

A case of oesophageal cancer with low back pain: the accidental finding of skeletal muscle metastasis



Ann. Ital. Chir., 2013 84: 193-195

pii: S0003469X12018581

www.annitalchir.com

Giovanni Leuzzi*, Alfredo Cesario**/, Stefano Margaritora**/, Anna Mariantonia Parisi*, Venanzio Porziella*, Elisa Meacci*, Maria Letizia Vita*, Maria Teresa Congedo*, Marco Chiappetta*, Pierluigi Granone*

*Department of General Thoracic Surgery, Catholic University of Sacred Heart, Rome, Italy

**Deputy Scientific Director, IRCCS San Raffaele Pisana, Rome, Italy

***Department of Thoracic Surgery, Centro Oncologico Fiorentino, Florence, Italy

A case of oesophageal cancer with low back pain: the accidental finding of skeletal muscle metastasis

Skeletal muscle metastasis is a very rare event in patients with oesophageal cancer. We herein report and discuss a case of a 65 years old man with history of gastro-oesophageal reflux disease referred to our department for pyrosis associated to persistent low back pain. Oesophageal endoscopy and transoesophageal endo-sonography showed a tumour localized in the lower third of the esophagus, histologically proved to be adenocarcinoma. Clinical staging procedures detected a two centimetres vascularized nodular lesion placed into right para-vertebral muscles at the level of L4 as the only sign of potential distant disease (versus a differential diagnosis of primitive sarcoma). The muscle lesion was completely removed and confirmed as secondary adenocarcinoma. Due to this evidence a chemotherapy protocol was initiated. After nine months the patient underwent transhiatal oesophagectomy.

To the best of our knowledge this is the first reported case of a soft tissue metastasis from oesophageal cancer resected with radical intent.

KEY WORDS: Low back pain, Oesophageal adenocarcinoma, Skeletal muscle metastasis

Introduction

In general, soft tissue metastases are rare. Sporadic reports exist of cancers from the gastrointestinal tract, the lung, leukaemia and lymphomas and urinary bladder metastasizing at the level of the muscle. [1]. Oesophageal cancer (OC) does metastasise, normally, in regional lymph nodes. So far and to the best of our knowledge only

four cases have been reported in the English literature of muscle metastasis from OC and none of these involved surgical treatment of the secondary lesion with radical intent.

Case report

A 65 years old man was admitted to our department because of pyrosis associated with a two month history of gradually increasing and persistent low back pain (LBP). His medical history included gastro-oesophageal reflux disease (GERD) and multiple rib fractures because of previous chest and spinal blunt trauma. He was an active smoker, denied dysphagia and recorded recent unintentional weight loss, with no significant changes in his dietary habits. Neurological examination and standard lab tests were unremarkable. Oesophageal endoscopy and trans-oesophageal endo-sonography showed a 2 cen-

Pervenuto in Redazione Novembre 2011. Accettato per la pubblicazione Gennaio 2012

Correspondence to: Giovanni Leuzzi, MD, Department of General Thoracic Surgery, Catholic University of Sacred Heart, Rome, Italy. Largo F. Vito 1, 00168, Rome, Italy (e-mail: gio.leuzzi@yahoo.it)

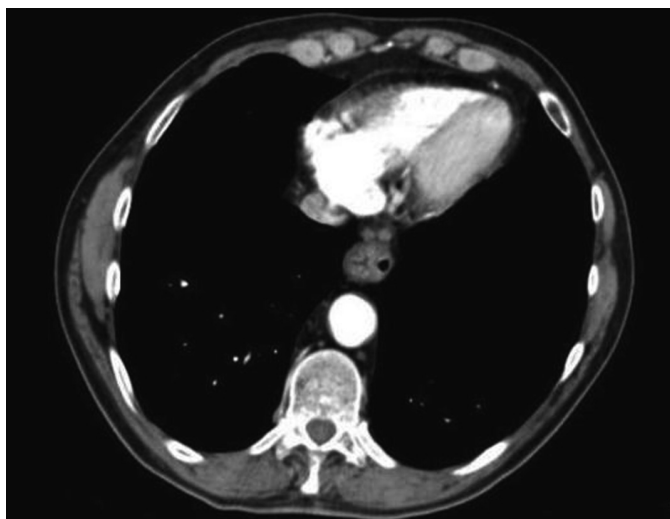


Fig. 1: A whole body CT scan shows the oesophageal adenocarcinoma of the lower third of the oesophagus (as confirmed by oesophageal endoscopy and transesophageal endosonography) and reveals a 2 centimetres intramural mass with 4 centimetres longitudinal extent occupying 50 % of the circumference of the oesophagus.



Fig. 2: CT demonstrates also an incidental 22 millimetres vascularized nodular lesion localized into right para-vertebral muscles at the level of L4. Radiological diagnostic consideration includes a soft tissue metastasis or a primary muscle sarcoma.

timetres intramural mass with 4 centimetres longitudinal extent, occupying 50 % of the circumference of the lower third of the oesophagus. No pathological lymph nodes were detected. Histopathological examination of endoscopic biopsy demonstrated an adenocarcinoma infiltrating the *muscularis propria*. Because of worsening LBP, a spinal X-ray was performed and this did not show any pathological finding. Staging procedures (including whole body computed tomography -CT- scan) confirmed the evidence of the lesion in the lower third of the oesophagus (Fig. 1) and an incidental 22 millimetres vas-

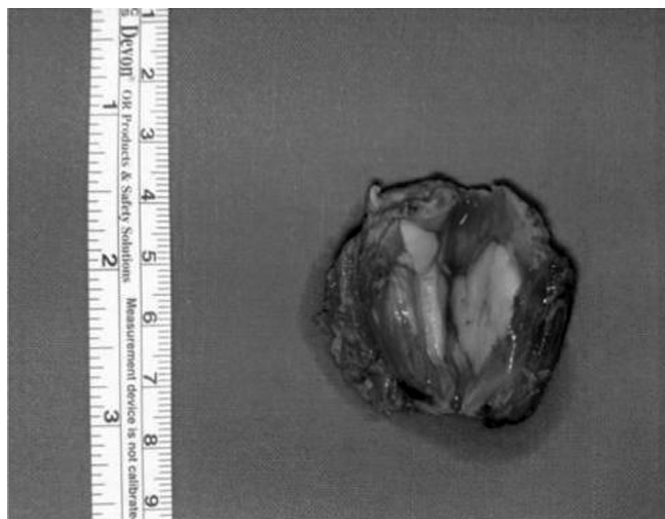


Fig. 3: Macroscopic findings: muscle nodule appears as a 2 centimetres whitish unencapsulated sclerotic lesion. Final histological examination confirmed the presence of glandular epithelial cells proliferation with pattern of cytokeratins compatible with oesophageal adenocarcinoma metastasis.

cularized nodular lesion localized into the right para-vertebral muscles at the level of L4 (Fig. 2). Radiologically, the differential diagnosis of the muscular lesion was among a metastasis or a primitive sarcoma. A radionuclide bone scan was negative for elements of interest. Circulating tumour markers (CEA, CA 19.9, alpha-FP, CA 15.3, etc.) were all within normal range. After a multidisciplinary meeting, a surgical approach was indicated for the muscle lesion, deemed to be resectable. Oesophagectomy was planned in a staged approach after verification of the origin of the para-vertebral soft tissue lesion.

Therefore, the patient underwent radical excision of muscle nodule, which appeared as a 2 centimetres whitish sclerotic lesion without any evident capsule (Fig. 3). Intra-operative histology was strongly suggestive for metastatic adenocarcinoma; this diagnosis, supported by the presence of glandular epithelial cells proliferation with pattern of cytokeratins at final pathology, was confirmed and determined the subsequent therapeutic approach consisting of first step chemotherapy.

The patient was discharged at the first pod and, after nine months, he underwent transhiatal oesophagectomy.

Discussion

Oesophageal cancer ranks eighth among the most common cancers worldwide and represent the sixth commonest cancer related cause of death. Parkin et al. showed that each year 462,000 people are diagnosed with OC and 386,000 people die from it ². Most of the OCs are squamous cell carcinomas; however there is an increase in western countries of the relative and absolute

number of adenocarcinomas of the lower third of the esophagus, related to the increasing prevalence of Barrett's esophagus³.

Overall 242 cases only of skeletal muscle metastasis (every primary) have been reported and these more often occur when the primary is in the gastrointestinal tract, the lung and in the urinary bladder. To a minor extent, leukaemias and lymphomas are concerned as well¹. A muscle metastasis from OC (adenocarcinoma) is a very rare event and, to the best of our knowledge it has been described in four cases only⁴⁻⁷.

Herring et al. retrospectively reported about 15 patients with metastatic spread to skeletal muscle and speculated about the relevance of different factors in relation with the rarity of this phenomenon. In fact, although skeletal muscle has a rich blood supply, blood flow is variable and influenced by modifications in pH (due to production of lactic acid which is different at rest and during exercise) temperature, blood pressure and beta-adrenergic receptor expression⁸.

Muscle metastases are generally accompanied by evident clinical signs: palpable mass, pain, etc. However they can be asymptomatic, depending on their localization. As reported in previously mentioned case reports⁴⁻⁷, 3 out of the 4 cases presented with pain. In the case reported by ourselves, the patient developed a progressive LBP, caused by metastatic growth into the paravertebral muscle. Surgical removal completely resolved this symptom.

So far, even if MRI is considered superior to CT in featuring and characterizing soft tissue tumors⁹, imaging cannot resolve the differential diagnosis among primitive sarcoma and secondary lesions. For this reason we did not perform MRI (or further characterize the mass by fine needle aspiration biopsy) and decided for surgical removal with the double aim of getting a final diagnosis and curing the symptoms. This behavior would have been entirely appropriate even if the mass had proven to be a sarcoma¹⁰. In our subsequent clinical work-up we then concurred with Herring^[8] on the criteria to apply post-operative radiotherapy (combined with chemo) on the metastatic site only if the disease is extensive or the resection is deemed incomplete. None of these scenarios applied to our case, and then no adjuvant treatment was applied.

In conclusion, muscle metastasis is a rare event, rarest in oesophageal cancer. It should be ruled out, with targeted tests within the clinical staging of any tumour, in all cases where muscular pain is reported. Surgery on the metastatic site should be considered in the comprehensive scenario of the treatment of the primitive cancer and according to its staging. However, a complete resection in cases like the one reported herein could achieve a very good control of the symptoms (and therefore impact positively on the patient's quality of life). It provided, as well, accurate staging and final pathological definition.

Riassunto

Le metastasi muscolari sono estremamente rare nei pazienti con tumore dell'esofago.

Si riferisce del caso di un uomo di 65 anni con storia di reflusso gastro-esofageo che è stato ricoverato presso la nostra Unità Operativa per la comparsa di pirosi e dolore lombare destro. Il paziente è stato sottoposto ad esofagogastrosopia ed eco-endoscopia che hanno documentato un adenocarcinoma del terzo inferiore dell'esofago. Gli esami radiologici preoperatori hanno evidenziato inoltre la presenza di un'unica lesione nodulare ipervascolarizzata di 2 cm localizzata all'interno dei muscoli paravertebrali di destra a livello di L4 compatibile con metastasi muscolare o con sarcoma primitivo. Il paziente è stato quindi sottoposto ad asportazione radicale della lesione suddetta il cui esame istologico definitivo ha documentato una metastasi da adenocarcinoma. In considerazione di questa evidenza isto-patologica, il paziente ha intrapreso un trattamento chemioterapico e, dopo nove mesi, è stato sottoposto ad intervento chirurgico di esofagectomia transiatale.

In letteratura, questo è il primo caso in cui è stata resecata una singola metastasi muscolare da tumore esofageo con intento radicale.

References

1. Menard O, Parache RM: *Les metastases musculaires des cancers (Muscle metastases of cancer)*. Ann Med Interne (Paris), 1991; 142:423-28.
2. Parkin DM, Bray F, Ferlay J, Pisani P: *Global cancer statistics, 2002*. CA Cancer J Clin, 2005; 55:74-108.
3. Vizcaino AP, Moreno V, Lambert R, Parkin DM: *Time trends in incidence of both major histologic types of esophageal carcinomas in selected countries, 1973-1995*. Int J Cancer, 2002; 99:860-68.
4. Rehman SU, Cope DW, Basile JN: *Metastatic gastroesophageal adenocarcinoma to skeletal muscle: a unique event*. South Med J, 2002; 95:1076-78.
5. Heyer CM, Rduch GJ, Zgoura P, Stachetzki U, Voigt E, Nicolas V: *Metastasis to skeletal muscle from esophageal adenocarcinoma*. Scand J Gastroenterol 2005; 40:1000-4.
6. Wu G, Bybel B, Brunken R, Lin H, Neumann D: *PET detection of solitary distant skeletal muscle metastasis of esophageal adenocarcinoma*. Clin Nucl Med, 2005; 30:335-37.
7. Norris WE, Perry JL, Moawad FJ, Horwhat JD: *An unusual presentation of metastatic esophageal adenocarcinoma presenting as thigh pain*. J Gastrointest Liver Dis, 2009; 18:371-74.
8. Herring CL Jr, Harrelson JM, Scully SP: *Metastatic carcinoma to skeletal muscle. A report of 15 patients*. Clin Orthop Relat Res 1998; (355):272-81.
9. Berquist TH: *Magnetic resonance imaging of musculoskeletal neoplasm*. Clin Orthop, 1989; 244:101-18.
10. Lawrence W Jr, Donegan WL, Natarajan N, Mettlin C, Beart R, Winchester D: *Adult soft tissue sarcomas, a pattern of care survey of the American College of Surgeons*. Ann Surg, 1987; 205:349-59.

