CURRENT CONCEPTS REVIEW

Primary and Metastatic Bone Tumors of the Patella: Literature Review and Institutional Experience

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Abstract

Background: Patellar tumors are rare but certainly must be considered in the differential diagnosis in patients with knee pain. Diagnosis can be challenging as often patellar neoplasms are confused with benign conditions and their clinical presentation is usually not specific. We performed an institutional and a literature review to determine what are the most common tumors affecting the patella and what is the best management.

Methods: This is a case series from our institution including all patients with benign, malignant, and metastatic patellar neoplasms. Charts were reviewed for patient demographics, clinical presentation, pathology characteristics, radiographic classification, and oncologic and functional outcomes.

Results: Twenty-four patients were identified; twelve patients had benign lesions, 10 metastatic and 2 primary malignant tumors. Chondroblastoma and Giant Cell Tumor were the most common tumors. Management of benign lesions with intralesional curettage and packing with bone graft or cement demonstrated excellent results with no local recurrence. In terms of malignant tumors, the spectrum of treatment is variable; it could range from medical management alone or in combination with surgical procedures to total patellectomy with reconstruction of the extensor mechanism.

Conclusion: Patellar tumors should be part of the differential in patients with chronic knee pain that does not respond to initial conservative interventions. Recurrence rate with intralesional curettage and bone grafting or cement packing is very low and therefore should be the treatment of choice for benign intraosseous neoplasms. Resection with negative margins in malignant neoplasms or bone metastasis decreases local recurrence but only in the former group there is a potential impact in survival.

Level of evidence: IV

Keywords: Patellar tumors, Metastatic bone lesions, Patella, Primary bone tumors, Knee tumors

Introduction

Benign and malignant patellar neoplasms are rare, and their clinical presentation is not specific, making their diagnosis challenging (1). Very often they are mistaken for other benign lesions and malignant lesions are often confused with benign conditions (1,2). Of all patellar neoplasms, the primary benign tumors comprise the vast majority of cases, while the malignant tumors and metastatic disease are far less common (3). The patella can be the source of a wide variety of tumors including Giant Cell Tumor of Bone

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(GCT), Chondroblastoma, Aneurysmal Bone Cyst and enchondroma (3). GCT is the most common representing approximately 30% of the cases followed by chondroblastoma which accounts for nearly 16% of the cases (1,3). The rest of the patellar tumors have relatively very low incidence rates. In terms of malignant tumors, the most common include osteosarcoma and chondrosarcoma (4)(5). Primary malignant visceral neoplasms with osseous tropism such as breast, thyroid, lung, prostate and renal cancer are expected to be the most common cause of patellar bone metastasis (6)(7). A slightly higher incidence might be observed with lung and renal cell carcinoma given their tendency to cause acrometastasis (metastasis distal to the elbow and the knee joints) (8).

To our knowledge, in the literature, the reports have been limited to case reports and very small case series with very little or no data regarding the optimal management of these lesions. Thus, we performed an institutional review of our experience with cases of primary and metastatic tumors of the patella as well as a literature review of all the published literature to determine: 1) What are the most common tumors in the patella and 2) What is the best surgical treatment for benign lesions, malignant and metastatic tumors in the patella.

Methods

Series of patients treated at our institution

Under Institutional Review Board (IRB) approval, we extracted from our musculoskeletal oncology registry database all patients who underwent surgical treatment for benign primary patellar tumors, malignant primary patellar tumors, and patellar metastatic lesions. Review time frame was from 1972 to 2017. The data of this case series was collected retrospectively and based on surgery, pathology, radiology, billing and clinical reports which were searched with a registry tool. A retrospective chart review was performed for patient demographics (age, sex, affected side, race), clinical presentation variables (duration of symptoms, level of pain, time to treatment, type of treatment including surgical and non-surgical interventions, history of trauma and ability to ambulate), histopathologic tumor characteristics (histology, grade, size, and metastatic presentation if malignant), radiographic characteristics according to the modified Lodwick radiologic classification(9), and oncologic outcomes including local recurrence free survival, metastatic disease-free survival, disease-specific survival, and overall survival.

Method of Literature Review

We also performed a literature review using PubMed and EMBASE search engines. We used the search terms “patella tumors”, “patellar tumors”, “patella neoplasms”, patellar neoplasms”, and “knee tumors”, retrieving 722 results written in English. After cross-reference comparison and removing all duplicates and title screening, we identified a total of 117 papers for review, including case reports, case series, abstracts and literature reviews. We used as time frame 1950 up to 2019.

After a full text screening of these 117 papers, we included 94 that had the minimum data necessary for this review which included: demographics; clinical presentation; tumor characteristics including histology, grade, recurrence after treatment and metastatic presentation if malignant; surgical management.

Results

Institutional Cohort: Characteristics

Twenty-four patients were identified in our institutional database, including 12 with primary benign tumors, 2 with primary malignant tumors, and 10 with metastatic lesions. Of the primary benign tumor group, seven patients were females and 5 were males. Average age at presentation was 37.25 years (Range: 23 to 66 years old). As expected, patients with metastatic disease were older, on average 48 years old (Median: 51 years old, Range: 8 to 69 years old), as were patients with primary malignant tumors, average 48 years old (Range: 34 to 61 years old). Tumor location for all comers was in the left patella for 13 patients (54%) and it was in the right for 11 patients (46%). In terms of race/ethnicity, most patients were Caucasian. [Tables 1-4]

Based on the modified Lodwick radiographic classification for bone tumors, we found that 5 patients had tumor type I (2 Type I-A and 3 Type I-B), 6 were found to be type II and 12 were found to be type III, one patient was excluded fromthis Classification since the tumor was described as an “exostosis” and not an intraosseous neoplasm [Table 4]. For the malignant tumors both patients had a Lodwick radiographic grade III at moment of presentation. The metastasis group patients were classified as Lodwick II (3 patients) and Lodwick III (7 patients; 2 type III-A and 5 type III-B) [Table 4]. The higher the biologic aggressiveness of the tumor, the higher the grade in the modified Lodwick classification was. Despite anatomic location within the patella being described as a marker of biologic behavior (benign bone tumor tumors are reported to occur more frequently in the superior quadrants of the patella), the anatomic locations of the lesions were variable, with 5 tumors presenting in the inferior pole or apex, 6 presenting in the superior pole, 2 in the medial aspect, and the other 2 in the lateral aspect of the patella. Those with malignant characteristics on imaging presented with a diffuse patellar involvement (9 patients). Metastatic disease patients had also inconsistent location and aggressive and diffuse involvement characteristics on radiographs [Table 4].

Institutional Cohort Results: Characteristics by Tumor Histology

Benign bone tumors

Our experience matches the literature. Local pain and swelling are the primary symptoms that patients describe. Given the benign nature of the tumor, these clinical findings increase along with prolonged time, which in many cases can be months [Table 1].

Table 1. Benign tumors of the patella: Institutional Experience

Diagnosis	Symptoms	Level of Pain (1-10)	Duration of symptoms until surgery	Gender	Age	Affected Patella	Radiologic Classification	Location	Trauma	Management	Recurrence
GCT	Pain+Swelling	--	4 m	M	36	L	III	N/E	Yes	Patellectomy	No
GCT	Pain+Swelling	--	24 m	F	23	L	I-B	Medial	Yes	Curettage+Packing	No
GCT	Pain+Swelling	3	12 m	M	32	L	I-B	Inferior pole	Yes	Patellectomy	No
GCT	Pain+Swelling	--	12 m	F	48	L	I	Superior	No	Patellectomy	No
GCT	Pain+Swelling	--	10 years	F	37	L	IIB	Sup/Inferior Pole	No	Curettage+BG	No
CBL	Pain+Swelling	--	15 m	M	32	R	II	Inferior Pole	No	Patellectomy	No
CBL	Pain	--	N/E	M	27	L	II	Superior Pole	N/E	Curettage+BG	No
CBL	Pain+Swelling	--	6 m	M	30	R	I-A	Inferior Pole	No	Curettage+BG	No
Ganglion Cyst	Pain+Swelling	--	N/E	F	40	R	III	Sup/Lateral Pole	No	Curettage+BG	No
Hemangioma	Pain+Swelling	--	12 m	F	35	R	I-B	Medial Pole	No	Curettage+BG	No
Angioleiomyoma	Pain	--	24 m	F	66	L	Exostosis*	Lateral Pole	No	Exostosis excision	No
Bone cyst	Pain	5	12 m	F	41	R	II	Superior Pole	Yes	Curettage	N/E

Abbreviations: BG: Bone Graft; CBL: Chondroblastoma; GCT: Giant Cell Tumor; N/E: No Evidence Note: Radiologic classification is based on Revised Lodwick Classification

Table 2. Malignant tumors of the patella: Institutional Experience

Diagnosis	Symptoms	Level of Pain (1-10)	Duration of symptoms until surgery	Gender	Age	Affected Patella	Radiologic Classification	Location	Trauma	Management	F/U in 1 Year	Recurrence	DFS
Chondrosarcoma	Pain+Swelling	5	27 m	F	34	R	III	Diffuse	Yes	Patellectomy	Yes	No	<3 Y
Synovial Sarcoma	Pain+Swelling	N/E		M	61	R	III	Diffuse	No	Patellectomy	Yes	No	< 10 Y

Abbreviations: DFS: Disease Free Survival; F/U: Follow Up; N/E: No Evidence Note: Radiologic classification is based on Revised Lodwick Classification

Table 3. Metastatic Tumors of the patella: Institutional Experience

Primary Tumor	Symptoms	Duration of symptoms until management	Gender	Age	Affected Patella	Radiologic Classification	Location	Time to MT	Management	F/U in 1 Year	DFS
Lung. AC.	Pain	30 days	M	51	L	III	N/E	1 m	Radiotherapy	No	<3 Y
Lung. AC	Pain	5 weeks	M	52	L	IIIB	Diffuse	12 m	Patellectomy	No	N/E
Lung.AC	Pain	5 months	M	67	R	II	Inferior pole	5 m	En Block Resection	Yes	<3 Y
Lung. AC	Pain	12 months	F	69	L	N/E	N/E	12 m	Radiotherapy	Yes	<3 Y
Melanoma	Swelling	24 months	M	64	R	II	Diffuse	10 m	Radiotherapy	Yes	<3 Y
Breast	Pain	--	F	51	R	IIB	Superior pole	16 m	None	Yes	N/E
Osteosarcoma	Swelling	2 months	M	8	L	III	Diffuse	2 m	Amputation	Yes	<3 Y
E.M.C.	Pain	14 months	M	37	L	III	Diffuse	14 m	Amputation	Yes	> 10 Y
Fibromyxoid Sarcoma	Pain	26 months	M	44	R	III	Diffuse	26 m	Tibial Turn-up	Yes	<10 Y
Lymphoma	Pain	17 days	F	40	L	II	Inferior Pole	1 m	Patellectomy	Yes	<10 Y

Abbreviations: AC: Adenocarcinoma; DFS: Disease Free Survival; E.M.C.: Extra-skeletal myxoid chondrosarcoma F/U: Follow Up; N/E: No Evidence.
Note: Radiologic classification is based on Revised Lodwick Classification

Table 4. Lodwick Classification

Radiologic Classification (n)	Pooled Cohort	Benign	Malignant	Metastatic
I-A	2	2	NA	NA
I-B	3	3	NA	NA
II	6	3	NA	3
IIIA	3	1	NA	2
IIIB	9	2	2	5
Affected Patella, % (n)				
Right	45.8% (11)	41.7% (5)	100% (2)	40% (4)
Left	54.2% (13)	58.3% (7)	0%	60% (6)
Location (n)				
Superior Pole	6	5	0	1
Inferior Pole	5	3	0	2
Medial Pole	2	2	0	0
Lateral Pole	2	2	0	0
Diffuse	9	0	2	5

Giant Cell tumor of Bone

We identified 5 patients with Giant Cell Tumors of Bone (GCT). Two were male and 3 were female. Average age at presentation was 35 years of age (Median: 36 years old, Range: 23 to 48 years old.) As expected, this tumor was more prevalent in females. GCT often presents with swelling, joint effusion, tenderness and redness of the knee, diminished range of motion and pain with active and passive motion. Diagnosis can be challenging on occasion as infection is usually considered first in the differential. Three out of the five patients had history of trauma in the affected patella [Table 5]. The average time between onset of symptoms and treatment was about 13 months. The pain was described between 3 and 5 in a pain scale from 1 to 10. Two patients were treated with total patellectomy due to tumor extension. Two were treated with curettage and cement packing, and one with curettage and allograft bone packing due to younger age. None of the patients experienced local recurrence, metastatic disease to the lungs or bone, or died because of the disease.

Table 5. Trauma and Management

Trauma (n)	Pooled Cohort	Benign	Malignant	Metastatic
Yes	9	4	1	4
No	16	9	1	6
Management (n)				
Curettage	8	8	0	0
Patellectomy	6	4	2	3
Bone Graft	5	5	0	0
Packing	2	2	0	0
Excision	1	1	0	0
Resection	1	0	0	1
Radiation	1	0	0	2
Amputation	0	0	0	2

Chondroblastoma

Three patients presented with chondroblastoma, all of them reported mild to moderate pain with active motion. There was no history of trauma on the affected patella. The average time between onset of symptoms and treatment was about 16 months. Two patients were treated with curettage and allograft bone packing and 1 patient was treated with patellectomy given the size of the tumor and close involvement of the subchondral bone. None of these patients presented with local recurrence after treatment. None of the patients had metastatic lung disease or died of the primary disease. As expected, this tumor was identified more commonly in male patients of younger age (Average: 29 years old, Median: 30, Range: 27 to 32 years old).

Hemangioma

We reported a rare case of hemangioma of bone and one Angioleiomyoma arising from the patella. Clinical presentations were similar to other patellar benign neoplasms: pain and swelling. Both patients were treated with excision and curettage and bone grafting. None of the patients reviewed reported local recurrence after treatment.

Others benign tumors

Aside from the most common tumors that we have mentioned, we also identified a case of an intraosseous ganglion cyst. This patient presented with pain with active motion. Ambulation was diminished due to pain.

This patient underwent excision and curettage with allograft bone packing. There was no local recurrence after treatment. A bone cyst patient is also reported; this patient had history of trauma aroundthe patella, long standing pain (5/10) and was managed with excision and curettage. No local recurrence was reported after treatment. The mean time since the onset of symptoms until surgery was 22 months (range 4 to 100 months).

Malignant bone tumors

Chondrosarcoma

A 34 years old female patient presented with pain with ambulation. The pain was described as a level 5 on a 1-10 Likert pain scale. The patient did not have history of trauma but had as significant risk factor a concurrent diagnosis of Maffucci syndrome. She was treated with an above the knee amputation due to tumor intra-articular, femoral, tibial and fibular diffuse extension and morbid large hemangiomas in the lower leg. Imaging studies revealed a complete loss of anatomy of the lower limb bones. She subsequently presented with multifocal chondrosarcoma sites after surgery, as well as innumerable lytic lesions with soft-tissue component and disseminated lung metastases. Subsequently, an expansive lesion in the right iliac bone was also identified along with intraabdominal and pelvic organs involvement. Disease free survival was 16 months after treatment [Table 2].

Synovial sarcoma

A 60 year-old male patient with synovial sarcoma presented to the office complaining of knee pain and swelling. First, he underwent arthroscopy for mass excision originating from bone where pathology was positive for "Spindle cells grade 2 out of 3 most likely Synovial Sarcoma". Subsequently the patient was treated with post-operative radiation and chemotherapy. Due to post-radiation articular degeneration the patient underwent a total knee replacement. Due to multiple postoperative infections the patient underwent insertion of an antibiotic spacer after explantation and a two-stage reconstruction in multiple occasions. Ultimately, he had an above the knee amputation due to a recalcitrant periprosthetic joint polymicrobial infection.

Metastatic Tumors

We identified 10 cases of metastatic disease to the patella treated at our institution, including 7 patients (70%) who were male and 3 (30%) who were female. The average age was 48.3 years old (range 8-69). The location of the tumor was 6 (60%) in the left patella and 4 (40%) in the right. The primary tumors included lung adenocarcinoma (4), Melanoma (1), Breast carcinoma (1), Metastatic Osteosarcoma (1), lymphoma (1) and Myxoid Liposarcoma (2). The clinical presentation was predominantly pain.

The patients underwent different treatments ranging from only radiation to wide resection including partial patellectomy. Of the 4 patients treated non-operatively, 2 had metastatic lung cancer, 1 had melanoma and 1 had breast cancer. All these patients received palliative radiation of the affected knee due to poor prognosis and terminal disease. Two cases underwent above knee amputations (AKA): one case of oligometastatic Osteosarcoma and one of oligometastatic Myxoid Liposarcoma. We also report a patient with a fibromyxoid sarcoma which was treated with a Tibial turn-up due to massive intra-articular infiltration. Two patients with metastatic lung cancer (Adenocarcinoma sub-type) were treated with "en bloc resection" along with a hinged Total Knee Replacement due to concomitant severe osteoarthritic disease along with bone loss. We also report a rare case of lymphoma with extension in to the patella; this patient underwent patellectomy after failed radiation [Table 3]. There was no specific quadrant location in the patella for metastatic disease. Most patients presented with a tumor radiographic type II or type III according to the Ludwick classification [Table 4].

Literature Review Outcome: how it compares to our data

A total of 94 publications were suitable for review. They included a total of 254 patients of which 85 were females (34 %) and 169 were males (66 %). A hundred ninety-two (75%) patients had a benign primary bone tumor with the majority being males n=129 (67%). Average age 29 years old (Median: 24 years-old, Range: 9 to 67 years-old). Thirty eight patients (15%) had a malignant primary bone tumor, average age 44 years old (range 17 to 77 years old) and 24 (10%) had metastatic lesions, average age 61 years old (range 13 to 86 years old).

Combining our experience and the review, it was observed that local pain and swelling are the primary symptoms that the patients describe.

Benign Tumors

In benign tumors, pain and swelling, increase along with time, which in many cases, can last for months. GCT often presents with swelling, effusion, tenderness and redness of the knee, diminished range of motion, and pain to active and passive motion. In our review of the published literature, 57 cases of giant cell tumors have been reported, with a mean follow up of 46 months. Only 2 patients reported to experience local recurrence, one after patellectomy(10) and one after curettage and bone grafting(11). Sixty-five cases of chondroblastoma have also been reported with a higher rate of local recurrences n=4 (6%) after curettage and bone grafting with an average of 25 months of follow-up. Compared to our series, chondroblastoma patients reported in the published literature match the traditional 13 to 15 year-old male patient with knee pain (known by some authors as the Bar-mitzvah tumor). In our series, patellar chondroblastoma patients were older, 20 to 30 years of age instead. The third most commonly-reported benign tumor was the aneurysmal bone cyst with 18 cases identified in our review. This group of patients did not experience local recurrence after an average follow up of 26 months. These tumors may present with either swelling without other inflammatory

symptoms or just pain.

Despite the literature describing the incidence of GCT and Chondroblastoma local recurrence around 25%(12–14), our experience and review of the literature revealed a much lower recurrence rate when these tumors occur in the patella. An extensive curettage with allograft or cement packing or less likely, patellectomy, were the main treatment modalities on these patients. [Table 6]

Malignant Tumors

Based on the literature(15–17), osteosarcoma is the most common cause of primary malignant tumors on the patella but represents only 6% of the total of the patella tumors. Eleven cases have been reported (7 were Males and 4 were females) with an average age of 35 years. (Range: 18 to 54 years old). The clinical presentation of these tumors is pain and swelling of rapid progression. Four of these patients underwent patellectomy and other 4 needed amputations due extensive intra-articular extension. All resections had negative margins.

We did not identify any case of an osteosarcoma as a primary malignant tumor of the patella in our series.

Chondrosarcoma is the second most common type of malignant tumor in the patella, accounting for 20% to 27% of the malignant tumors of the patella.(5,18) However, only 4 cases have been reported in the literature so far. Three of these patients were female and 1 male with an average age of 55 years of age (Range: 32 to 68 years old). Chronic, dull pain of the knee was the main cause of consultation. No recurrence was reported in these patients after being treated with patellectomy within an average of 31 months of follow-up.

There are also two cases reported of Ewing's Sarcoma of the patella in a 26 year old female and a 41 years old male (19)(20) who presented to the clinic with pain and swelling of rapid evolution. The first patient, a 26 years old female, was treated with patellectomy in combination with neoadjuvant and adjuvant chemotherapy with no signs of local recurrence after a follow up of 24 months. The 41 year old male patient was treated with chemotherapy with a combination of vincristine, Adriamycin and cyclophosphamide (VAC).

Other malignant tumors have also been reported, including lymphoma, the previously known Malignant Fibrous Histiocytoma (MFH) and Angiosarcoma. [Table 7].

Metastatic Tumors

The review of the literature demonstrates that around 50% of the patellar metastasis come from a primary lung carcinoma (21–24), Following the expected osseous tropism of lung malignancies(8). Ten cases of metastatic lung carcinoma to the patella have been reported, 8 were males and 2 females with an average age of 67 years of age (Range: 51 to 86 years old). Clinically, these patients presented with knee pain and respiratory symptoms. Our patients varied regarding the type of lung carcinoma where 4 patients had adenocarcinoma.

All patients had a history of smoking. Most of the patients succumbed to the lung disease not too long after the diagnosis; mean follow-up was 6 months (Range: 3 to 12 months). For the primary malignant tumors, procedures capable to offer negative margins (total patellectomy, wide resection, and in block resection) are the recommended treatment options taking into consideration patient function. Chemotherapy is useful in the adjuvant and neoadjuvant setting as well as for management of metastatic disease. Radiotherapy post-surgery might also be found to be helpful, especially if an intra-lesional procedure is needed for palliation in patients with poor prognosis and health status. Radiation in this setting helps to improve local control. Radiation can also be used a single therapy in the setting of palliation according to the radiosensitivity of the tumor.

Renal Cell Carcinoma is another type of tumor reported to metastasize to the patella (25–28). From a total of five patients, four were male and 1 female with an average age of 62 years of age (49 to 74 years old). Pain was the chief complain at the time of presentation. Average follow-up was 10 months.

Carcinoma of the breast is very uncommon to metastasize in the patella, corresponding to 2% of the total patellar metastasis. On imaging studies, the patella presented with a lytic destructive lesion similar to the presentation on the literature (21). In just 2 cases that have been documented so far, the age, the symptoms at presentation, and the treatment were similar to our experience (29,30). Both females, average age 59 years old (Range: 48 to 70). Chronic, long standing knee pain was the main symptom at the moment of presentation.

Malignant melanoma is a well-known tumor for this capacity to metastasize to bone.

Table 6. Benign Tumors of the Patella. Literature Review

Author	Year	Type of study	Tumor	No. Pts	Gender Distribution	Age	Symptoms	Duration	Treatment	Follow Up (NED)	Recurrence
Cameron GW[43]	1955	Case Report	GCT	1	M	35	Pain	1 y	NM	NM	NM
Henelt et al[44]	1961	Case Report	GCT	1	F	24	Pain	1 y	Patellectomy	14 m	No
Copeland et al[45]	1966	Case Report	ABC	2	M	~26	Pain+Swelling	~1 y	Patellectomy	~48 m	No
Lammot TR[46]	1968	Case Report	ECh	1	F	20	Pain+Swelling	1 m	Patellectomy	NM	NM
Bansal et al[47]	1974	Case Series	HG	2	1 M 1 F	27	Pain+Swelling	4 y	Patellectomy	1.5 y	No
Lewis & Bullough[4	1976	Case Report	CBL	1	M	20	Pain+Swelling	2 y	Patellectomy	6 y	No

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Wilson et al[49]	1976	Case Report	GCT	1	F	56	Pain+Swelling	2 y	En block resection	NM	NM
Wientroub et al[50]	1979	Case Report	SBC	1	M	52	Pain	2 y	Patellectomy	NM	No
Gottschalk et al[51]	1985	Case Series	CBL	3	2 M 1 F	~23	Pain+Swelling	~5 m	Patellectomy	12 m	Yes
Moser et al[52]	1988	Case Series	CBL	16	13 M 3 F	~23	Pain	NM	NM	NM	NM
De Coster et al[53]	1989	Case Report	OB	1	M	29	NM	NM	NM	NM	NM
Kransdorf et al [54]	1989	Case Series	CBL	16	13 M 3 F	23	Pain	NM	NM	NM	NM
			GCT	8	6 M 2 F	26	Pain	NM	NM	NM	NM
			SBC	6	5 M 1 F	25	Pain	NM	NM	NM	NM
			HG	3	2 M 1 F	15	Pain	NM	NM	NM	NM
			Och	2	M	46	Pain	NM	NM	NM	NM
			LP	2	1 M 1 F	16	Pain	NM	NM	NM	NM
			OB	1	F	25	Pain	NM	NM	NM	NM
Bulas et al[55]	1992	Case Report	OO	1	F	16	Pain	4 m	Resection	NM	NM
Pevny et al[56]	1994	Case Report	ABC	1	F	15	Pain+Swelling	3 m	Curettage	NM	NM
Saglik et al[57]	1995	Case Report	SBC	1	F	33	Pain+Swelling	5m	Patellectomy	11 m	No
Wolfe et al [58]	1995	Case Report	CBL	1	F	13	Pain+Swelling	8 m	Curettage+BG	18 m	No
Castro et al[59]	1996	Case Report	ABC	1	M	NM	NM	NM	Curettage	NM	NM
Tam et al[60]	1996	Case Report	G	1	M	78	Pain+Swelling	NM	Curettage	NM	NM
Ferguson et al[38]	1997	Case Series	GCT	5	2 M 3 F	~24	Pain+Swelling	NM	Curettage	~35m	No
			CBL	1	1 M	19	Pain+Swelling	NM	Curettage	28 m	No
Ferguson et al[38]	1997	Case Report	CBL	1	M	19	Pain+Swelling	NM	Curettage+BG	28 m	NM
Connell et al[10]	1998	Case Report	GCT	1	M	44	Pain	NM	Patellectomy	10 m	Yes
Ghekiere et al[61]	1998	Case Report	CBL	1	F	11	Pain+Swelling	1 y	Curettage +BG	60 m	Yes
Obrebski et al[62]	1998	Case Report	GCT	1	M	26	Pain	NM	Patellectomy	12 m	No
Chaudhary et al[63]	2000	Case Report	SBC	1	F	24	Pain+Swelling	6 m	Patellectomy	3 y	No
Macdonald et al[64]	2001	Case Report	GCT	1	F	36	Swelling	~10 y	Patellectomy	15 m	No
Mercuri et al[2]	2001	Case Series	GCT	6	2 M 4 F	~31	Pain+Swelling	~7 m	Patellectomy Curettage	~ 20 y	No
			G	2	2 M	~3	Pain+Swelling	~7 m	Curettage	14 y	No
			OO	2	2 M	0	Pain+Swelling	~7 m	Excision	43 y	No
			Och	1	1 F	13	Pain+Swelling	~7 m	Excision/Flap	44 y	No
			CH	1	1 M	56	Pain+Swelling	~7 m		51 y	No
			ABC	2	2 M	54	Pain+Swelling	~7 m	Curettage/Patellectomy	~14 y	No
			CBL	8	6 M 2 F	42 19	Pain+Swelling	~7 m	Curettage/Patellectomy	~12 y	No
Shen et al[65]	2001	Case Report	OB	1	M	34	Pain+Swelling	NM	Curettage	2 y	No
Trebse et al[66]	2001	Case Report	CBL	1	M	24	Pain	5 y	Curettage+BG	30 m	No
Agarwal et al[67]	2002	Case Series	GCT	11	8 M 3 F	~24	Pain	~5 m	Patellectomy	63 m	No
Koos & Than[68]	2005	Case Report	OO	1	F	17	Pain	2 y	En-bloc resection	2 y	No

Marudanayagam et al[69]	2006	Case Report	ABC	1	M	20	Pain+Swelling	3 y	Curettage	24 m	No
Ofluoglu & Donthineni [11]	2007	Case Report	GCT	1	F	22	Pain+Swelling	4m	Curettage and BG	4.5 y	Yes
Oh JH et al[70]	2007	Case Report	ABC	1	M	30	Pain+Swelling	10 m	Curettage	24 m	No
Bhagat et al[71]	2008	Case Series	GCT	2	1 M 1 F	~63	Pain+Swelling	6 m	Patellectomy	5 y	No
			OB	1	1 M	38	Pain+Swelling	6 m	Excision	4 y	No
			Myl	1	1 M	13	Pain+Swelling	6 m	Radiotherapy	3 y	No
			CBL	2	2 M	29	Pain+Swelling	6m		2 y	No
			OFC	1	1 F	57	Pain+Swelling	6m	Curettage Excision	3 y	No
Chakravertty & Chakravertty[72]	2008	Case Report	GCT	1	F	23	Swelling	4 y	Patellectomy	4 y	No
Gudi et al[73]	2008	Case Report	CBL	1	M	24	Pain+Swelling	1 y	Patellectomy	24 m	No
Malhotra et al[39]	2009	Case Report	GCT	1	F	37	Pain	6 m	Wide resection	36 m	No
Reddy & Sathi[74]	2009	Case Report	ABC	1	F	27	Pain+Swelling	9 m	Curettage	4 y	No
Singh et al[75]	2009	Case Series	GCT	11	8 M 3 F	34	NM	NM	NM	NM	NM
			CBL	9	8 M 1 F	23	NM	NM	NM	NM	NM
			Ch	1	M	13	NM	NM	NM	NM	NM
			CMF	1	M	41	NM	NM	NM	NM	NM
			OO	1	M	9	NM	NM	NM	NM	NM
			ABC	6	3 M 3 F	29	NM	NM	NM	NM	NM
			OFC	3	F	49	NM	NM	NM	NM	NM
			SBC	2	1 M 1 F	17	NM	NM	NM	NM	NM
			G	2	F	67	NM	NM	NM	NM	NM
			O	2	1 M 1 F	42	NM	NM	NM	NM	NM
Balke et al[76]	2010	Case Report	ABC	1	M	56	Pain	2 m	Curettage	16 m	No
Ozan & Toker[77]	2010	Case Report	CBL	1	M	26	Pain+Swelling	3 y	Curettage+BG	~ 2 y	No
Ma et al[78]	2011	Case Series	OO	2	M	22	Pain+Swelling	1.5 y	Curettage	3 y	No
Traore et al[79]	2011	Case Report	ABC	1	F	28	Pain	1 y	Curettage	31 m	No
Yoshida et al[14]	2012	Case Report	GCT	1	M	39	Pain	2 w	Curettage	5 m	No
Kaymaz et al[80]	2014	Case Report	CH	1	M	16	Pain	3 m	Excision	3 m	No
Tan et al[81]	2014	Case Report	CBL	1	M	35	Pain	2 y	Curettage+BG	1 y	No
Shibata T et Al[82]	2015	Case Report	GCT	1	F	25	Pain	1 y	Curettage	16 m	No
Baptista et al[37]	2016	Case Series	GCT	3	2 M 1 F	~27	Pain+Swelling	NM	Partial Patellectomy	NM	No
			CBL	1	1 M	17	Pain+Swelling	NM	Curettage	NM	No
			OFC	1	1 F	41	Pain+Swelling	NM	Curettage	NM	No
			OO	1	1 M	12	Pain+Swelling	NM	Partial	NM	No
			ABC	1	1 M	19	Pain+Swelling	NM	Patellectomy	NM	No
Cetinkaya M[83]	2016	Case Report	ABC	1	F	32	Swelling	NM	Curettage Patellectomy	22 m	No
Lang et al[84]	2019	Case Report	CBL	1	M	15	Pain	NM	Curettage	21 m	No

We presented one case in our series with similar presentation to that four cases reported in the literature: swelling and pain due to a pathologic fracture. According to the literature, the management of melanoma is still controversial (21,31). In our experience, we treated the patient with ORIF and postoperative radiotherapy. The cases reported in the literature were 4 in total, 2 male and 2 females, average age of 44 years (Range 13 to 52 years-old). Pain was the chief complaint of these patients (32–34). These patients were treated with chemotherapy and patellectomy. With the advent of effective chemotherapy, the role of treatment in bone metastasis secondary to melanoma has decreased being limited to lesions that required biomechanical stabilization while medical response to treatment is awaited [Table 8].

Table 7. Malignant Tumors of the Patella. Literature Review

Author	Year	Study	Diagnosis	No. Pts	Gender	Age	Symptoms	Duration	Treatment	Follow Up	Recurrence
Goodwin et al[15]	1961	Case Report	OS	1	M	24	Pain+Swelling	5 m	Amputation	6 m	NM
Kransdorf et al[54]	1989	Case Series	HE	1	M	20	Pain	NM	NM	NM	NM
			L	3	2M 1F	49	Pain	NM	NM	NM	NM
					1						
Lopez-Barea et al[85]	1991	Case Report	MFH	1	F	84	Pain	5 m	Resection	NM	NM
Nagai et al[16]	1993	Case Report	OS	1	F	34	Pain+Swelling	8 m	Patellectomy	NM	NM
Okada et al[86]	1994	Case Report	OS	1	M	54	Pain	3 m	Patellectomy	24 m	No
Roger et al[87]	1994	Case Report	MFH	1	M	22	Pain+Swelling	6 m	Lower Leg Amputation	18 m	No
Ferguson et al[31]	1997	Case Series	MFH	1	1 M	17	Pain+Swelling	NM	Patellectomy	9 m	DOD
			OS	1	1 F	18	Pain+Swelling	NM	Resection/Prosthesis	25 m	No
Boyle & Walton[88]	2000	Case Report	CS	1	F	32	Pain	3 y	Patellectomy	72 m	No
Chandra & Eilender[89]	2000	Case Report	L	1	F	52	Pain	NM	Chemotherapy	13 m	No
Agarwal et al[90]	2001	Case Report	L	1	M	72	Pain	NM	Excision	NM	NM
Inoue et al[91]	2001	Case Report	LMS	1	F	59	Pain+Swelling	10 y	Patellectomy	20 m	No
Mercuri et al[2]	2001	Case Series	HE	5	1 F 4 M	~38	Pain+Swelling	7m	Patellectomy/Radiotherapy	~11 y	No
			AS	1	1 M	52	Pain+Swelling	7m	Radiotherapy	~11 y	No
			P	1	1 F	51	Pain+Swelling	7m	Patellectomy	~11 y	No
			L	1	1 M	53	Pain+Swelling	7m	Amputation/Radiotherapy	~11 y	No
McGrath et al[92]	2006	Case Report	OS	1	M	53	Pain+Swelling	3 m	Amputation	15 m	NM
Kumagai et al [18]	2008	Case Report	CS	1	F	64	Pain+Swelling	4 y	Patellectomy	20 m	No
Shehadeh et al[93]	2008	Case Report	OS	1	M	22	Pain+Swelling	NM	Amputation	NM	NM
Cho et al[17]	2009	Case Report	OS	1	F	53	Pain+Swelling	3 y	Patellectomy	26 m	No
Singh et al[75]	2009	Case Series	OS	2	M	18	Pain+Swelling	NM	NM	NM	NM
			MFH	1	M	42	Pain+Swelling	NM	NM	NM	NM
			CCCS	1	M	25	Pain+Swelling	NM	NM	NM	NM
			L	1	M	77	Pain+Swelling	NM	NM	NM	NM
			PS	1	M	70	Pain+Swelling	NM	NM	NM	NM
Chida et al[94]	2012	Case Report	OS	1	F	30	Pain	2 y	Amputation	48 m	No
Gorelik et al[20]	2013	Case Report	ES	1	M	41	Pain+Swelling	4 m	Chemotherapy	7 m	Yes
Aoki et al[4]	2014	Case Report	OS	1	M	47	Pain+Swelling	1 y	Patellectomy	6 m	No
Valsalan et al[19]	2015	Case Report	ES	1	F	26	Pain+Swelling	2 m	Patellectomy	24 m	No
Ye et al[5]	2017	Case Report	CS	1	F	68	Swelling	6 m	Patellectomy	3 m	No

Table 8. Metastatic Tumors of the Patella. Literature Review

Author	Year	Study	Primary Tumor	No. Pts	Gender	Age	Symptoms	Duration until MT	Treatment	Follow Up	Recurrence/Deceased
Taylor G H[30]	1964	Case Report	Breast	1	F	70	Pain+Swelling	NM	Patellectomy	NM	NM
Benedek TG[95]	1965	Case Report	Lung. SCC	1	M	66	Swelling	5 m	None	5 m	Deceased
Klenerman L[29]	1965	Case Report	Breast	1	F	48	Pain	5 y	Patellectomy	3 m	Deceased
Stoler et al[33]	1969	Case Report	Melanoma	1	M	48	Pain+Swelling	NM	None	NM	NM
Kwa & Nade[96]	1989	Case Report	RCC	1	F	64	Pain	NM	NM	NM	NM
Pazzaglia et al[97]	1989	Case Report	Lung. SCC	1	M	71	Pain	NM	NM	NM	NM
Jaeger et al[31]	1992	Case Report	Melanoma	1	F	65	Pain	NM	Patellectomy	3 w	Deceased
Aktas et al[22]	1998	Case Report	Lung. SCC	1	M	55	Pain+Swelling	3 m	Patellectomy	8 m	Deceased
Ganjoo et al[98]	1999	Case Report	Lung. AC	1	M	65	Pain+Swelling	2 m	Radiotherapy	5 m	Deceased
Warner et al[28]	1999	Case Report	RCC	1	M	NM	NM	NM	NM	NM	NM
Cooper & Mess[41]	2000	Case Report	Lung. AC	1	M	51	NM	12 m	Curettage	NM	NM
Mercuri et al[2]	2001	Case Series	OS	1	M	21	Pain	NM	Knee	NM	Deceased
Sun et al[24]	2001	Case Report	Lung. SCC	1	M	76	Pain+Swelling	5 m	Patellectomy	3 m	Deceased
Siddiqui et al[99]	2003	Case Report	Lung. AC	1	F	64	Pain	2 m	Radiotherapy	3 m	Deceased
Lim C et al[25]	2007	Case Report	RCC	1	M	49	Pain	7 m	Curettage	12 m	No
Singh et al[52]	2009	Case Series	NM	3	2 M 1 F	64	Pain+Swelling	NM	NM	NM	-NM
Burk et al[32]	2010	Case Report	Melanoma	1	M	13	Pain	4 y	Patellectomy+RT+CT	3.5 y	No
Tudor et al[23]	2010	Case Report	Lung. ASC	1	M	86	Pain	3 m	Patellectomy+RT	1 y	No
Codreanu et al[100]	2012	Case Report	Lung. AC	1	F	57	Pain+Swelling	NM	Patellectomy+RT+CT	NM	No
Tas & Keskin[34]	2013	Case Report	Melanoma	1	F	52	Pain	NM	Chemotherapy	15 m	Deceased
Broomfield et al[26]	2014	Case Report	RCC	1	M	74	Pain	24 m	RT+BP	24 m	NM
Feng et al[101]	2015	Case Report	Lung. SCC	1	M	65	Pain	NM	Patellectomy	NM	NM

Abbreviations—AC: Adenocarcinoma; ASC: Adeno-squamous Carcinoma; BP: Bisphosphonates; CT: Chemotherapy; NM: Not Mentioned; OS: Osteosarcoma; RCC: Renal Cell Carcinoma; RT: Radiotherapy; SCC: Squamous Cell Carcinoma

Discussion

Embryologically and anatomically, the patella is a sesamoid bone that optimizes the biomechanics of the patellar tendon (35). As such, it is considered an apophysis or an epiphyseal equivalent in terms of tumor location (35). The incidence and prevalence of primary bone tumors originating from the patella are both, extremely low with primary benign neoplasms being far more common than their malignant counterparts. As expected, giant cell tumor of bone and chondroblastoma are the most common benign neoplasms in this location given the apophyseal equivalent characteristics of this bone (3). A similar phenomenon is seen in the calcaneus (36). The epidemiology of malignant neoplasms of the patella follows that of primary bone sarcomas in general, with osteosarcoma and chondrosarcoma being the most common (1,3). Metastatic lesions are far less common, the location is random and matches the statistics of the most common metastatic cancers with osseous predilection such as lung cancer or breast cancer (8,24,29).

Atraumatic knee pain with or without knee effusion is one of the most common orthopaedic complaints along with other symptomatology around the knee. Unfortunately, the lack of specificity of these symptoms may delay the diagnosis of oncologic processes that by default are not high in the differential diagnosis when these symptoms present (37). The majority of patients receive an initial trial of treatment with anti-inflammatories, physical therapy, rest, elevation and icing that has minimal to no impact in terms of recovery. Delay in diagnosis also occurs as the clinical progression might be insidious and slow lessening the level of concern of the treating physician and /or the patient (38).

We consider radiographs at an early stage to be useful in fastening diagnosis. Radiographs are particularly helpful in patients with mechanical symptoms, pain at night and acute inability to bear weight. Axial imaging is relevant when conventional radiographs are non-diagnostic or to narrow the differential diagnosis obtained from initial radiographs. CT scans are valuable in assessing bone integrity while MRI imaging studies are more helpful in evaluating bone marrow edema and the soft-tissue component or extension of the tumor.

Our experience with benign tumors was similar to the published literature with GCT of bone and

chondroblastoma being the most common and prevalent entities. We could not identify, based on our review of the published literature, a site of predilection based on quadrant location in the patella. Results through the published literature and our experience, confirm satisfactory and predictable clinical results with excision and curettage and cement or bone graft packing(39) [Table 5]. Our preference is to use bone graft in younger patients and in the treatment of tumors particularly close to the articular surface. Conversely, cement is our preference for treatment in older patients. An interesting finding in our cohort was the age of presentation in patients with chondroblastoma. This tumor considered to be commonly found in the second decade of life, was found in older patients in our institution.

In terms of malignant tumors, the most frequent neoplasm in our review of the literature was osteosarcoma. As expected, it is more common in males than females and it has, as in other locations, a bimodal distribution in terms of age (40). In intermediate and high grades, it is traditionally treated with preoperative chemotherapy and surgery with negative margins followed by postoperative chemotherapy. The chemotherapy consists in high doses of Adriamycin, Cisplatin and Ifosfamide in combination with high-dose Methotrexate (41). Resection with negative margins implies a partial or total patellectomy if there is not intra-articular extension. If there is, resection of the knee joint is necessary to attain oncologic control. In the setting of metastatic disease, the burden of metastatic involvement defines the extent of treatment. In patients with isolated metastatic disease a resection with negative margins may improve oncologic outcome (17). In patients with extensive metastatic disease, a less invasive surgery focusing of palliation might be more appropriate. For example, in a patient with extensive lung metastatic disease and a patellar metastatic lesion, a palliative treatment such as curettage and cement packing followed by radiation might be sufficient for pain control.

In second place in prevalence, we identified in our review primary chondrosarcomas. The previously know "low grade chondrosarcoma" is considered in the new WHO tumor classification a "Low-grade cartilaginous tumor" (42). The removal of the word chondrosarcoma is an attempt to diminish the misinterpretation of the aggressiveness of this tumor. This neoplastic process carries an extremely low risk for both, metastatic disease and malignant transformation to a more aggressive tumor when it is located in the extremities (42). The treatment in this particular scenario is the same as for primary benign bone tumors, excision and curettage with subsequent packing with either cement or bone graft (42). An important consideration is that the risk of recurrence is higher after surgical treatment than enchondromas. Our literature review proved this tumor to be among the most common type of "chondrosarcoma" in the patella. The role of chemotherapy and radiation therapy is limited given the intrinsic biologic resistance of the tumor to these treatment modalities (5). Higher grade chondrosarcoma tumors, grade two and three, represented 50% of the primary malignant bone tumors identified in our review (5). Both grades are considered now high-grade primary bone sarcomas according to the WHO classification. They possess the same resistance to conventional chemotherapy and radiation therapy. The main stem for treatment is surgical resection with negative margins in which the same principles described for osteosarcoma resection apply. Controversially, this tumor was found in a female patient whereas it is more commonly expected in male patients (18).

Bone is a common site of metastasis, but the patella due to its low blood flow is a very uncommon site from other tumors to spread (24). However, it could be expected due to its ossification process which is similar to the long bones. There is a significant lack of literature reviewing metastatic patellar tumors (21). In our review of the literature and in our experience, we identified as prevalent histology those with osseous tropism and high prevalence for acrometastasis such as primary lung cancers and primary renal cell carcinomas. The management of these lesions is based on the primary histology of the tumor and the soft-tissue involvement. Range of treatment varies on the spectrum from no actual treatment to amputation or total knee reconstruction pending on the function of the patient, presence and severity of osteoarthrosis, burden of disease and survival expectancy on each individual patient.

Our findings and results are limited due to the rare nature of these tumors and the low level of evidence published in the literature which intrinsically increases selection bias. Nevertheless, our review is systematic and comprehensive summarizing a large body of literature of a rare condition. Not all papers were included as selection was based on the information reported in the screen and reviewed papers increasing furthermore selection bias.

Conclusion

In summary, primary patellar bone tumors, benign, malignant or metastatic, are rare but should be part of the differential in patients with chronic knee pain that does not respond to initial conservative interventions. This tumor-related pain tends to be chronic, intermittent, mostly accompanied by swelling and these are very non-specific symptoms. Radiographs must be the first step in diagnosis as valuable information in terms of what the tumor is doing to the bone and how the bone is reacting to the tumor can be attained. Axial imaging is necessary when radiographs are negative or demonstrate suspicious findings such as bone destruction, periosteal reaction and pathologic fractures. Keeping in mind oncologic processes in the differential diagnosis of knee pain, may reduce delay in treatment and misdiagnosis. This delay in treatment correlates with worse outcomes when the surgical decision has been made. Recurrence rate with intralesional curettage and bone grafting or cement packing is very low and therefore these are first treatment choice for benign neoplasms. Resection with negative margins in malignant neoplasms or bone metastasis decrease local recurrence but only in the former group there is an impact in survival. Multidisciplinary care is necessary in this group of patients.

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