

Acute monoparesis onset in recent aortic valve replacement: a case report

E. Satta, C. Soavi, M.A. Miselli, A. Passaro, G. Zuliani

Clinical and Experimental Medicine Department, Section of Internal and Cardio-Pulmonary Medicine, University of Ferrara, Italy

Vertebral metastases are frequent in patients with cancer. They are much more frequent in higher age groups (> 50 years); the lesions can be asymptomatic despite a setting of widespread metastatic disease and may become symptomatic due to bone pain, pathological compression fractures, or extension into spinal canal with cord compression ensuing neurological symptoms.

We report the case of a patient, without known history of malignancy, in which shoulder pain was attributed for few months to sequela of recent cardiac surgery for ascending aortic aneurysm. She was admitted to our ward because of acute onset of lower limb monoparesis which evolved in several days in paraparesis. Only a dorsal magnetic resonance study revealed the presence of a bulky vertebral lesion at D1-D2 level involving the peri-dural space. Neuro-surgical decompression was performed obtaining specimens for histological analysis, which suggested the presence of metastatic adenocarcinomatous lesions of gastrointestinal origin. A computed tomography study partially supported this hypothesis showing only a thickening of rectal wall, even if endoscopic exploration did not show macroscopic mucosal abnormalities. Surgical and medical therapies did not improve the patient's clinical course and she died few months later.

Key words: Spinal metastasis, Rectal cancer, Paraparesis, Spinal cord compression

CASE REPORT

A 72 year old female presented to Emergency Room referring acute onset of left leg weakness started two days before; she could not walk. She first underwent a neurological evaluation (left leg monoparesis) and brain CT scan (negative); then, the patient came to our ward for clinical observation.

Her medical history revealed arterial hypertension in good pharmacological control and a previous surgical correction of cystocele. Moreover, six months before she had undergone surgical correction of ascending aortic aneurysm associated with aortic valve insufficiency with reconstruction by composite valve graft. Post-operative period was characterized by pain to

left upper extremity and shoulder; the symptom was considered a normal surgical sequela and was treated with analgesics. She was currently treated with anti-platelet therapy, anti-hypertensive drugs, minor opiates, acetaminophen, pregabalin and paroxetine to control chronic pain.

At admission to hospital, the patient was cooperative and orientation in time and space were good. Neurologic examination revealed left leg paresis with associated tactile hypoesthesia below the knee, but normal deep tendon reflexes. In the first hours after admission the patient referred inability to voluntarily pass urine, with evidence of urinary retention. Her vital signs were stable, with only asymptomatic bradycardia attributable to beta-blockade therapy; pulmonary and

cardiovascular examination were normal. Initial laboratory tests showed no abnormalities in haematological parameters, coagulation, renal and hepatic function.

In order to investigate the monoparesis we asked for carotid ultrasound examination (negative), brain scan at 48 h from admission (negative) and electromyography (altered central motor conduction at lower limbs, in particular at the left side).

Because of the persistence of left shoulder and upper extremity pain, we speculated on aortic dissection related to the recent surgical correction, involving medullar vessels and asked for chest and abdominal CT scan; no aortic aneurysms or dissections were found, while at D1 it is observed a collapse of the vertebral body with extended replacement of bone by new tissue; also multiple lymphonodes in mediastinic and peritoneal space were described, together with thickening of rectal wall. In suspicion of a malignancy, additional laboratory tests were performed and rectal endoscopic studies were performed. A significant elevation of Ca 19.9 (798 U/ml, n.v. 27) was observed; colonoscopy was negative while sigmoidoscopy revealed normal mucosal pattern; thus, the endoscopist decided not to obtain bioptic specimens.

In addition the patient underwent cervical and dorsal MRI study. A bulky vertebral lesion was revealed at D1-D2 level involving the peri-dural space. The medullary cord on T2 weighted sequences appeared hyperintense, suggesting a parenchymal suffering from C7 to D2 (Fig. 1).

The lesion also involved the left conjugation foramen, reached the spinous process and, in part, the soft paravertebral tissue. In the meanwhile we noticed an evolution of neurological picture, with appearance of hypostenia at right lower limb and global anesthesia below D4 dermatome.

After a high dose steroid course of 4-5 days and discontinuance of antiplatelet therapy, D1-D2 laminectomy was performed. Pathologic examination showed neoplastic cells with CDX2 +/- CK7 +/- CK20 + phenotype, suggesting a gastrointestinal-pancreatic origin. Further molecular analysis documented the presence of *KRAS* mutation in exon 2 with G12V substitution.

The patient was taken in charge by oncologists. A first line therapy was made but during the hospitalization patient's condition quickly impaired because of the onset of flaccid paraparesis, thus she was assigned to an Oncology Palliative Care Unit where she died four months later.

DISCUSSION

Spinal metastasis is common in patients with cancer, constituting the third metastatic site, following the lung and the liver^{1,2}. Approximately 5-30%³ of patients with systemic cancer will have spinal metastasis. Generally, only 10% of patients are symptomatic⁴, and the majority of them present with epidural and/or vertebral involvement. Back pain is the most common presenting

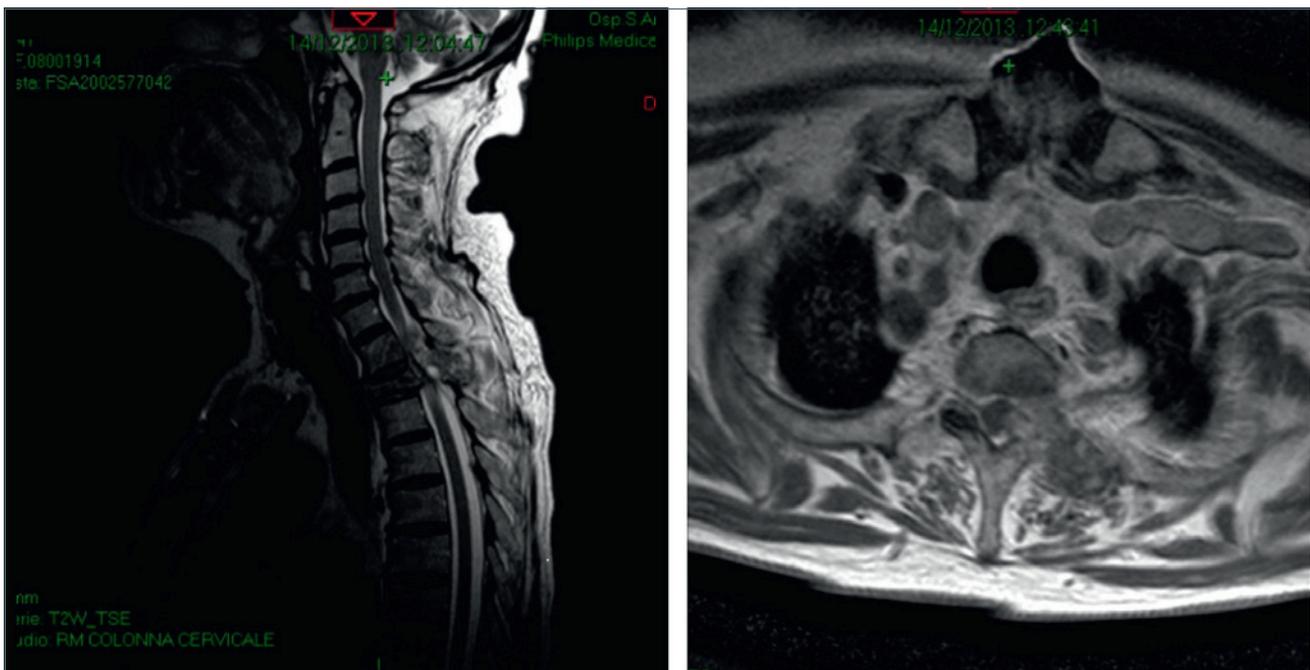


Figure 1. Cervical dorsal spine RMN shows vertebral (D1) involvement and spinal cord compression.

symptom and can precede the development of neurologic symptoms by weeks or months. Intradural extramedullary and intramedullary seeding of systemic cancer is unusual; this condition accounts for 5-6% and 0.5-1% of spinal metastases, respectively. Bone marrow metastases are an uncommon mode of onset of malignant disease (10%). Furthermore in only about 7% of symptomatic patients, the primary tumour remains occult⁵. The most common primary neoplasm involving vertebrae include breast, lung, prostate cancer, lymphoma, melanoma, and renal cell carcinoma.

In our case the diagnosis was difficult, due to two aspects: 1) the recent heart surgery was misleading for the understanding of the pain reported by the patient, thereby causing only a symptomatic therapy for few months; 2) the absence of symptoms suggesting a specific organ involvement, particularly change in bowel habit, intestinal bleeding, and weight loss made it difficult to identify the primary tumour.

Based on the instrumental relief of thickening of the rectal walls, and the phenotype of the cytological metastatic tissue, we hypothesize the presence of a rectal cancer. Isolated bone metastases from colon-rectal cancer are unusual, but some cases have been reported in literature⁶⁻⁸, although bone involvement is rarely described as a manifestation of colon-rectal cancer onset⁹⁻¹¹. Skeletal metastases generally appear when the disease is in an advanced stage; in this context the bone involvement is mostly multifocal and typically liver and lung metastases coexist¹². The lack of involvement of liver and lung may be explained by the existence of the venous plexus of Batson¹³. This is a system extended from sacrum to cranium that forms a large capacitance venous system and communicates with the other venous systems through segmental vessels; it is widely anatomised with the spinal venous system, the azygos vein, the dorsal and the intercostal veins. The plexus has not valves; thus, each increase in the pressure in the system of vena cava might result in an increased flow level of the plexus. In our patient, the heart failure secondary to aortic valve insufficiency may have played a pathophysiological role.

We also speculated that the rapid course of disease is well explained by *KRAS* mutation and above all by G12V substitution; *KRAS* phenotype is known to be associated with poorer prognosis in gastrointestinal tumor, increased risk of recurrence and death¹⁴. Especially the presence of a valine in codon 12 adversely affected overall survival¹⁴ and it has recently been analyzed in a murine experimental model¹⁵: increased metastatic propensity and a higher growth rate in lymph node metastases was observed in this oncogene mutation in agreement with clinical higher aggressiveness observed in patients with CRC.

CONCLUSIONS

Our case draws attention to several key-points. First, the presence of a persisting musculoskeletal pain, even if a plausible aetiology is identified, requires a careful diagnostic work-up to rule out a neoplastic involvement. Second, the absence of liver and lung metastases in a patient with suspected colorectal cancer must not represent an exclusion criteria, since other ways of spread can by-pass these organs (i.e. Batson's plexus). Of consequence, colon-rectal cancer should be included in the differential diagnosis of a solitary bone metastasis.

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