Case Report

Complex Regional Pain Syndrome-like Pattern in a Case of Pancoast Tumor

Abstract

We present the case of a 52-year-old male who was recently diagnosed case of a Pancoast tumor and presented to the pulmonary outpatient department with a complaint of pain in the shoulder and chest region which was burning type, associated with shoulder abduction weakness and poor hand grip. Subsequently, he was referred for a ^{99m}-Tc bone scan for metastatic workup, which showed increased uptake in all the joints and long bones of the ipsilateral upper limb. This case highlights the importance of considering nontraumatic cause of pattern similar to complex regional pain syndrome.

Keywords: *^{99m}-Tc bone scan, complex regional pain syndrome, Pancoast tumor*

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Introduction

Pancoast tumors are also known as Pancoast-Tobias tumors, "superior sulcus tumors,"[1] "superior pulmonary sulcus tumors," or "non-small cell lung cancer."[2] Their location is adjacent to the apical pleuropulmonary groove, near the thoracic inlet, and adjacent to the subclavian vessels.[3-5] The major presenting features are shoulder and arm pain (in the distribution of the C8, T1, and T2 dermatomes), Horner's syndrome (ipsilateral ptosis, miosis, and anhidrosis; caused by injury to the sympathetic nerve chain), and weakness and atrophy of the hand muscles. Extension of tumor to the C8 and T1 nerve roots leads to upper limb neurological symptoms in about 8%–22% of cases.[5,6] These constellations of symptoms are known as Pancoast syndrome or Pancoast-Tobias syndrome.[5] Pulmonary symptoms such as cough, hemoptysis, and dyspnea are uncommon until late in the disease due to the peripheral location of the tumor. This results in a delay in diagnosis of 5–10 months in some cases and patients frequently receive treatment for presumed cervical osteoarthritis or shoulder bursitis.[7-10] Ipsilateral complex regional pain syndrome (CRPS) (reflex sympathetic dystrophy [RSD]) may occur rarely.[11,12]

CRPS is a multifactorial disorder. There are features of neurogenic inflammation that cause hypersensitivity to pain or severe allodynia. Other associations include blood flow problems, swelling, skin discoloration, and maladaptive neuroplasticity due to vasomotor disorders.[13-15]

There are two subtypes of CPRS according to the international association for the study of pain (IASP).[16]

Type I is also known as RSD, Sudeck's atrophy, reflex neurovascular dystrophy, or algoneurodystrophy. No neurological lesion is proven to be associated with this type. Most cases of CRPS fall into this category.

Type II, also known as causalgia, is associated with neural injury. It is more painful.[17] In Type II, the cause of nerve damage is known or obvious, although the causal mechanism remains unknown, as is in Type I. Several studies have investigated the etiology of this condition, but the cause remains unknown. The role of associated factors such as the limb immobilization technique and genetics has been reported in the development of this complication. A three-phase bone scan is complementary to the clinical history in diagnosing CRPS. It is characterized by increased accumulation of the radiotracer in the juxta-articular region of the affected joint on all three phases, with the delayed images being the most sensitive for confirming the diagnosis.

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Case Report

A 52-year-old man presented to the pulmonary medicine outpatient department with dyspnoea on exertion, weight loss, fever, and loss of appetite. This was associated with left shoulder pain associated with left-sided chest pain which was a burning type and radiating to the left upper limb. The patient also had shoulder abduction weakness and worsening left-hand grip. A contrast-enhanced computed tomography (CT) chest was done, which revealed a Pancoast tumor of the left lung. CT-guided biopsy from the lesion showed adenocarcinoma of the lung. The patient was referred for a bone scan to rule out any bone metastasis. The bone scan showed increased ^{99m}-Tc uptake in the left shoulder, left elbow, small joints of the left hand, and long bones of the left upper limb (black arrows in Figure 1a and b which show anterior and posterior images, respectively). This was CRPS-like pattern, however, the patient had no pain hypersensitivity/allodynia or sudomotor symptoms. The single-photon emission CT-CT of the upper thorax was acquired to look for metastatic involvement of ipsilateral upper limb bones which showed the involvement of D1-D3 vertebral bodies by the primary tumor and no other upper limb bony metastasis. The increased tracer uptake in the bones did not reveal any corresponding CT changes.

Discussion

Sympathetic dystrophy, aka CRPS presents with clinical features of hypersensitivity to pain or severe allodynia, blood flow problems, swelling, skin discoloration, and maladaptive neuroplasticity due to vasomotor disorders.[13-15] CRPS is commonly observed in patients with upper limb trauma. Several studies have investigated the etiology of this condition, but the cause remains unknown. The role of associated factors such as the limb immobilization technique and genetics has been reported in the development of this complication. A Pancoast tumor rarely develops the features of CRPS and respiratory symptoms are known to be uncommon until the late stages of Pancoast's tumor.[18] Three-phase bone scan in CRPS is characterized by increased accumulation of the radiotracer in the juxta-articular region of the affected joint on all three phases. The delayed images are the most sensitive for confirming the diagnosis. It is complementary to the clinical history in diagnosing CRPS. This case presenting with the features of burning type shoulder and chest pain may be attributed to the involvement of nerves by the apical and paravertebral extension of the tumor. The involvement of adjacent sympathetic nerves may be taken as the reason for developing the CRPS-like pattern. Although the increased ^{99m}Tc–methylene diphosphate uptake is seen in the areas of increased osteoblastic activity, here, it can be attributed to the increased blood flow to the left upper limb because of the sympathetic disturbance. Joint changes are usually not appreciated on the plain X-ray images in CRPS. This case highlights the importance of nontraumatic cause of pattern like that of CRPS. As CRPS is a debilitating condition with severe, chronic pain, autonomic dysfunction, and motor abnormalities, an early diagnosis and comprehensive management strategy should highly be considered in these cases.

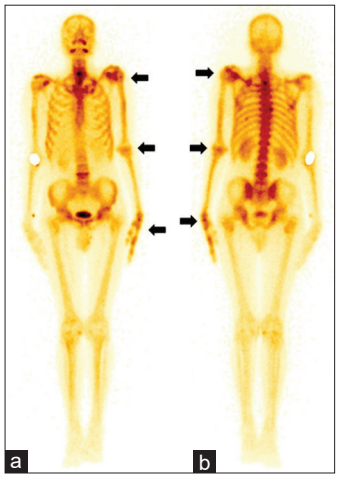

Figure 1: ^{99m}Tc methylene diphosphate bone scan. (a and b) Represent anterior and posterior images, respectively, which show increased tracer uptake in the left shoulder, left elbow, small joints of the left hand, and long bones of the left upper limb (black arrows)

Declaration of patient consent

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

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Nil.

Conflicts of interest

There are no conflicts of interest.

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