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OESOPHAGEAL CARCINOMA WITH DELAYED DISTANT METASTASIS IN SKELETAL MUSCLE: A RARE PRESENTATION

General Surgery

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ABSTRACT

Introduction Oesophageal carcinoma is the sixth most common cancer in the world, most commonly affects mid to late adulthood. Only 5-10% patients who diagnosed survive for five years, rest of the patients die in the due course of the disease. the most common pattern of esophageal cancer metastasis (ECM) is to the lymphnode, lung, liver , boones , adrenal gland and the brain. Sometimes unexpected distant metastasis also occur to unusal sites other then expected that is skeletal muscle in our case.

CASE REPORT We present a case of oesophageal carcinoma in a elderly gentleman with distant skeletal metastasis –a very rare delayed presentation of oesophageal carcinoma, & discuss about the various presentation and diagnostic modelities and the treatment options, with literature review.

In this case report we presented a case of oesphageal carcinoma who treated initially with three field oesophegectomy and with adjuvant chemoradaition. Later on in routine follow-up patient presented with hemetmesis, and on PET-SCAN incidental finding was an nodule in theright thigh suspecting of sarcoma-a new primary or a distant metastasis from the primary oesphageal carcinoma that later on turned out into metastatic adenocarcinoma from the primary oesophageal carcinoma. By doing through study and review of literature of more then 1000 pub med cases only 7-8 cases till date are reported for oesophageal carcinoma with distant metastasis to skeletal muscle, our case is very unique one in presentation, as none of the 7-8 reported cases , presented with delayed skeletal muscle(in our case almost after 3 yrs of curing the primary lesion), all are reported at the very initial presentation or with in 4-6 weeks of initial presentation, and probably one of the first case with such presentation(particulary in right thigh muscle).

CONCLUSION Oesopageal carcinoma mainly present distant metastasis to lymph nodes, lung, liver, bone while SMM represent a rare presentation in patients with oesophageal carcinoma, the incidence may be increasing because of better diagnostic tool available now a days, and because of better control of primary, a high clinical suspicion is needed along with PET-CT and histopathoogical examination to diagnose delayed skeletal muscle distant metastasis, an active therapeutic multidisciplinary approach including surgery, radiation and chemotherapy may result in better survival that is longer in comparison to other metastaic esophageal carcinoma cases.

KEYWORDS

Oesophageal carcinoma, distant metastasis, SMM(skeletal muscle metastasis), PET-SCAN

Introduction

Esophageal carcinoma is the sixth most common cancer in the world, most commonly affects mid to late adulthood. Only 5-10% patients who diagnosed survive for five years, rest of the patients die in the due course of the disease. the most common pattern of esophageal cancer metastasis (ECM) is to the lymphnode, lung, liver, bones, adrenal gland and the brain, pleura, pericardium or peritoneum, less frequent sites of metastasis include thyroid, pancreas, spleen, ovary kidney, heart, skin, gallbladder or small and large bowel[1-2] Sometimes unexpected distant metastasis also occur to unusual sites other then expected that is skeletal muscle in our case.

CASE REPORT

We present a case of esophageal carcinoma in a elderly gentleman with distant skeletal metastasis –a very rare presentation of esophageal carcinoma, & discuss about the various presentation and diagnostic modalities and the treatment options, with literature review.

In this case report we presented a case of 64 year gentleman initially presented with complaints of swallowing of solid food, gaseous distension of abdomen and generalized weakness and weight loss, on doing detailed work-up including clinical examinations and certain investigation (CT Abdomen with enterography , gastroduodenoscopy which showed GE Junction growth, biopsy showed Adenocarcinoma GE junction. Initial PET-CT Scan (nov. 14) done which showed no distant metastasis except some peritoneal and mediastinal lymp nodes. Patient was managed with robotic assisted transthoracic total esophagectomy with partial gastrectomy with gastric pull up esophago-gastrostomy with feeding jejunostomy under GA (NOV. 14).surgical pathology report showed well differentiated adenocarcinoma with nodal metastasis(PT3N3Mx).patient also received adjuvant chemoradiation. Patient tolerated the procedure well, & was apparently asymptomatic,on routine follow-up detailed investigation including PET-CT whole body performed at various stages, nothing abnormality detected until he presented in surgical emergency with complaints of hematemesis and weakness, UGI Endoscopy was done which showed an active bleeding duodenal ulcer which was managed with endoscopic hemostatic plug application and inj. Sclerotherapy, as the patient was known case of carcinoma esophagus, PET-CT Scan also performed to detect any new primary , remanant or any delayed metastasis, which shows FDG avid lesions in anterior compartment of right thigh suspecting of sarcoma-a new primary or a distant metastasis from the primary oesphageal carcinoma that later on doing FNAC –consistent with metastatic adenocarcinoma and on histopathological examination of excised lump confirmed delayed distant metastasis from esophageal carcinoma to right thigh skeletal muscle.PET-CT also showed FDG avid mediastinal nodes and FDG avid lesions in right ribs also likely metastasis, but no recurrence to primary anatomical site. later on patient eventually died because of disease morbidity and as he developed bilateral pulmonary embolism with hemodynamic instability.

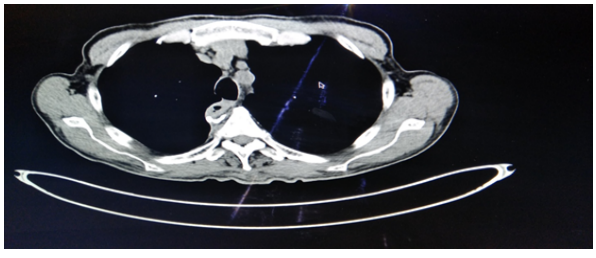

Fig.1-showing no remnant or recurrent disease at primary site

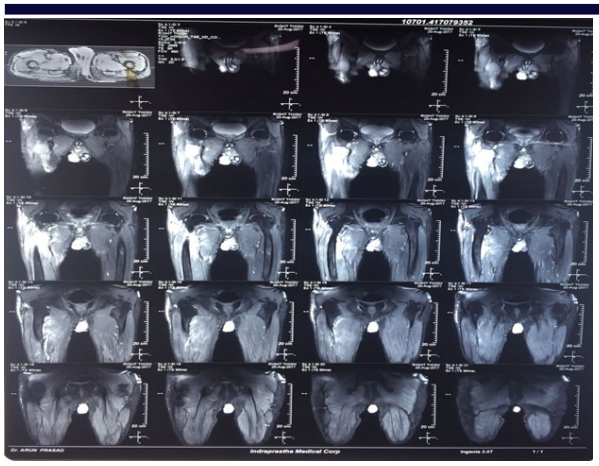

Fig.2-showing right thigh muscle mass(metastasis)

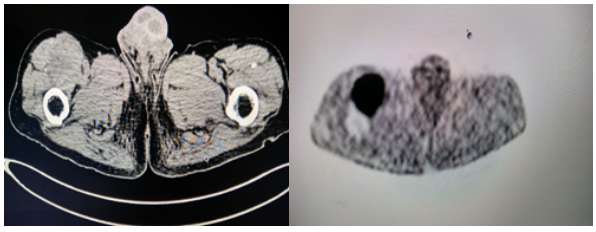

Fig.3,4-showing hypoechoic, heterogenous mass showing tracer accumulation in right anterior thigh muscle

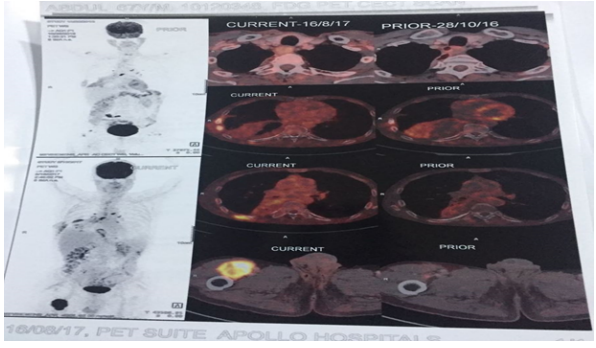

Fig.5-showing FDG avid lesion on PET-CT, also showing comparison to previous one

DISCUSSION

The prognosis of esophageal carcinoma is very poor, very few patients (10%) survive for a longer time. carcinoma esophagus usually metastasizes to lymph nodes, liver, lung, adrenals and bone & less frequently to kidney, heart or gall bladder. Skeletal muscle metastasis is very rare from carcinoma esophagus and so from solid tumours (<1%)[3-6]. Most common primary tumour sites for skeletal muscle metastasis are lung(35%), gastrointestinal(23%) and kidney(19%)[7]. Other primary sites included melanoma, head and neck cancer, thyroid, breast, uterus, cervix, prostate, bladder,ovary , pancreas, and liver[7-9]. By doing through study and review of literature of more than 1000 pub med cases only 7-8 cases till date are reported for esophageal carcinoma with distant metastasis to skeletal muscle, our case is very unique one in presentation, as none of the 7-8 reported cases, presented with delayed skeletal muscle(in our case almost after 3 yrs of curing the primary lesion), all are reported at the very initial presentation or with in 4-6 weeks of initial presentation, and probably one of the first case with such presentation (particulary in right thigh muscle). A study done by Jan Cincibuch et al reported only 2% incidence (all were male) of skeletal muscle metastasis while studying of 205 patients for more than period of 4 years, in out of 5 cases 3 were diagnosed at the same time while doing staging FDG PET –CT, one after 5 months of initial diagnosis and one after 2 years of initial diagnosis, two were reported as adenocarcinoma same as in our case and rest of the three as squamous cell carcinoma, in their study SMMs were the first manifestation of metastatic disease as in our case, SMM involvement usually preceed the involvement of many other organs in metastatic diseases by several months, and survival time is relatively better in the context of esophageal carcinoma[10]. In only 1-2% of all patients with metastatic disease are reported to SMM, with metastatic involvement limited to skeletal muscle in less than 10 % of cases[11]. In cases of isolated SMM, best results may be obtained with the surgery alone, while in case of inoperable or recurrent SMM, external beam radiation, or in cases of multiple metastasis it should be combined with platinum based chemotherapy. A study done by Shaheen O et al.showed metastasis to unexpected sites in esophageal carcinoma, they concluded metastasis were disseminated to five main anatomical sites; the head and neck(42%), thoracic(17%), abdomen and pelvis(25%), extremities(9%) and multiple skin and muscle metastasis(7%)[12]. A Study done by Wang YX et al. in 395 patients showed a overall failure rate of 75.7%(299/395) with locoregional recurrence found in 208 patients, 26.9%(56) recurred in supraclavicular /neck, 69.7% in mediastinum and 19.7% in upper abdomen, so they concluded upper mediastinum is the most common site of recurrence, followed by supraclavicular and para-aortic regions, they didn't find even a single case of skeletal muscle metastasis[13]. Lymph node metastasis is most common pattern of recent relapse after esophagectomy[14] while most common site of distant metastasis was the lung and bone[15]. Similar study done by Dminguez et al. showed increased tracer uptake in subcutaneous node in abdomen initially ,later on in several muscles as isolated distant haemtaogenous spread of metastasis[16], as most of the skeletal muscle metastasis are asymptomatic as same in our study, so it is recommended that physician should have high index of clinical suspicion and every focal FDG uptake should be thoroughly evaluated by histopathological examination, because they may be the first sign of disease spread, so earlier the diagnosis, better could be the treatment & survival. Locoregional recurrence is due to tumour progress related to the extent of lymph node metastasis ,distal recurrence usually depends upon oncological behavior of the tumor, and for this vascular invasion found to have been the most important prognostic factor[17]. Fujita H et al. in their study divided the recurrence after resection of thoracic esophageal cancer into 5 categories according to site of recurrence-1. Local recurrence 2. Recurrence at the anastomotic sie, 3. Recurrence in cervical or mediastinal lymph nodes 4. Recurrence in abdominal lymph nodes and 5. Distant organ metastasis[18]. Makoto Sohda et al reported a similar case of primary esophageal carcinoma with distant skeletal muscle metastasis to skeletal muscle with in 3 weeks of primary presentation, taken up by FDG-PET[19]. Up to that study only 4 cases have been described in the literature[20-23]. Although direct muscle invasion is very well known but distant metastasis to skeletal muscle is not so very common, soft tissue mass found in clinical finding on routine follow-up is usually mistaken as sarcoma or some other benign tumors. In our patient FDG-PET proved very useful in detection of right thigh metastasis. This case demonstrates importance of FDG-PET in a clinically high suspicion case for wide spread metastasis, histopathological examination is mandatory to further establish the diagnosis. So, FDG-PET is very important diagnostic tool for cancer staging and for effective treatment. Skeletal muscle metastasis often presents with painful mass in a known primary case[24]. Skeletal muscle metastasis is very rare from esophageal carcinoma, exact cause is unknown, some study suggested that contractile activity, changes in pH, accumulation of metabolites, intramuscular blood pressure or local temperature might be the cause behind this rarity[25-27]. Study done by Norris WE et al. also reported a case of right hip pain that on detailed investigation ultimately turned out into primary esophageal carcinoma with distant metastasis to skeletal muscle[28], while Wu G in a separate case report showed primary esophageal carcinoma with distant metastasis to right gluteus minimus muscle[29], the details in these cases reported here are consistent with our findings, only difference is that in our case it presented as delayed presentation almost after 3 years of curing the primary, and it presented as asymptomatic incidental finding on proper work-up. 2 Similar cases of skeletal muscle metastasis one in forearm and one in right gluteus maximus as primary distant metastasis from esophageal carcinoma reported by Keijii et al[30].

Osama Shahen et al in their study of "esophageal cancer metastases to unexpected sites: a systemic review discussed that small vessels form abundant submucosal plexus which could explain the existence of distal unexpected metastasis[31] while complex anatomical pathway of esophagus lymphatic network including the lymphatic nodal skip(retrograde and bidirectional) spread is also a possible explanation of the random distribution of metastasis in esophagus cancer[32-34]. Bruzzi et al. showed importance of integrated CT-PET in detecting distant metastasis those are unusual in appearance and locations[35] while Nguyen et al. showed in their results 7.7% of unexpected soft tissue metastasis while using F-18 FDG PET/CT[36]. So both these

studies and our case study hypothesized that limited scanning may underestimate the true extent of soft tissue metastasis, because sometime they have very unusual and atypical presentation as in our case. Olga N. et al reported a case of esophageal carcinoma with multiple asymptomatic metastasis to skeletal muscles[37] and as in our study supports that PET is superior to CT in detecting metastasis.

CONCLUSION

Esophageal carcinoma mainly present distant metastasis to lymph nodes, lung, liver, bone while SMM represent a rare presentation in patients with esophageal carcinoma, the incidence may be increasing because of better diagnostic tool available now a days, and because of better control of primary, a high clinical suspicion is needed along with PET-CT and histopathological examination to diagnose delayed skeletal muscle distant metastasis, an active therapeutic multidisciplinary approach including surgery, radiation and chemotherapy may result in better survival that is longer in comparison to other metastatic esophageal carcinoma cases. Since most Esophageal carcinoma related deaths are related to distant metastasis , an extensive study is further needed to understand the pattern of the disease. The metastasis to skeletal muscle is very late event in a carcinoma and in esophageal carcinoma overall survival is very poor , so early detection is very important and palliative treatment with multimodality approach should be done with minimal discomfort .

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