

Atypical Presentation of Metastatic Carcinoma Causing Patellar Destruction and Synovial Carcinomatosis: A Case Report

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Learning Point of the Article:

In the context of a patient with high-stage carcinoma and the sub-acute onset of mono or oligoarticular inflammatory arthritis, physicians should consider synovial metastases on their differential.

Abstract

Introduction: This case report describes the third documented example of primary esophageal carcinoma metastasizing to the patella and the first documented example of esophageal carcinoma metastasizing to synovium.

Case Report: A 67-year-old man with a history of metastatic esophageal carcinoma presents with right knee pain and an aggressive, destructive lesion involving the superior patella. Biopsy revealed esophageal carcinoma. After ineffective radiation, he underwent resection of the tumor-filled bone and quadriceps advancement. Two months later, a recurrent tumor involving the entire patella and significant knee synovitis was observed. He underwent a total patellectomy with a radical anterior synovectomy. Further assessment showed that the entire synovium was replaced with metastatic carcinoma.

Conclusion: This report describes an atypical presentation of metastasis with patella and synovium involvement.

Keywords: Esophageal carcinoma, patellar metastases, synovial metastases.

Introduction

Primary patellar tumors are rare, and patellar metastases are even rarer [1, 2]. A recent literature review reported only 44 cases of patellar metastasis in the literature [2]: They found that patellar metastasis was most commonly of lung origin (~40%) and only rarely from gastrointestinal primary cancers –with esophageal carcinoma comprising <5% of patellar metastases.

Although patellar metastasis is rare, even more rare is metastasis to the knee joint synovium. There are only approximately 50 reported cases of metastasis to synovium [3] with primary lung cancer being the most common cause of synovial metastasis, followed by cancers of colorectal origin, and no reported cases of upper gastrointestinal tumors metastasizing to the synovium.

The purpose of this case report is to describe the third documented example of primary esophageal carcinoma metastasizing to the patella and the first documented example of esophageal carcinoma metastasizing to synovium.

Case Report

The patient, who provided consent for this report, is a 67-year-old male with a past medical history of hypertension, hypothyroidism, and esophageal carcinoma with known lung metastases who presented with 3 months of worsening right knee pain. In addition, he reported that the pain worsened with any range of motion activities, as well as direct pressure, and he was unable to find any relieving factors.

Author's Photo Gallery



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Figure 1: Anterior posterior and lateral radiographs of the right knee taken at the time of presentation demonstrated a radiolucent destructive bone lesion with a large expansile soft-tissue mass at the superior pole of the patella.

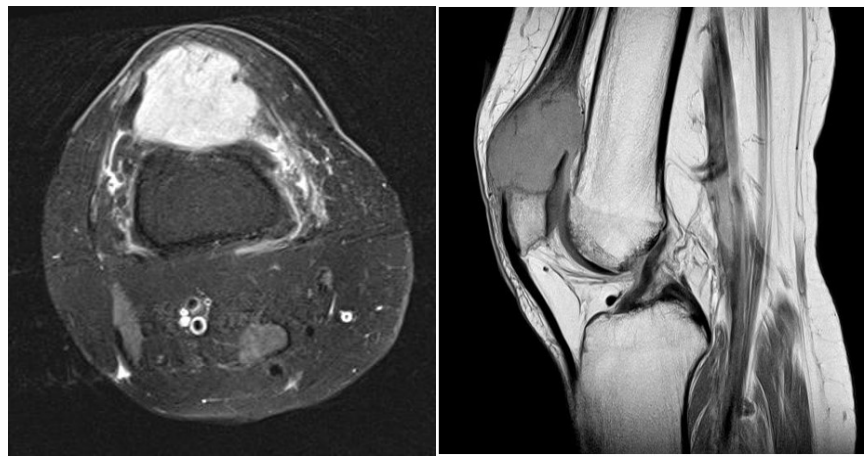


Figure 2: Magnetic resonance imaging done before presentation with Axial STIR (a) and Sagittal T1 (b) demonstrated a 4.2 cm expansile destructive lesion originating at the superior pole of the patella, with soft-tissue involvement of the quadriceps tendon.

On examination, the patient was generally deconditioned and able to stand with significant pain. He had decreased active (10–70°) and passive range of motion (0–90°) and was otherwise neurovascularly intact without lymphadenopathy. Magnetic resonance imaging (MRI) before presentation demonstrated a 4.2 cm destructive lesion at the superior pole of the patella, invading inferiorly into the patella, along with soft-tissue involvement of the quadriceps tendon (Fig. 1). Radiographs at presentation demonstrated a radiolucent bone lesion at the superior pole of the patella (Fig. 2). Due to the rarity of patellar metastases, the patient was scheduled for a biopsy with a frozen section with a plan for exploration, curettage with bone grafting and quad augmentation if the lesion was found to be metastatic carcinoma.

Due to medical issues, 6 weeks later the patient underwent the procedure described above. A 2-cm incision was made over the patella using fluoroscopic guidance. On dissecting through the retinaculum, abnormal tissue was noted at the superior patella. This tissue was biopsied and histologic analyses of the frozen section confirmed metastatic carcinoma. The rest of the patella was then exposed by dissecting through the retinaculum and intralesional curettage was performed. Approximately 40% of the patella was left intact. Deep into the synovial tissue, there was intact cartilage connected to the distal pole of the patella, which was left intact. Hydrogen peroxide was used to clean the wound and the joint was visualized with no significant abnormalities noted. The quadriceps tendon was then debrided and advanced to the superior pole of the patella using No. 5 Fiberwire (Arthrex, Naples, FL) in a Krackow fashion and secured distally through transosseous drill holes. The patient's wound was closed, and he was transferred to the PACU in stable condition.

Two weeks later, the patient returned to the office for a routine follow-up. He was in considerable pain but otherwise was doing well. The wound was clean, dry, and intact. Official pathology results confirming metastatic squamous cell carcinoma to the bone were reviewed with the patient.

Within the next month, the patient was admitted to an outside hospital for COVID-19 pneumonia. During the admission, there was a concern for septic arthritis as there was a poke-hole sinus tract on the anterior knee. However, arthrocentesis produced a negative cell count and culture. He also had a computed tomography scan which was read as concerning for osteomyelitis versus worsening malignancy. On transfer to our institution, radiographs (Fig. 3a and b) demonstrated that the patient had continued bony destruction. Repeat MRI (Fig. 4a and b) demonstrated homogenous enhancement of the distal pole of the patella with a differential diagnosis of continued metastatic disease, osteonecrosis, or osteomyelitis of the patella. The knee joint showed a large diffuse hypertrophic synovitis. Due to persistent pain, the patient was offered knee exploration and palliative completion patellectomy.

The patient had his sinus tract excised. Dissection revealed a small proximal remainder of the patella attached to the quadriceps tendon in the area of the previous advancement with significant distal bone loss. All remaining patella was removed as it had been completely replaced with a tumor. On removing the bone, the synovium was noted to have an abnormal red, beefy, nodular appearance. An extensive synovectomy was performed, including extensive superior proliferation in the suprapatellar pouch as well as the medial and lateral gutters and intercondylar notch. No purulence was encountered. Hydrogen peroxide was used to gain control of the tumor. A V-Y



Figure 3: (a and b) Radiographs demonstrated that the patient had continued bony destruction.

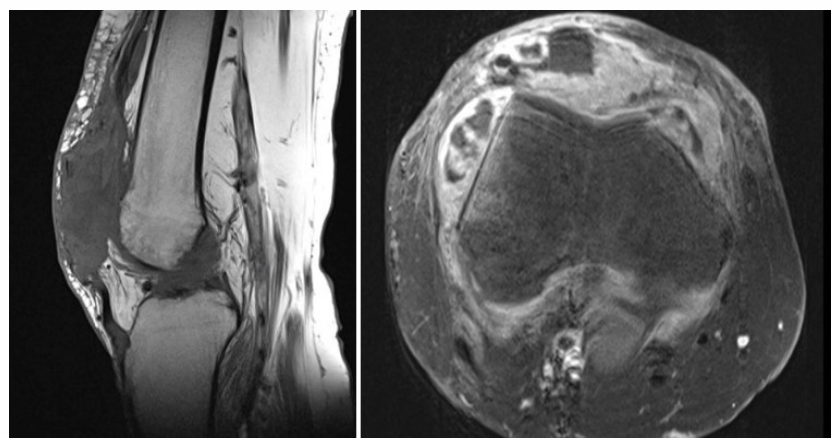


Figure 4: Repeat Magnetic resonance imaging Sag T1 (a), and contrast-enhanced Fat suppressed Ax T1 (b) demonstrated a homogenous enhancement of the distal pole of the patella and complex effusion with thickened, enhancing synovium through the joint space. Differential diagnosis included continued metastatic disease, osteonecrosis, or osteomyelitis in the patella with joint inflammation or infection.

quadricepsplasty was performed to advance the quad tendon to the patellar tendon and close the joint. The patella and synovium were sent for pathology. The patient was kept in extension and allowed to weight bear. His pain was better controlled although his ambulation was limited. He expired 3 months post-operation due to pulmonary complications.

Final pathology showed irregular, tan-brown fragments of lobulated fibrofatty tissue, dense fibrous tissue, hemorrhagic tissue, and a single piece of $3.9 \times 3.2 \times 2.0$ cm of cortical bone, all replaced with tumor (Fig. 5). No normal synovial tissue was noted, with all abnormal periarticular soft-tissue consistent with disseminated metastatic squamous cell carcinoma.

Discussion

To the best of our knowledge, this case report is the fourth well-documented case of patellar metastasis and the first well-documented case of synovial carcinomatosis. Metastasis with esophageal carcinoma is common with up to 50% of diagnosed patients having metastasis to other organs [4]. Lung metastasis as seen in this case is fairly common, occurring in 6.5% of patients diagnosed with esophageal carcinoma [5]. Although bone is the second-most common place for metastasis of esophageal carcinoma, rarely does this occur in the patella or knee synovium [4].

Patellar tumors in general are rare entities and are rarely metastatic in origin. Prior reviews from 1900 to 2000 reported 384 cases, and only 12% of these tumors were metastatic in origin [6]. An epidemiological study showed that patellar metastasis is most commonly of lung (~35%), breast (~10%), or kidney (~10%) origin [6]. Esophageal carcinoma reportedly occurs at an incidence of 7%, with only three other cases found

in the literature (Table 1) [7-9]. Consistent with prior reports, our patient presented with disabling knee pain, a radiolucent lesion in the patella, and MRI-revealed non-specific hypertrophic synovitis. Despite aggressive initial surgical treatment, the patient developed tumor progression to the inferior pole of the patella, requiring intervention for persistent pain.

During subsequent patellectomy with V-Y advancement, the synovial tissue was noted as grossly abnormal and friable, and a full synovectomy was performed. Pathologic analysis confirmed the presence of synovial metastasis with a special note of no remaining normal synovial tissue. Metastasis of solid tumors to synovium is similarly a rare event with approximately 50 cases described in the literature (Table 2) [3, 10]. These are most commonly of lung or colorectal origin [3]. To the best of our knowledge, this is the first case report on esophageal carcinoma metastasizing to synovium. Prior studies have suggested two mechanisms by which tumors metastasize to synovial tissue including direct spread from periarticular masses (likely what happened in our case) versus hematogenous spread [11, 12], while others have suggested prior surgical intervention played a role in spread to synovium [6]. Although the etiology and timeline of synovial metastasis, in this case, is unknown, the pathologist was unable to identify any normal synovial tissue in the specimen, suggesting the synovial metastasis was either present for a long period of time or was rapidly aggressive, representing a case of synovial carcinomatosis. The initial and follow-up MRI scans showed enhancement throughout the synovium, which could be consistent with both synovitis versus disseminated synovial metastases (Figs. 1, 4a and b). Physicians treating such patients

Conclusion

Patellar and synovial metastases are rare events, and their co-incidence is even rarer. As a result, the approach to diagnosing and treating either metastatic event remains understudied. This highlights the importance of publishing detailed reports on the treatment of patients with these conditions. Here, we have presented such a case, possibly the third to be published, and the first to be diagnosed definitively as concurrent patellar and synovial metastases, with the synovium completely replaced with carcinoma. Both metastases appear to present similarly, albeit non-specifically, with the sub-acute onset of mono or oligoarticular inflammatory arthritis. However, in the context of a previous diagnosis of high-stage carcinoma, physicians should consider either patellar or synovial metastases in their differential.

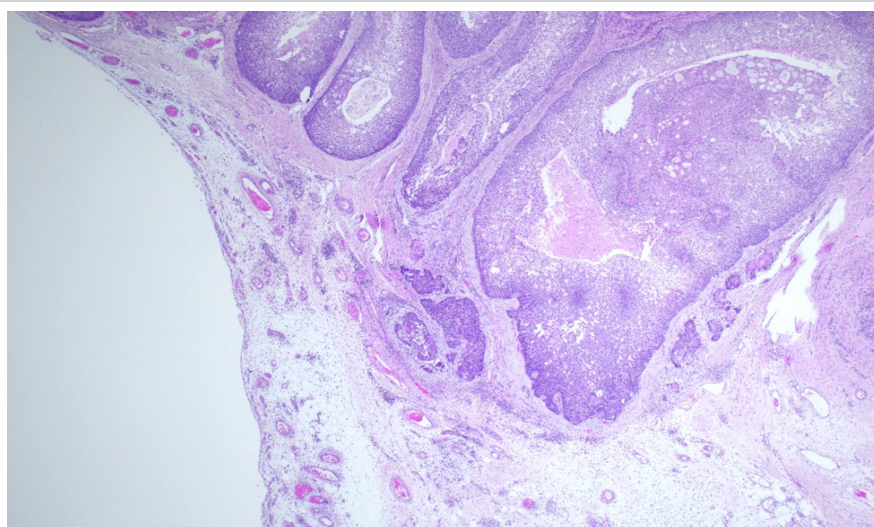


Figure 5: A low magnification photomicrograph showing residual synovium in the left lower half of the image and large, infiltrating nests of metastatic squamous cells carcinoma with focal necrosis.

should consider co-incidence of synovial metastasis and be sensitive to synovial biopsy or prophylactic synovectomy in patients undergoing surgical intervention. Furthermore, as disseminated carcinoma in other body cavities has been well described (for example, pleural or peritoneal carcinomatosis), we suggest a new diagnosis of “synovial carcinomatosis.”

previous diagnosis of high-stage carcinoma, physicians should consider either patellar or synovial metastases in their differential.

Cases	Age	Gender	Presentation	Diagnosis	Synopsis/studies	Treatment/findings
Stoler and Staple [7] (Note. Though this case is reported as a patellar metastasis from an esophageal primary, their arthrogram findings suggest this may be a case of both patellar and synovial metastases from an esophageal primary.)	52	M	Progressive dysphagia, weight loss, monoarticular inflammatory arthritis.	Esophageal carcinoma; patellar metastasis (and metastases to other joints).	X-ray demonstrated a lytic lesion of the patella and a mass in the suprapatellar bursa.	Patient was treated “conservatively” and discharged.
Arthrogram showed “irregular synovium compatible with metastatic nodules.”						
Ashby and Dappen [8] (Note. Though this case is reported as a patellar metastasis from an esophageal primary, their findings on gross inspection of the joint suggest this may be a case of both patellar and synovial metastases from an esophageal primary.)	55	M	Progressive dysphagia, weight loss, monoarticular inflammatory arthritis.	Esophageal carcinoma; patellar metastasis.	Arteriography showed “marked vascularity in the areas of synovium containing the tumor.”	After treating the patient with antibiotics and failing to see any improvement, an arthrotomy was performed, and a tumor within the patella and “extending into the suprapatellar bursa” was found. In addition, the “synovium was studded with metastatic nodules.” A total patellectomy was performed, and radiation was applied to the knee.
X-ray demonstrated “lytic destruction of the patella.”						
Zhao et al [9]	50	M	Acute pain and immobilization of the right knee after a fall 2 months after receiving a diagnosis of esophageal carcinoma.	Esophageal carcinoma; patellar metastasis.	A 99 m Tc-methylene diphosphonate bone scintigraphy scan showed “increased radioactivity in the right patella.”	Patient denied therapeutic intervention for his patellar lesion and died one month later due to a pulmonary embolism.
					Biopsy of the patella confirmed patellar metastasis from esophageal carcinoma.	
Muran et al. (Note. Case of esophageal carcinoma patellar metastases and synovial metastases.)	67	M	Progressively worsening monoarticular knee pain	Esophageal carcinoma; patellar and lung metastases	X Ray at presentation demonstrated a radiolucent bone lesion at superior pole of patella with soft tissue invasion of the quadriceps tendon.	Biopsy, exploration, and curettage: approx. 40% of the patella was left intact.
					Arthrotomy showed abnormal tissue noted at the superior pole of the patella.	Eventually the patient returned with knee pain concerning for osteomyelitis or worsening malignancy: Xray showed a transverse patella fracture and the patient underwent a total patellectomy and synovectomy (because the synovium appeared nodular, inflamed, and abnormal on gross inspection consistent with SC).
					Biopsy confirmed metastatic carcinoma.	
					Pathology findings of patellectomy and synovectomy demonstrated irregular, tan-brown fragments of lobulated fibrofatty tissue, dense fibrous tissue, hemorrhagic tissue, and a single piece of cortical bone. No normal synovial tissue was noted and all of this was consistent with metastatic squamous cell carcinoma.	

Table 1: Cases of esophageal carcinoma patellar metastases and/or synovial metastases.



Cases	Age	Gender	Presentation	Diagnosis	Synopsis/studies	Treatment/findings
Khurram et al. [3]	67	M	Hematuria and monoarticular knee pain and swelling.	Transitional cell carcinoma of the bladder; synovial and lung metastases.	X-ray produced no additional findings.	The patient was treated with palliative external beam radiotherapy to the left knee and received a total of two single doses of 8 Gy radiation therapy. He was also initiated on pembrolizumab. For the treatment of bone pain, he was prescribed zoledronic acid.
					Computed tomography scan "depicted a permeative, granular appearance of the bones at the left knee with erosive changes.	The patient passed away in January 2018 due to the progression of his disease, approximately 6 months after being diagnosed with synovial metastasis.
					Ultrasound identified synovial thickening	
					Aspiration and culture demonstrated no bacterial growth, but cytologic findings of: "three-dimensional clusters of atypical epithelial cells, scattered within a hemorrhagic background along with synovial cells"	
					Immunohistochemistry confirmed this as metastatic transitional cell carcinoma of the bladder.	
Tapia Rico et al. [13]	31	F	Diarrhea and rectal bleeding; progressively worsening monoarticular knee pain and swelling.	Colorectal carcinoma; lung, liver, and synovial metastases.	Knee aspirate showed no organism or crystals and rheumatologic serologies were negative	Open right synovectomy showed exuberant and friable synovial tissue on gross inspection
					X-ray – no additional findings	Histopathology confirmed metastatic adenocarcinoma from the rectal primary lesion
					MRI: showed a large joint effusion with diffuse thickening and nodularity of the synovium without bone abnormalities	
					Repeat MRI: showed increased synovial thickening	
Levine et al. [14]	61	M	Monoarticular knee pain and swelling 4 months after left pneumonectomy for lung cancer.	Large-cell lung carcinoma; synovial metastasis.	Aspiration showed inflammatory white blood cells but no microorganisms.	
					Gadolinium-enhanced MRI demonstrated: "diffuse enlargement of the knee joint space secondary to a multiloculated and heterogenous mass; most of the joint space was replaced by hyper-enhancing synovium."	
					Surgical pathology confirmed the diagnosis of synovial metastasis from primary large-cell lung carcinoma.	
McConnell et al. [15]	83	M	Hematuria and progressively worsening monoarticular knee pain and swelling.	Renal cell carcinoma; synovial metastasis.	X-ray: large intra-articular soft-tissue mass with small calcifications.	
					MRI: multilobulated intra-articular tumor with bone erosions at distal femur.	
					Pathology of ultrasound guided biopsy of synovial mass demonstrated poorly differentiated metastatic carcinoma.	

MRI: Magnetic resonance imaging

Table 2: Other representative cases of knee joint synovial metastases within the past 10 years.

Clinical Message

Synovial metastases are rare occurrences, but in the context of a patient with previously diagnosed high-stage carcinoma, and sub-acute to chronic inflammatory arthritis, they should be considered. Our case is one of a few well-documented descriptions of the diagnostic workup and management of a patient with synovial and patellar metastases from esophageal carcinoma - offering insight into how these patients might present, the diagnostic tests that can be used, pathologic descriptions of these lesions, and discussion of their surgical management. Though definitive diagnosis of synovial metastases requires a biopsy of the tissue, soft-tissue involvement can be visualized with MRI and help to narrow the differential to a process involving periarticular soft tissue. Lastly, due to the rarity of synovial and patellar metastases in the literature, and the significant diagnostic and treatment challenges they pose, it is important for physicians who encounter patients with these conditions to publish detailed case reports on how they were diagnosed and treated.



Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

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