrence. Results: In total, 70 patients were included—58.5% were women, with a mean age at the time of diagnosis of 21.66 years. Among all cases, 76% were bone tumor, 14% were soft tissue tumor, and 10% were infection. The mean age at the time of diagnosis was 21.7 ± 2.29 years. The overall prevalence of pain was 77.1%. In total, 55.6% patients had a general local volume increase 13.4% had fractures. The mean time from symptoms to treatment was 17.4 ± 4.61 months and the mean diagnosis time was 10.13 ± 0.86 months. Of all cases, 73.44% underwent surgical treatment and 22.64% had recurrence. Conclusion: In this series, ankle tumors corresponded mainly to bone tumors. Benign tumors were the most prevalent type of tumor and the highest occurrence was among young people. ***Level of Evidence IV, Case Series.***

Keywords: Neoplasms. Sarcoma. Ankle. Amputation. Neoplasm Recurrence, Local.

RESUMO

Objetivos: Caracterizar tumores da região do tornozelo apresentando o perfil epidemiológico destas lesões. Métodos: Estudo observacional retrospectivo de série de casos para avaliação dos resultados de neoplasias do tornozelo submetidos a tratamento clínico e/ou cirúrgico em que o primeiro atendimento tenha ocorrido entre 1990 e 2020. As variáveis dependentes foram: tumor ósseo benigno, tumor ósseo maligno, tumor de partes moles benigno, tumor de partes moles maligno e infecção. As variáveis independentes foram: sexo, idade, presença de sintomas (dor/aumento de volume local/fratura), tempo de sintomas até o atendimento, diagnóstico, tratamento e recidiva. Resultados: Foram analisados 70 pacientes, sendo 58,5% do sexo feminino, com média de idade no momento do diagnóstico de 21,66 (21,7 ± 2,29) anos. As neoplasias ósseas correspondem a 76% dos casos, seguidas de tumor de partes moles com 14% e de infecção com 10%. A prevalência geral de dor foi de 77,1%. O aumento geral de volume local ocorreu em 55,6% pacientes e presença de fraturas em 13,4%. A média de tempo de sintomas até o atendimento foi de 17,4 ± 4,61 meses e a média de tempo para o diagnóstico foi de 10,13 ± 0,86 meses. O tratamento cirúrgico ocorreu em 73,44% dos casos e a recidiva em 22,64%. Conclusão: Os tumores ao nível do tornozelo nesta série correspondem majoritariamente a tumores ósseos, com prevalência do benigno e maior ocorrência em jovens. ***Nível de Evidência IV, Série de Casos.***

Descritores: *Neoplasias. Sarcoma. Tornozelo. Amputação. Recidiva Local de Neoplasia.*

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INTRODUCTION

About 3% to 4% of cases of bone tumors affect the ankle, mostly soft tissue tumors,[1] of which about 8% are benign and 5% are malignant.[2] Malignant lesions and especially secondary lesions (metastatic lesions) are extremely rare.[3]

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The study was conducted at Institute of Pediatrics Oncology (IOP/GRAAC), Universidade Federal de São Paulo.
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Among studies in the literature addressing neoplastic lesions of the foot and ankle, sarcomas of the foot show a distinct biological behavior when compared with the same type of tumor in other skeletal sites. Moreover, knowing incidence and distribution patterns of tumors of the foot and ankle helps in their evaluation, diagnosis, and treatment.[4-6]

The distal anatomy of the lower limb is of great complexity and, along with the rarity of this type of lesion, its diagnosis becomes difficult, especially for general orthopedists.[2] This region is anatomically small, with little muscle and subcutaneous tissue, which prevents neoplastic lesions from growing for long periods unnoticed.[1] The anatomical peculiarity of this region is a challenge to limb-salvage surgery in the context of obtaining broad oncological margins.[7]

Patients with ankle tumors should be evaluated in a systematic and judicious way with a good anamnesis and physical examination, followed by complementary imaging tests to formalize a diagnostic hypothesis and, when necessary, schedule a biopsy.[2,8,9] In general, the initial imaging test is a simple X-ray, but ultrasound and computed tomography may also be necessary. In case of local staging of the lesion, magnetic resonance imaging with contrast remains the best imaging test for diagnostic evaluation and treatment planning.[1]

Inadequate treatment of these lesions can cause a great impairment regarding prognosis and functional outcome of patients. Unplanned resection and local recurrences generally occur with the need for more aggressive approaches.[4]

To the best our knowledge, no study in the Brazilian literature characterizes ankle tumors. Thus, this study aimed to characterize ankle tumors, presenting the epidemiological profile of lesions treated in an oncology reference center in Brazil.

METHODS

This retrospective observational case series study was performed to evaluate the results of ankle musculoskeletal tumors. Data were collected from medical and imaging records of patients and a specific database was constructed for this study with total protection regarding the identification of patients. This study was approved by the institutional Research Ethics Committee and is registered in the Brazil Platform under no. 41308920.9.0000.5505.

Medical records of 101 patients diagnosed with neoplasm of the distal end of the lower limbs, whose first visit occurred from 1990 to 2020, undergoing clinical and/or surgical treatments were analyzed. The inclusion criteria were patients of both sexes, with no age limit, followed up at the institution, with an ankle musculoskeletal tumor, which could be defined as:

i. Bone lesion of the talus;
ii. Bone lesion in the distal fibula to the upper limit of the tibial Heim triangle, including lateral malleolus;
iii. Bone lesion of the distal end of the tibia, defined by the metaphyseal region (upper limit of the Heim triangle) to the articular surface of the distal tibia, including the medial malleolus;
iv. Extraosseous lesion (soft tissue) covering the distal anatomical regions to the upper limit of the metaphyseal region of the distal tibia and in proximity to the distal limit of the talus.

Musculoskeletal infections (osteomyelitis, soft tissue infection) that did a differential diagnosis with musculoskeletal neoplasms in this region were included in the study.

All patients with tumors of lower limbs proximal or distal to the area of interest or lesions with extension by contiguity to the ankle, but an epicenter that was not in the region to be studied were excluded. The lack of patient compliance to participate in the study, at any time, was also considered an exclusion criterion.

All patients were evaluated according to general epidemiological variables: (1) sex; (2) age; (3) presence of symptoms (pain/local volume increase/fracture), (4) duration of symptoms until treatment; (5) diagnosis; (6) treatment; (7) recurrence of the lesion after surgical treatment. In this study, ankle lesions were considered a dependent variable: benign bone tumor, malignant bone tumor, benign soft tissue tumor, malignant soft tissue tumor, and infection.

The differential diagnosis of bone infection was confirmed by culture and/or anatomopathological exam. The studied variables were collected by analysis of medical records. Bone tumors were confirmed by biopsy.

The construction of the database and graph creation was performed using the Excel (Microsoft®) software. For statistical analysis, the SPSS® software (IBM, V21) was used. Descriptive analyses are presented in absolute number (n) and relative frequency (%), mean and standard deviation. The Fisher Exact test was used to compare the relative frequency lower than five. ANOVA was used to compare the means of numerical variables of three or more groups. Epidemiological analyses of the variables studied were performed, with a description of categorical and continuous variables.

RESULTS

In total, 70 patients with ankle lesions were included. Among them, 41 (58.6%) were women and 29 (41.4%) were men. The mean age of patients with ankle tumors at the time of diagnosis was 21.7 ± 2.29 years. A total of 47 (77.1%) patients reported pain, 35 (55.6%) had a local volume increase, and nine (13.4%) had fractures. The mean duration of symptoms until treatment was 17.4 ± 4.61 months (range of 0 to 180 months). The mean diagnosis time was 10.13 ± 0.86 months. In this study, 47 patients (73.44%) underwent surgical treatment. Regarding the side affected by the lesion, 49.23% (n = 32) had its right side affected and 46.15% (n = 30) its left side. Recurrence occurred in 22.64% (n = 12) of the cases (Table 1).

Regarding the prevalence of ankle lesions, benign bone tumors represented 55.71% (n = 39) of cases, malignant bone tumors 20% (n = 14), malignant soft tissue tumors 8% (n = 6), infections 10% (n = 7), and benign soft tissue tumor 6% (n = 4). Figure 1 shows the distribution of diagnoses of ankle lesions. Among benign bone tumors, non-ossifying fibroma was the most prevalent (14%; n = 10), followed by osteochondroma (10%; n = 7), Trevor disease (7%; n = 5), and aneurysmal bone cyst (6%; n = 4). Among malignant bone tumors, osteosarcoma (9%; n = 6) and Ewing sarcoma (6%; n = 4) were the most prevalent. Among malignant soft tissue tumors, synovial sarcoma was the most prevalent (6%; n = 4). Figure 2 presents X-ray images of patients with chondroblastic osteosarcoma, chondrosarcoma, osteomyelitis, and Trevor disease. Table 1 also shows the prevalence and means of ankle lesions according to the variables studied and association information. The local volume increase of lesions was different (p = 0) and benign bone tumors presented an increase greater than other lesions (34.29%; n = 12). Moreover, the mean diagnosis time was also different (p = 0).

DISCUSSION

This study presents essential information and contributions to the field of studies on ankle musculoskeletal tumors. Tumors of the foot and ankle are generally uncommon. However, despite the rarity, orthopedic surgeons must know the epidemiological profile, diagnostic criteria, and therapeutic options of patients, as each tumor varies in presentation, degree of aggressiveness, and natural history of the disease.[2]

In this study, benign bone tumors presented higher prevalence, with no statistical differences between women and men. This type of tumor affected mainly adolescents and non-ossifying fibroma and osteochondroma were the most prevalent diagnoses. This type of tumor presented higher prevalence of pain, local increased volume,

Table 1. Sample characterization, prevalence, and mean/standard deviation of ankle lesions and their association with the studied variables (n = 70).

	Total n (%)	BBT n (%)	MBT n (%)	Benign STT n (%)	Malignant STT n (%)	Infection n (%)	
		39 (55.71%)	14 (20%)	4 (5.71%)	6 (8.57%)	7 (10%)	
Sex							0.082*
Women	41 (58.57%)	22 (53.66%)	12 (29.27%)	1 (2.44%)	2 (4.88%)	4 (9.76%)	
Men	29 (41.43%)	17 (58.62%)	2 (6.9%)	3 (10.34%)	4 (13.79%)	3 (10.34%)	
Age (mean/SD)	21.66 2.29	17.08 1.84	27.31 7.75	42.75 13.87	35 6.06	12.57 6.35	0.064**
Pain							0.852*
No	14 (22.95%)	9 (64.29%)	1 (7.14%)	1 (7.14%)	2 (14.29%)	1 (7.14%)	
Yes	47 (77.05%)	26 (55.32%)	8 (17.02%)	3 (6.38%)	4 (8.51%)	6 (12.77%)	
Local volume increase							0.000*
No	28 (44.44%)	25 (89.29%)	2 (7.14%)	0 (0%)	0 (0%)	1 (3.57%)	
Yes	35 (55.56%)	12 (34.29%)	8 (22.86%)	3 (8.57%)	6 (17.14%)	6 (17.14%)	
Fracture							0.180*
No	58 (86.57%)	34 (58.62%)	11 (18.97%)	4 (6.9%)	5 (8.62%)	4 (6.9%)	
Yes	9 (13.43%)	4 (44.44%)	1 (11.11%)	0 (0%)	1 (11.11%)	3 (33.33%)	
Duration of symptoms until treatment (mean/SD)	17.40 4.61	20.04 6.69	5.78 1.10	72 48	30 6	2.57 0.68	0.539**
Diagnosis time (mean/SD)	10.13 0.86	5.31 0.80	13.85 0.85	16.5 4.79	17.5 0.34	19.57 0.30	**0.000***
Surgical treatment							0.299*
No	17 (26.56%)	13 (26.56%)	3 (17.65%)	1 (5.88%)	0 (0%)	0 (0%)	
Yes	47 (73.44%)	23 (73.44%)	11 (23.4%)	3 (6.38%)	6 (12.77%)	4 (8.51%)	
Affected side							0.230*
Left	30 (46.15%)	14 (46.67%)	5 (16.67%)	1 (3.33%)	4 (13.33%)	6 (20%)	
Right	32 (49.23%)	19 (59.38 %)	8 (25%)	2 (6.25%)	2 (6.25%)	1 (3.13%)	
Both	3 (4.62%)	2 (66.67%)	0 (0%)	1 (33.33%)	0 (0%)	0 (0%)	
Recurrence							0.955*
No	41 (77.36%)	20 (48.78%)	9 (21.95%)	3 (7.32%)	5 (12.2%)	4 (9.76%)	
Yes	12 (22.64%)	7 (58.33%)	3 (25%)	1 (8.33%)	1 (8.33%)	0 (0%)	

BBT: benign bone tumor; MBT: malignant bone tumor; Benign STT: benign soft tissue tumor; Malignant STT: malignant soft tissue tumor; SD: standard deviation; *Fisher exact test; **ANOVA; $p < 0.05$: statistically significant.

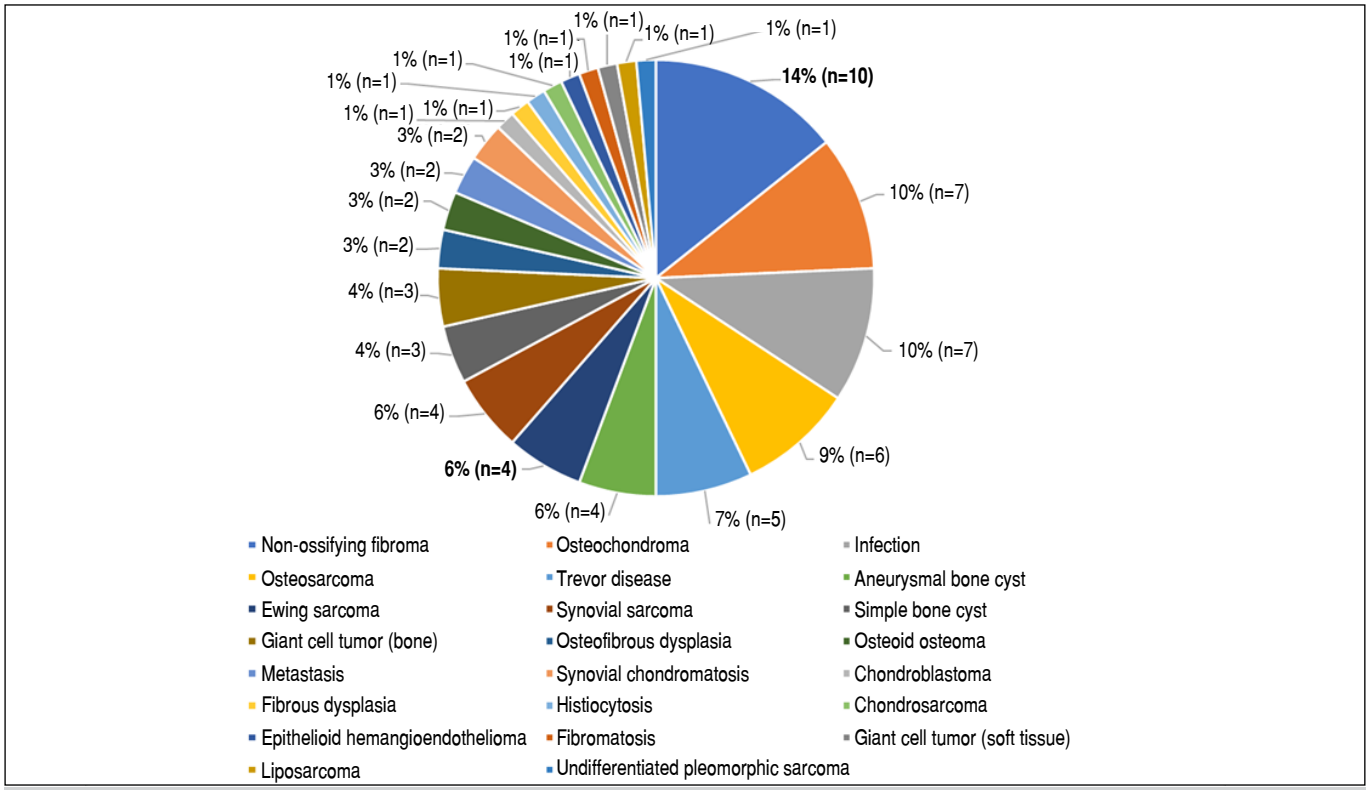

Figure 1. Distribution of diagnoses of ankle lesions.

Figure 2. X-ray images: A) woman, 23 years of age, chondroblastic osteosarcoma; B) man, 63 years of age, grade II chondrosarcoma; C) man, 11 months of age, osteomyelitis; D) woman, eight years of age, Trevor disease.

and recurrence in comparison with other types of tumor. Our findings are in line with other studies.[1,3] Ozdemir et al.[1] reported 1,786 bone and soft tissue tumors, of which 87.2% were benign. Moreover, Chou, Ho, and Malawer[3] reported a prevalence of 60.8% of benign ankle and foot lesions.

Malignant bone tumors were more prevalent among young women (27.31 ± 7.75 years). It was the second type of tumor with the highest prevalence of pain, local increased volume, and recurrence. Osteosarcoma and Ewing sarcomas were the most prevalent diagnoses for this type of tumor. Brotzmann et al.[6] showed that for malignant bone tumors, the first symptoms, such as pain and edema, are nonspecific and often misinterpreted as inflammatory or post-traumatic in nature. The authors also stated that the variety of differential diagnoses explains the long delay in the diagnosis of bone tumors in general, but not the marked difference between foot tumors and those in other skeletal sites.

In our study, 5.71% of the cases were benign soft tissue tumors, with lower prevalence of pain, local increased volume, and risk of fractures. The diagnoses of this type of tumor were synovial chondromatosis (3%), extra-abdominal fibromatosis (1%), and giant cell tumors (1%). In a study by Ruggieri et al.,[10] 189 patients (16.15%) were diagnosed with soft tissue lesions, of whom 91 (48.15%) were non-malignant (pseudotumors or benign tumors).

Malignant soft tissue tumors were more predominant among adult men (mean age of 35 years) and synovial sarcoma (6%), liposarcoma (1%), and undifferentiated pleomorphic sarcoma (1%) were the most prevalent diagnoses. In a study by Toepfer et al.,[4] 78 cases (18.8%) were malignant tumors. Soft tissue tumors showed a malignancy rate (29.2%) higher than bone lesions (13.1%).

In our study, diagnoses of infections were osteomyelitis and infected Charcot osteoarthropathy. Charcot osteoarthropathy is a relatively painless, progressive, and degenerative arthropathy of one or more joints caused by underlying neurological deficits. In contrast to Charcot osteoarthropathy, osteomyelitis is a bone infection that can reach the bloodstream from nearby tissues.[11]

The strength of this study refers to information on the epidemiological profile of ankle lesions. However, this study had limitations, such as its design (case series) and data obtained from a single center, which limits the interpretation at the national level of the findings.

CONCLUSION

In this series, ankle tumors corresponded mainly to bone tumors. Benign tumors were the most prevalent type of tumor and the highest occurrence was among young people. Knowing the epidemiological profile of ankle lesions can help to improve the understanding of the pathology and, consequently, the therapeutic success.

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