Treatment trends of benign bone lesions in a suburban New York healthcare system

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ABSTRACT

Introduction: The management of benign bone lesions is controversial as it is dependent on a multitude of factors such as age, anatomic location, comorbidities, lesion metabolic activity, surgeon preferences, and goals of care, among others. Thus far, many studies have attempted to report on these lesions; however, most are heterogeneous compilations of benign and malignant lesions with nearly all failing to report patient treatment and none of which have originated from a suburban area of the United States. The goal of this study was to establish a modern database dedicated solely to benign bone tumors to reflect current diagnosis and treatment trends in suburban New York. **Materials and Methods:** This was a multicenter retrospective observational study with inclusion criteria limited to benign bone lesions of all ages. Malignant lesions were excluded. Patients were drawn from both primary care provider and surgeon records, with documentation of their associated management. **Results:** A total of 689 patients met inclusion criteria. The overall operative rate for this cohort was 71.6%. In agreement with current literature, aneurysmal bone cysts, giant cell tumors, and osteochondromas underwent surgery more frequently than enchondromas; older patients underwent surgery less frequently; benign bone lesions were more commonly found in younger males, and the distal femur and proximal tibia were the most common locations for lesions ($P < .05$ for all findings). **Conclusion:** This study demonstrates the management of a globally representative variety of benign bone lesions in a diverse suburban population of New York and should facilitate future research on how lesion type, location, management, and other factors relate to patient outcomes.

Keywords: Benign bone tumor, demographics, epidemiology, lesion, orthopedic oncology

Introduction

The prevalence of benign bone lesions (i.e., any nonmalignant pathologic process or neoplasm located within the bone) is difficult to estimate, as these lesions typically present without symptoms and are most often found incidentally on radiographic studies,[1-3] but there is evidence indicating a rate approaching 20%.[4] Examples of benign bone lesions include osteoid osteoma, osteochondroma, solitary bone cyst (SBC), aneurysmal bone cyst (ABC), nonossifying fibroma (NOF), giant cell tumor of bone (GCT), fibrous dysplasia (FD), chondroblastoma, Langerhans cell histiocytosis (LCH), and enchondroma.

The treatment of benign bone lesions is controversial with no clear consensus on a gold standard of care.[5] The reasons for this controversy are multifactorial, with management being driven by the underlying diagnosis, lesion metabolic activity, age, location, comorbidities, surgeon preferences, and goals of care. Regionality, access to care, and institutional and societal culture may also play a role in management. A lack of large patient databases with complete management data dedicated solely to

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Quick Response Code:

Website:
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DOI:
10.4103/jfmpc.jfmpc_5_23

How to cite this article: Muran A, Fallon J, Jung B, Dzaugis P, Zhang A, Fitzgerald M, *et al*. Treatment trends of benign bone lesions in a suburban New York healthcare system. J Family Med Prim Care 2023;12:1979-83.

benign bone lesions is another contributing factor to the absence of consensus on management.

There have been several studies that have generated large bone lesion databases, but most are heterogeneous mixtures of both malignant and benign lesions that lack specific operative details, and none to date have been from a significantly diverse suburban area comparable to New York.[6-12] Establishing a sound differential diagnosis and early referral to a musculoskeletal oncologist in the primary care and family medicine setting is the first step in management, thereby minimizing potential long-term sequelae that may result in osteoarticular deformities, leg-length discrepancies, fractures, and/or gait abnormalities.[2]

In this retrospective study, we aimed to create a large database of patients dedicated solely to benign bone lesions with a focus on operative versus nonoperative management to elucidate the demographic data and management patterns of patients in a suburban New York hospital system, thereby adding to the existing body of knowledge of these complex lesions globally. With our database, we addressed the following questions: 1) What are the demographic patterns of patients with benign bone lesions in suburban New York? 2) How often are patients undergoing surgery? and 3) Are these trends in agreement with globally reported data?

Materials and Methods

Data acquisition

This was a single institution, multicenter retrospective observational study, approved by our institutional review board, of patients diagnosed with a benign bone lesion during the period from 2007 to 2021. The following 9th and 10th International Classification of Disease codes (ICD9 and ICD10) were included: D16.0-D16.9, M85.0, M85.4-M85.6, D48.0, 213.7, 213.9, 733.9. Inclusion criteria included patients of all ages with a diagnosis of a benign bone lesion. Exclusion criteria included all malignant bone tumors (except atypical cartilaginous neoplasms), soft tissue tumors, or tumors involving the skull, or maxillofacial bones.

Statistical analysis

Descriptive analyses included frequency data for the variables gender, anatomic location, age (categorized within brackets of 1-18 years old, 19-40 years old, and greater than 40 years old), diagnosis, treatment (categorized as operative vs nonoperative), and laterality of lesion. To investigate how these variables related to each other, cross-tabulations (CROSSTABS) with a bootstrapping procedure were performed. All analyses were conducted using SPSS (IBM SPSS Statistics Version 28.0.0.0). Two-tailed tests with *P*- values of. 05 were used to establish statistical significance.

Results

A total of 689 patients met inclusion criteria: 391 (56.7%) patients were male and 298 (43.3%) were female. The most common diagnoses were osteochondroma (47.0%), followed by enchondroma (12.9%), and osteoid osteoma (9.4%) [Figure 1]. A total of 380 patients ranged from 1 to 18 years old (55.2%), 167 from 19 to 40 (24.2%), and 142 were greater than 40 (20.6%). Benign bone lesions were more commonly found in males under 18 and in females over 40. As expected, aneurysmal bone cysts (ABCs), nonossifying fibromas (NOFs), osteochondromas, and solitary bone cysts (SBCs) were more commonly found in patients under 18. Giant cell tumors (GCTs) were more commonly found over age 18 and enchondromas were more commonly seen over age 40. The distal femur was the most common location (185/26.9%), followed by the proximal tibia (99/14.4%), proximal femur (77/11.2%), and proximal humerus (77/11.2%) [Figure 2]. 195 (28.3%) had nonoperative management, and 494 (71.6%) had surgery [Figure 3]. Patients over the age of 40 were less likely to undergo surgery than younger patients. As expected, the most common diagnoses undergoing surgery were, ABC, GCT, and osteochondroma, with diagnoses of enchondroma, NOF, and osteoid osteoma more frequently managed nonsurgically. All of these findings were statistically significant ($P < .05$).

Discussion

This study characterized the epidemiology of benign bone lesions in the suburban population of New York based on lesion type, location, and management. We believe the results of this study will serve to aid primary care providers in early
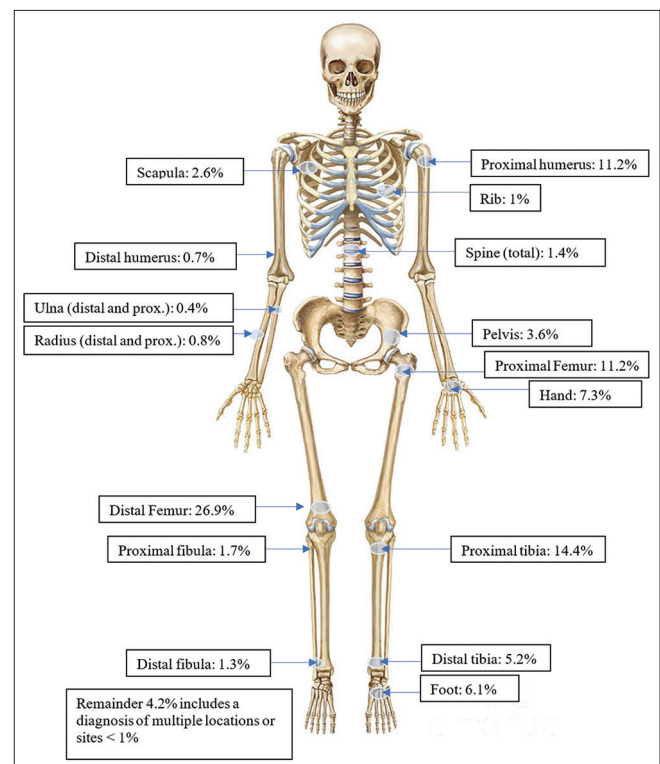
Figure 1: Anatomic location and frequency of benign bone lesions. Most common locations included the distal femur (26.9%) and proximal tibia (14.4%)
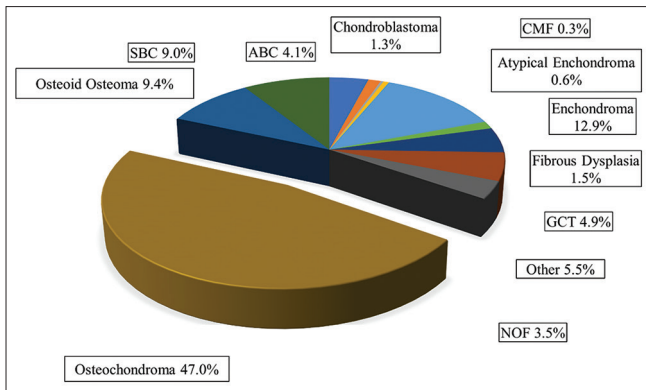
Figure 2: Diagnosis frequency. Three most common diagnoses were osteochondroma (47.0%), enchondroma (12.9%), and osteoid osteoma (9.4%)

recognition, raising awareness, and empowering them to offer initial counseling to affected patients. Operative management was more frequent than nonoperative management, likely owing to an inherent selection bias of symptomatic patients rather than incidentally found lesions that may otherwise have never presented for evaluation. ABCs, GCTs, and osteochondromas underwent surgery more frequently than enchondromas, NOFs, and osteoid osteomas, presumably due to more active/aggressive bony involvement in the former as compared to the latter group. Older patients were less likely to undergo operative treatment than younger patients, which is expected, given that these lesions are more active during skeletal growth and generally reach stability in adults. Osteochondromas were the most frequently diagnosed benign bone lesion, with benign bone lesions in general seen more commonly in younger males. As expected, the distal femur

			non-op	operation	Total
Diagnoses Coded	Aneurysmal Bone Cyst	Count	3	25	28
		Expected Count	7.9	20.1	28.0
		Adjusted Residual	-2.1	2.1	
	Chondroblastoma	Count	0	9	9
		Expected Count	2.5	6.5	9.0
		Adjusted Residual	-1.9	1.9	
	Chondromyxoid Fibroma	Count	0	2	2
		Expected Count	.6	1.4	2.0
		Adjusted Residual	-.9	.9	
	Chondrosarcoma	Count	0	4	4
		Expected Count	1.1	2.9	4.0
		Adjusted Residual	-1.3	1.3	
	Enchondroma	Count	35	54	89
		Expected Count	25.2	63.8	89.0
		Adjusted Residual	2.5	-2.5	
	Fibrous Dysplasia	Count	2	8	10
		Expected Count	2.8	7.2	10.0
		Adjusted Residual	-.6	.6	
	Giant Cell Tumor of Bone	Count	4	30	34
		Expected Count	9.6	24.4	34.0
		Adjusted Residual	-2.2	2.2	
	Nonossifying Fibroma	Count	16	8	24
		Expected Count	6.8	17.2	24.0
		Adjusted Residual	4.2	-4.2	
	Osteochondroma	Count	59	265	324
		Expected Count	91.7	232.3	324.0
		Adjusted Residual	-5.5	5.5	
	Osteoid Osteoma	Count	48	17	65
		Expected Count	18.4	46.6	65.0
		Adjusted Residual	8.6	-8.6	
	Other	Count	9	29	38
		Expected Count	10.8	27.2	38.0
		Adjusted Residual	-.7	.7	
	Solitary Bone Cyst	Count	19	43	62
		Expected Count	17.5	44.5	62.0
		Adjusted Residual	.4	-.4	
Total		Count	195	494	689
		Expected Count	195.0	494.0	689.0

Figure 3: Operative vs. nonoperative management according to diagnosis. 494 out of 689 patients underwent surgery, resulting in a 71.6% operative rate

and proximal tibia were the most common locations for lesions, owing to the greatest site of skeletal growth, which occurs about the knee.

The management of benign bone lesions is controversial and lacks consensus due to the multifactorial nature of these cases and the lack of large dedicated benign bone lesion patient databases. Two particularly important drivers of this lack of consensus are: 1) the inherent complexity and variability of cases and 2) the lack of large, representative databases of benign bone lesions that contain detailed information needed to create guidelines and definitively inform care. The dataset produced in the current study contains information on patient demographics, specific pathologic confirmed diagnosis for operative cases, anatomic location, and management of the lesion. Other databases in the literature are also valuable, but none have been solely dedicated to benign bone lesions in a suburban and diverse location similar to New York.

Van den Berg *et al.*'s 2008 study included similar variables to our study, such as age, diagnosis, sex, and anatomic location; however, they did not include data on how the lesions were managed and the patient's eventual outcome.[8] Their study produced similar findings to ours with regard to the epidemiology of benign bone lesions. Namely, osteochondromas were the most common bone lesions, overall incidence of benign bone lesions was higher in males than females, and specific lesion diagnoses were more or less common based on certain age brackets. One notable difference compared to our findings was that they found aneurysmal bone cysts to be the second most common type of benign bone lesion, as opposed to our study which found enchondroma to be the second most common diagnosis (albeit a distant second to osteochondroma).

In their 2016 study of 3,482 patients from Croatia, Bergovec *et al.* found that the average age of their patients was 33.5 years, with an even distribution with regard to sex.[9] Roughly, 80% of the lesions in their study were benign, with osteochondroma being the most frequently occurring, and simple bone cysts and enchondromas being the second and third most common occurring lesions. Although they did report on a large number of patients, they lacked any treatment data; therefore, no recommendations of management could be drawn. In addition, the social demographics of Croatia may not be representative of more diverse areas of the world such as suburban New York. Though we found enchondroma to be the second most common type of benign bone lesion and simple bone cysts to be the fourth most common (with osteoid osteoma being the third most common), their findings regarding incidence of lesion type are similar to ours. In contrast to their demographic findings, our study found that benign bone lesions were more commonly found in males and that the average patient age was 25 years old (both of these findings are more consistent with the current literature on the incidence of benign bone lesions).[1-3]

In their 2018 study, Toepfer *et al.* found that of 409 patients with musculoskeletal lesions of the foot and ankle included in their database, 266 lesions involved the bone, and of these, 87% were benign and 13% were malignant (a finding not dissimilar to what was found by Bergovec *et al.*).[9,10] They also reported that within the foot and ankle region, the most common benign bone lesions were simple bone cysts, enchondroma, and osteochondroma in descending order. Though osteochondroma is not the most common lesion in their database, those three lesion types are consistently reported as amongst the three most common benign bone lesions in other studies. The major weakness of their study was the lack of generalizability due to the strict inclusion of the foot and ankle only.

In Liu *et al.*'s 2021 study on 643 patients, the median age of patients with benign lesions was 18 years old, further validating our finding that bone lesions are most often diagnosed within the first three decades of life.[11] They also found that both benign and malignant lesions occurred more frequently in men and that the most common benign bone lesion was osteochondroma, followed by osteofibrous dysplasia, enchondroma, aneurysmal bone cysts, and lastly, simple bone cysts. Though not identical to our findings in term of prevalence order, their findings regarding lesion frequency were similar.

In his 2021 study in the setting of a Turkish hospital, Demirbas found that benign bone lesions occurred more frequently in younger males (with a mean age of roughly 30 years), osteochondroma was the most common diagnosis followed by enchondroma, and the most common anatomic location overall was around the knee joint.[12] All of these findings were similar to ours, further validating the current data set. In their 2022 review, "Benign Bone Tumors: An Overview of What We Know Today," De Salvo *et al.* discussed the epidemiology of the most common types of benign bone lesions based on the available published literature.[5] In accordance with our findings, they report that the most common lesion type is osteochondroma, followed by enchondroma. Other common benign bone lesion types reported included simple bone cysts, osteoma, osteoid osteoma, osteoblastoma, giant cell tumor, aneurysmal bone cyst, and fibrous dysplasia. Though the incidence across sex and the specific ages at which each lesion most often arises vary based on lesion type, they found that overall, benign bone lesions occurred more frequently in males and before the age of 30.

In their 2015 study comparing a large Chinese bone lesion database from Ji Shui Tan Hospital (JST) to the Dahlin's bone tumor cohort, Niu *et al.* concluded that there are significant differences in the epidemiology of bone lesions (both benign and malignant) across these large Chinese and US cohorts, with giant cell tumor and osteosarcoma having higher incidences in the Chinese population and Ewing sarcoma, chordoma, fibrosarcoma, myeloma, and malignant lymphoma having higher incidences in the US cohort.[7] In accordance with previously discussed literature and our study, benign bone lesions most frequently occurred about the knee in both the Chinese and US cohorts, roughly 63% of patients were under 30 years old, and the frequency was higher in males. The most common benign

lesion types in the Chinese cohort were osteochondroma, giant cell tumor, simple bone cyst, and fibrous dysplasia, whereas the four most common in the US cohort were osteochondroma, giant cell tumor, fibrous dysplasia, and chondroma. This finding contributes to the author's conclusion that there is variability in the epidemiology of benign bone lesions across different populations and is also different than the findings of our paper and the review article by De Salvo *et al.*,[5] which both found osteochondroma and enchondroma to be the most common benign bone lesions.

One limitation discussed in the Niu *et al.* study is that both the Mayo Clinic and the JST conclusions were based on hospital based rather than population-based data, which could skew the findings to artificially increase the incidence of lesion types or anatomic locations that are more likely to be symptomatic. It is important to note that this limitation also applies to our study as well. Another weakness of our study, which is similarly shared by the previous databases, is a lack of outcome data. Obtaining these data would be very helpful in determining best practice guidelines, however, and would be difficult to compare due to lack of uniformity for this heterogeneous group of conditions.

Our findings are generally consistent with previous published literature, suggesting that global epidemiologic trends of benign bone lesions are similar to suburban New York trends, further validating our cohort as being representative of the true distribution for these conditions. The relatively high operative rate of 71.6% is likely due to a selection bias for symptomatic or active cases which get preferentially referred for further management. Additionally, our operative rate could not be compared to other studies, given the lack of this parameter from other databases.

Conclusion

The data generated from nearly 700 patients with benign bone lesions in suburban New York appear to echo the current characterization of benign bone lesions in the literature from other global regions. The observations of this study therefore contribute to validating previous findings and have the potential to serve as a valuable resource for future studies investigating the relationship among patient age, gender, socioeconomic status, lesion location, lesion type, and management.

In this study, we have successfully created one of the largest recent databases dedicated solely to benign bone lesions in suburban New York with inclusion of treatment data. Benign bone lesions are common findings which must be managed appropriately to avoid unnecessary patient morbidity. The best way to ensure appropriate management is by first promoting awareness and education for the healthcare providers on the front line who often initially detect a bone lesion. Definitive care depends on a multitude of factors and must be individualized on a case-by-case basis. If there is any concern, patients should be referred to an orthopedic oncologist for further care. Future steps will include collecting subjective and objective long-term outcome data for interdiagnosis comparison, which could help establish clinical guidelines for management.

Financial support and sponsorship

Conflicts of interest

There are no conflicts of interest.

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