Esophageal Cancer: Presentation With Unusual Bone Metastases And Review Of Relevant Literature

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Abstract
Bone metastases in oesophageal adenocarcinoma are rare. We present a case of solitary scapular metastasis from an oesophageal adenocarcinoma arising in the lower end of the oesophagus.

INTRODUCTION
Oesophageal cancer is one of the deadliest cancers with a lifetime risk of about 0.8% for men and 0.3% for women. Incidence of oesophageal cancer is 13 cases per 100,000 population for black American men. On the whole it is the sixth commonest cause of cancer related deaths in the world. More than 50% of the oesophageal cancers are unresectable or have radiographically visible distant metastases at the time of diagnosis. Majority of the patients die within one year of diagnosis of oesophageal cancer and the 5 year survival rate is 8-20%. Incidence of adenocarcinoma of oesophagus continues to increase by 5-10% per year. There has been an increase in the relative incidence of adenocarcinoma compared to squamous cell carcinoma in the last three decades but the total incidence of oesophageal adenocarcinoma remains the same. In 2002, approximately 60% of oesophageal malignancies were adenocarcinomas. Men were eight times more commonly affected than women and whites were five times more commonly involved than blacks.

He had signs of dehydration. Systemic examination of the body revealed an obvious soft tissue swelling involving whole of the left scapula. Swelling was tender with all movements preserved at the shoulder joints. It was a well defined swelling movable separately from the chest wall. There was no wasting of musculature involving the left shoulder or upper limb. On further questioning he admitted that the swelling existed for some months but he did not pay much attention to it as it was not troubling him.

Haematological investigations were within normal limits. Serum calcium was also normal. Routine Chest roentgenogram (Fig 1.) revealed destruction of the scapula including the scapular spine. Lung fields and costophrenic angles were clear. Barium meal study was unremarkable.

CASE REPORT
89 year-old male presented with complaints of refusal to feed and vomiting for four days. There were no other complaints related to either gastrointestinal or any other system.
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Figure 1

Figure 1: Frontal chest roentgenogram shows destruction of the scapula. Medial border of the scapula is not seen except inferomedially.

Upper gastrointestinal endoscopy was done. There was a growth involving one third of the circumference of the oesophagus at 37cms extending up to the gastro-oesophageal junction. Stomach was within normal limits and there was no involvement of the cardia by the oesophageal growth on retroflexion. Endoscopic biopsy from the oesophageal growth revealed poorly differentiated adenocarcinoma. Immunohistochemical stains were positive for Carcinoembryonic antigen (CEA) and low molecular weight cytokeratin.

Computed Tomography (CT) of the chest and whole abdomen was performed with oral and intravenous contrast. CT scan (Fig 2.) revealed a large soft tissue mass around the left scapula with destruction of the medial and superior borders of the scapula including the blade and the spine of the scapula. Rest of the study was unremarkable.

Figure 2

Figure 2: Contrast enhanced CT scan of the chest reveals soft tissue swelling around the scapula with destruction of the scapula including the scapular spine

Fine needle aspiration cytology (FNAC) from the periscapular soft tissue swelling was reported to be poorly differentiated epithelial malignancy possibly metastatic adenocarcinoma.

A diagnosis of poorly differentiated adenocarcinoma of the oesophagus with solitary distant bone metastases in the scapula, pending the bone scan at a higher centre was made and further management consisting of palliative therapy discussed with the patient. However patient refused any treatment and left against medical advice.

DISCUSSION

This is a case of oesophageal adenocarcinoma presenting as a solitary scapular mass. In a review of 1909 cases of oesophageal cancer with 145 bone metastases there were six scapular lesions. Majority of the bone metastases from oesophageal cancer are solitary lesions basically because of the short life span (rarely more than three months) in these patients. Incidence of bone metastases was 5.2% in this study. [4]

Bone metastases in scapula and other locations are described as presenting features of oesophageal cancer but are very unusual. On an average 3-9% of bone tumours involve scapula. Overwhelming majority of scapular tumours are metastases predominantly from breast cancer. Commonest primary malignancy is chondrosarcoma. Majority of the scapular lesions occur within the compartment involving the scapula and its muscular envelope. Extra compartmental spread to involve the axillary neurovascular bundle rarely occurs. Majority of the bone metastases occur bones containing the red marrow predominantly such as spine, ribs,
pelvis and ends of long bones. [\textsuperscript{5}]

Metastases represent only 1% of the malignant lesions in oral cavity. Bony metastases of oral cavity involve mandible in 80- 90\% cases. Maxilla is involved in rest of the 10- 20\%. In 15\% cases perioral soft tissues are involved. Adherent gums are the commonest locations involved among the soft tissues with tongue next in the order. Oral cavity metastases arising from oesophagus are very rare. Diagnosis of oral metastases is a terminal event with death occurring within few months. In 30\% cases oral metastasis is the presenting feature of a distant malignancy. [\textsuperscript{5}]

Paranasal sinuses can also be a site for metastases and patients present with sinonasal pathology [\textsuperscript{7}]. Oesophageal cancer spreading to temporal bone can cause isolated facial palsy [\textsuperscript{8}].

Metastases to the small bones of hands and feet from oesophageal carcinoma are unusual. FNAC was found useful to confirm the diagnosis. [\textsuperscript{9}] Acrometastases sometimes present with whitlow- like lesions. [\textsuperscript{10}] Micro metastases are more common in rib marrow (88\%) compared to iliac crest marrow (15\%). Micrometastases are independent of the histological type of underlying malignancy or the nodal status. They are tumorigenic and are resistant to neo-adjuvant therapy. [\textsuperscript{11}] Carcinosarcoma of the oesophagus is known to produce metastatic osteosarcoma in lung, kidney and iliac bone. [\textsuperscript{12}]

Oesophageal carcinoma has been reported to cause saucerization which is always due to bronchogenic carcinoma unless proved otherwise. [\textsuperscript{13}] Hypertrophic osteoarthropathy is one of the manifestations of the oesophageal carcinoma even in the absence of metastases. [\textsuperscript{14}]

Hypercalcemia in cases of oesophageal malignancy is linked to unfavourable prognosis even in the absence of detectable bone metastasis. Survival at the end of one year after surgery in these patients was found to be significantly low in patients with hypercalcemia compared the controls. [\textsuperscript{15}]

Commonest complications by contiguous spread are tracheobronchial or bronchoesophageal fistulas. Lungs and any of the mediastinal components may be involved. Lymphatic spread is seen in 70\% cases with mediastinal, abdominal, cervical and supraclavicular lymph node enlargement. Sub mucosal lymphatic spread is responsible for intraesophageal and intragastric metastases. Most common locations involved by hematogenous spread are liver and lung. [\textsuperscript{13}]

Among the distant metastases 45\% were seen in abdominal lymph nodes. Rest were as follows: liver (35\%), lung (20\%), cervical / supraclavicular lymph nodes (18\%), bone (9\%), adrenal (5\%), peritoneum (2\%), brain (2\%) and stomach, pancreas, pleura, skin / body wall, pericardium and spleen (each 1\%). Distant metastases other than those in lymph nodes appear to be relatively more frequent in adenocarcinoma. In a large series of patients with oesophageal cancers 18\% were detected to have M1 disease, of which majority were diagnosed prior to surgery by CT scan of the chest and abdomen. Reported sensitivity of CT scanning was 70\%. [\textsuperscript{16}]

Oesophageal cancer was the commonest cancer responsible to produce intragastric metastases in the cardia up to 25\%. In these patients there was a high rate of lymph node recurrence and distant metastases such as lung and bone metastases. [\textsuperscript{17}] Oesophageal carcinomas are known to implant in the gastric ulcers. [\textsuperscript{18}]

Splenic metastasis is very rare from oesophageal cancer. [\textsuperscript{19}] Malignant ascites may be the presenting feature for oesophageal cancer. [\textsuperscript{20}] Metastases from oesophageal carcinoma are known to cause bilateral ureteric obstruction. [\textsuperscript{21}] Solitary renal involvement was also noted.

Skin metastases may be the first sign of internal malignancy in about 0.8\% cases of which 60\% remain undiagnosed. Spread to the skin indicates terminal illness [\textsuperscript{22}]. Therefore any nonhealing ulcers, persistent indurated erythemas and/ or cutaneous nodules of unknown cause should be biopsied to identify the underlying malignancy. Cutaneous metastasis may present as ulcerated lesions for example at the tip of the nose [\textsuperscript{23}].

It usually results from adenocarcinomas. Incidence of Sister Mary Joseph's nodule due to oesophageal carcinoma was found to be (0.2\%). FNAC is an adequate method of diagnosis. Contrary to earlier belief, it was demonstrated that patients with umbilical metastasis survived more than one year and also that treatment prolonged survival in these cases from 2.3 to 17.6months. Search should be done for occult malignancy or relapse of the already known cancers. [\textsuperscript{24}] A rare instance of an oesophageal squamous cell carcinoma causing umbilical metastasis has been described. [\textsuperscript{25}]
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Oesophageal cancer is known to spread to skeletal muscles. Ossification of such metastases from oesophageal cancer is a rare event. [2]

Oesophageal adenocarcinoma metastases have been reported in orbit. [3] Extra ocular muscle infiltration due to metastases from carcinoma of gastrooesophageal junction has been described in literature. Metastases are the commonest intraocular tumours among adults. Breast and lung are the commonest malignancies responsible. [4] Metastases to retina have also been reported in retina.

Leptomeningeal carcinomatosis is very rare secondary to oesophageal malignancy with grave prognosis. [5] Metastasis has been reported to brain. [6]

Metastases to oesophagus are rare and difficult to diagnose except at autopsy or surgery because many of them are asymptomatic. Breast is the commonest organ to metastasize to oesophagus. [7]

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