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CASE REPORT

Referred Pain: A Case Report on Shoulder Pain Caused by a Mediastinal Tumor

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Abstract

Shoulder pain has many etiologies, with the most common causes being the shoulder joint itself and the surrounding soft tissues, such as tendons, bursae, or ligaments. However, rare pathologies can occur. This report presents a case of chronic right shoulder pain eventually diagnosed as a rare case of extra-skeletal osteosarcoma. The referred right shoulder pain was likely caused by tumor invasion of the pericardium, whose sensory innervation is provided by the phrenic nerve. The purpose of this case report is to highlight the fact that, although rare, neoplasm can be a cause of shoulder pain. Rather than just relieving symptoms, an accurate diagnosis is required to effectively treat the problem.

Keywords: Referred pain, Shoulder pain, Tumor

1. Introduction

The most common causes of shoulder pain arise from the shoulder itself including rotator cuff injury, adhesive capsulitis, shoulder instability, and arthritis.[1] Arm or shoulder activity usually cause the pain to worsen. However, various diseases and conditions affecting structures other than shoulder may result in pain in the shoulder area. This referred pain usually does not worsen with movement of the shoulder.

It has been noted that mediastinal tumor induced diaphragmatic irritation or phrenic nerve pathology can be a cause of referred shoulder pain. However, this is still not as common as pain originating from the joint or surrounding muscles, tendons, and ligaments. This report describes a patient with an extra-skeletal osteosarcoma, a rare malignancy accounting for 2%−4% of all osteogenic sarcomas,[2] who presented with right shoulder pain as an initial symptom.

2. Case report

A 48-year-old man, who worked as a university teacher, presented to the Department of Interventional Radiology on February 25, 2021 complaining of recurrent right shoulder pain for the past several weeks. He denied any systemic diseases such as hypertension or diabetes as well as any recent trauma. The pain was dull and more prominent at night with an intensity of 5/10 on the visual analog scale (VAS). There was no radiating pain to a distal part of the body. Physical examination revealed some tenderness over the posterior aspect of the right shoulder, and there was no limitation to range of motion (ROM). A radiograph of the right shoulder (Fig. 1) found no contour defects or discontinuities. Early stage of clinical frozen shoulder was impressed and a local intra-articular (IA) injection with Shincort 40 mg (1 ml) + Lidocaine 2% 5 ml was performed and an oral pain killer was prescribed. Further, a right shoulder MRI (Fig. 2a and b) performed on March 10, 2021 revealed suspected supraspinatus tendinitis.

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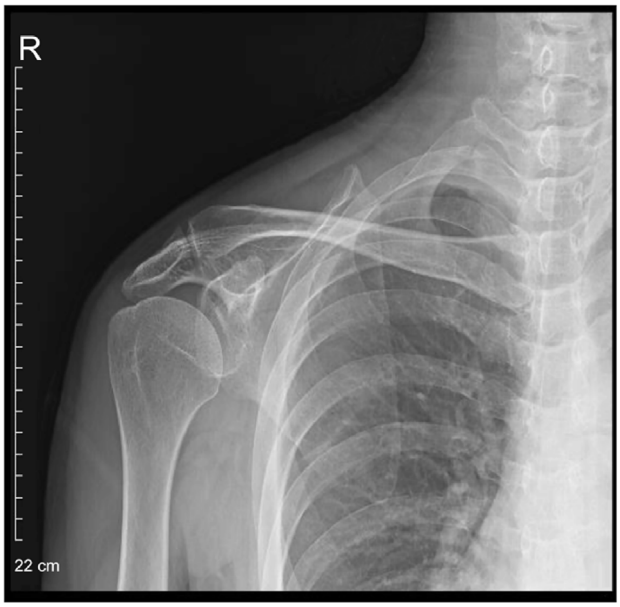

Fig. 1. Right shoulder x-ray revealing no contour defects nor discontinuities.

The patient responded poorly to the injection treatment and experienced persistent right shoulder pain with numbness over his right arm. He sought out acupuncture therapy, but the symptoms became worse after treatment. He visited the Department of Cardiovascular Medicine on April 1, 2021 over concerns of progressive right shoulder pain which might be caused by associated heart disease. A chest radiograph (Fig. 3) showed cardiomegaly with an elevated right hemidiaphragm, and an ECG showed normal sinus rhythm. An oral analgesic and muscle relaxant were prescribed. The patient was then advised to visit the Department of Rehabilitation for further pain control and rehabilitation training. However, he had never showed up at our clinic for interventions.

A chronic cough with white sputum and progressive dyspnea were reported in the following weeks. He denied any fevers, and there was no significant weight loss reported. Due to the new onset of worsening respiratory symptoms, he was seen in the Department of Chest Medicine on May 3, 2021. A chest radiograph disclosed a right lower lobe mass with ipsilateral pleural

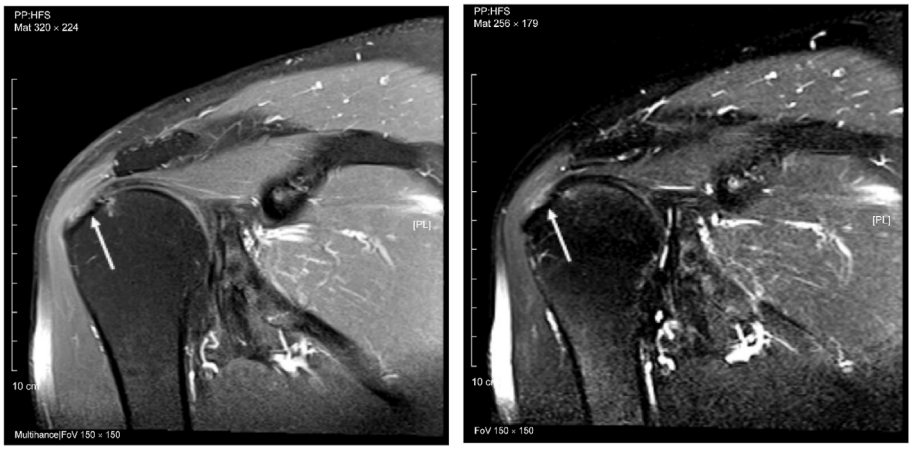

Fig. 2. a. T1. fs. cor. +C. b. T2. fs. cor. Focal increased signal intensity over insertion site of supraspinatus tendon & enhancement post IV contrast, the tendinitis is considered.

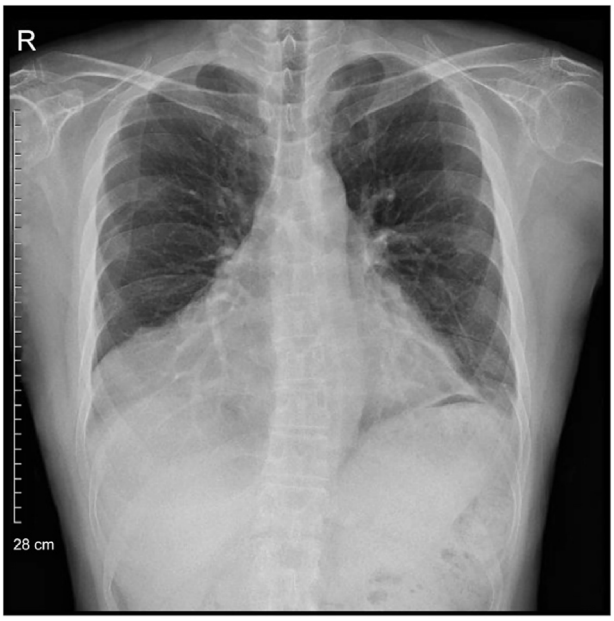

Fig. 3. Chest x-ray indicating cardiomegaly with elevated right hemidiaphragm and blunting of bilateral costophrenic angles.

effusion. Admission for a cancer survey was suggested, but the patient refused and returned home to discuss treatment plans with his family. Increasing dyspnea was noted that night, and he was brought to a nearby medical center for further evaluation and management.

A chest CT scan (Fig. 4) revealed a large, heterogenous, mediastinal tumor approximately 10 cm in size with invasion of the pericardium. Tumor biopsy noted a poorly differentiated tumor favoring a sarcoma. A bone scan and brain MRI were performed to rule out distant metastases, and the results of both studies were negative.

The patient was referred to the Department of Thoracic Surgery on May 21, 2021 for tumor resection and further treatment. A chest MRI (Fig. 5) revealed an infiltrative, heterogenous, contrast-enhancing mass with a cystic component approximately 14.6 × 10.0 cm in size in the right mediastinum with invasion of the superior vena cava, right atrium, right middle lobe, and anterior mediastinum. The treatment, which proceeded smoothly, included resection of the anterior mediastinal tumor and partial pericardiectomy via medial sternotomy on May 26, 2021. Operative findings revealed a 15 cm × 12 cm × 10 cm yellowish tumor over the anterior mediastinum with invasion of the pericardium. The pathologic report noted an extra-skeletal osteosarcoma (ESOS) of the anterior mediastinum, staging of pT2Nx, and the pleural biopsy was negative for malignancy (Fig. 6).

The patient's clinical condition stabilized, and he was discharged 5 days after surgery. Follow-up at the Department of Thoracic Surgery occurred one week after discharge, and adjuvant chemotherapy was suggested. He was also seen at the Department of Rehabilitation at his one-week follow-up, and improved right shoulder pain, 1−2/10 on VAS, was reported with no significant findings on physical examination. The patient was given education on chest mobility, breathing exercises, and aerobic exercise.

3. Discussion

Extra-skeletal osteosarcoma (ESOS), a rare variant of osteosarcoma with a poor prognosis, is an aggressive tumor consisting of approximately 1% of all soft tissue sarcomas. It is not considered as extensions of the primary bone osteosarcoma. The 5-year overall survival rate for all patients ranges from 37% to %1%. Poor prognostic factors include a later stage when diagnosed, a larger tumor size, and older age. Patients who develop ESOS are usually between 48 and 60 years of age with males being slightly more affected. This is consistent with this patient's

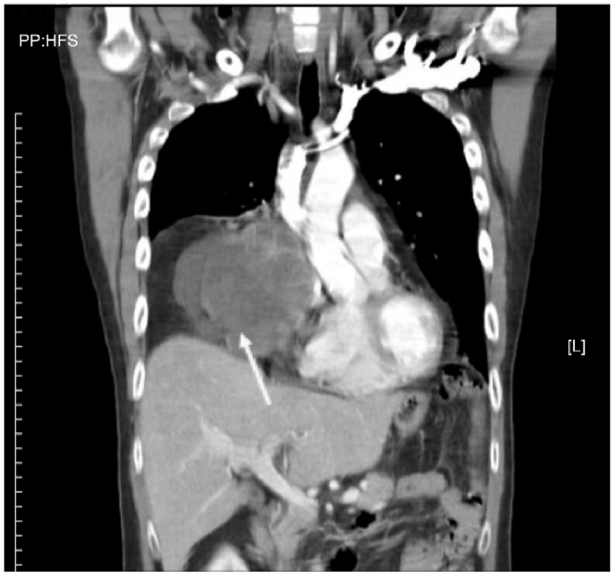

Fig. 4. Chest CT scan showing a heterogenous mediastinal tumor about 10 cm in size, with invasion into the pericardium.

characteristics. The malignancy most often affects the lower limbs followed by the upper limbs and retroperitoneum. However, this patient had a mediastinal ESOS, which was an even rarer case. To be noticed, despite MRI revealed right mediastinal tumor, the surgical result showed anterior mediastinal tumor instead, and should be the final diagnosis. ESOS commonly presents with pain and swelling of the affected region. On occasion there may be constitutional symptoms such as fatigue, anorexia, and weight loss. Risk factors include prior trauma, radiation therapy and underlying genetic conditions such as Paget's disease. Image modalities, for instances, x-ray, CT and MRI can help identifying the disease with variable degrees of calcification may be noticed. Biopsy is the gold standard for diagnosis and microscopically, ESOS displays variable amounts of

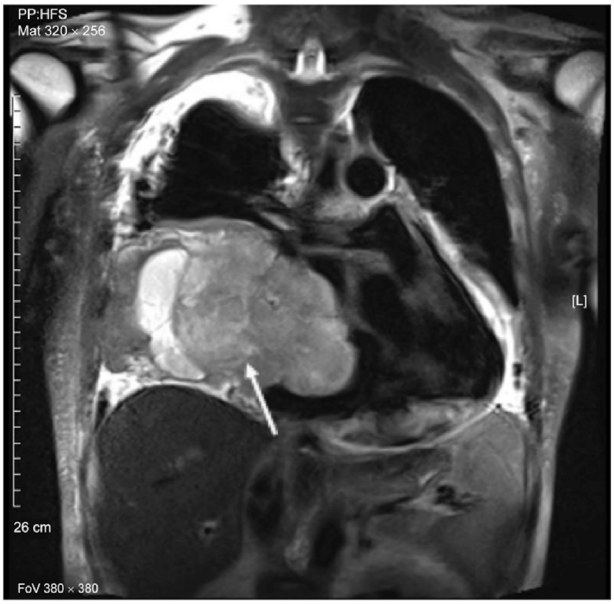

Fig. 5. Chest MRI indicating an infiltrative, heterogenous contrast enhancing mass with cystic component about 14.6 × 10.0 cm in size at right mediastinum with invasion to the superior vena cava, the right atrium, the right middle lobe and the anterior mediastinum.

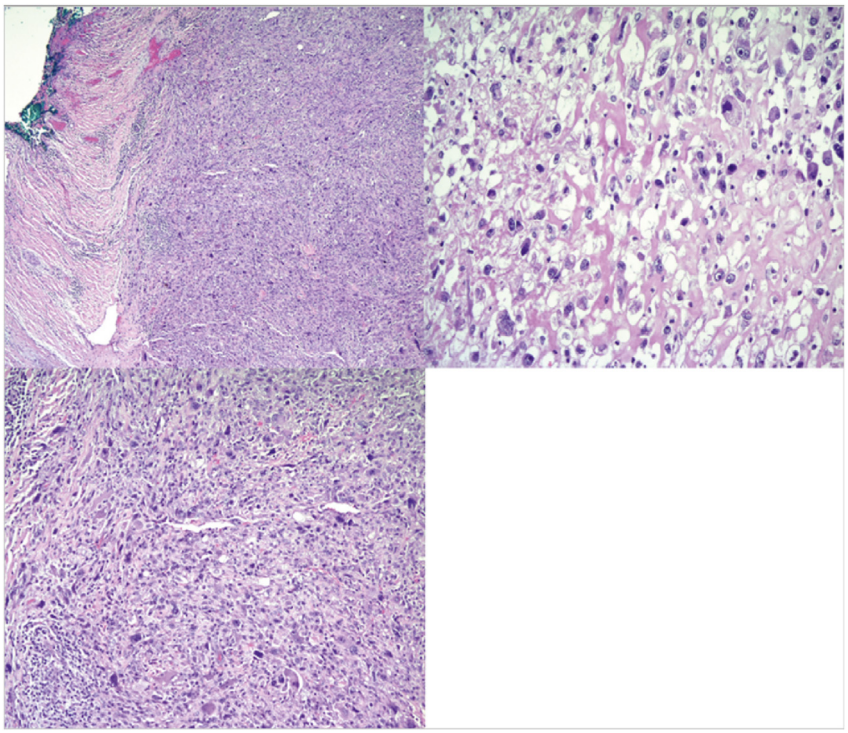

Fig. 6. Histopathology showed tumor cells demonstrating ovoid, pleomorphic spindle nuclei with prominent nucleoli and atypical mitosis, arranged in storiform and sheet pattern. Areas of small round cells and malignant cartilage are noted. Large areas of necrosis (>50%), tumor giant cells, osteoid with lacelike and trabecular pattern are present (hematoxylin and eosin stain).

osteoid. Treatments consists of wide surgical excision and adjuvant chemotherapy or radiation therapy. However, the role of chemotherapy, especially higher grade of ESOS, is still controversial.[2–4]

This patient, however, presented with an initial symptom of right shoulder pain with subsequent numbness developing over the ipsilateral arm. This is so called referred pain, which, by definition, is pain perceived at a location other than the site of the stimulus.[5,6,8] Referred pain follows dermatomal rules, meaning the structure where the pain originates and the structure to which the pain is referred developed from the same embryonic segment. Therefore, they are innervated by a common neural segment.[6–9]

This patient was initially diagnosed as frozen shoulder, also known as adhesive capsulitis, and received subsequent IA steroid injection at the Department of Interventional Radiology. Adhesive capsulitis is defined as painful, restricted ROM generally in more than 3 directions, with normal radiographic findings in patients. It is more commonly seen in women between 40 and 60 yeas of age. It is often idiopathic, but can be associated with autoimmune disease and endocrine etiologies such as diabetes mellitus. The pathophysiology is predominantly related to the inflammatory-fibrotic cascade. Adhesive capsulitis can be divided into 4 phases based on the degree of clinical pain and limitation of ROM. It is usually a clinical diagnosis but image studies, including ultrasonography, should also be arranged to rule out other possible causes such as rotator cuff injuries. Though early stage of frozen could be considered in this patient as he presented with shoulder pain and no obvious ROM limitations, it is more common for us to see patients present in later stage with more restricted shoulder ROM. In addition, more detailed physical examination should be documented. Was there any radiation pain, to what degree the patient's shoulder ROM limitation was, what was the end-feel of his symptomatic shoulder, and what were the results of the associated provocation tests, such as Empty can or Yocum test. All of the above examinations should be reported in

order to differentiate from other etiologies. Treatment of adhesive capsulitis consists of analgesics, physical modalities, ROM and scapular stability exercises, echo-guided injection or capsular hydrodilatation. More aggressive interventions such as manipulation under anesthesia or arthroscopic lysis of adhesions can be performed if non-operative treatment fails.[10]

This patient's right shoulder pain was likely caused by tumor invasion of the pericardium, whose sensory innervation is provided by the phrenic nerve. The phrenic nerve originates from C3 through C5 spinal nerve roots which also supply cutaneous innervation for the neck, shoulder, and upper arm. In addition to the pericardium, the phrenic nerve also supplies sensory fibers to the diaphragm, mediastinal pleura, and some abdominal structures such as the superior peritoneum.[1] This explains why pain is perceived in the shoulder despite the stimuli originating in visceral organs.

There are several distinct patterns that can help distinguish referred pain from local pain. First, referred pain is deep, a dull ache, and difficult to localize. It is sometimes described as an expanding pressure by the patient. In addition, it also has the features of spatial and temporal summation. With a stronger and longer duration of the noxious stimuli, the referred pain develops greater intensity and a wider area experiencing the pain. Also, secondary hyperalgesia and trophic changes of the skin and muscles can be seen over the area of referred pain.[9] However, it sometimes can be quite difficult to distinguish tumor-related visceral referred pain from visceral organ pain. Some clinical signs may help in differentiating the causes. In addition to the non-specific constitutional symptom, cancer pain can also contribute to neuropathic and somatic pain, which is burning, numbing and pinprick-like; and sharp, throbbing and well-localized in characteristic, respectively.[11] This may also explain why our patients experienced numbness over his right arm, which was not commonly seen as a symptom of referred pain. The neuropathic pain may be due to direct tumor invasion or compression of the related peripheral nerves that innervate the arm. Though, more common etiologies, for instance, cervical radiculopathy or facet joint arthropathy should be think of first.

Last but not least, it is worth noted that the chest x-ray is a simple, easily-approached and inexpensive tool to help identify possible visceral growth of tumor, which should be take into consideration whenever an atypical or intractable shoulder pain presents.

4. Conclusion

This report describes a patient whose chronic shoulder pain was the first symptom of a mediastinal tumor, in this case an extra-skeletal osteosarcoma. The pain was possibly due to involvement of the pericardium, which can sometimes be difficult to differentiate from pain originating in the shoulder itself. However, as physiatrists, it is important to identify the underlying cause of pain through a careful history, thorough physical examination and adjunctive image study. Hopefully this report will help to reinforce the need to consider a large differential diagnosis, including neoplasm, of the etiology of shoulder pain. An accurate diagnosis is necessary to be able to provide appropriate therapy as opposed to just relieving symptoms.

Conflict of interest

There is no conflict of interest.

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