Case report

Pyogenic spondylodiscitis and osteoradionecrosis of the cervico-thoracic spine: a rare complication after surgery and radiation for hypopharyngeal cancer

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Abstract. *Objective:* To describe a rare case of osteoradionecrosis and pyogenic spondylodiscitis of the cervico-thoracic spine after surgery and radiotherapy for hypopharyngeal cancer. *Methodology:* Diagnostic clues are presented and critically discussed with particular regard to computed tomography, magnetic resonance and positron emission tomography features advising the association between and osteoradionecrosis and spondylodiscitis. *Results and Conclusions:* Pathogen isolation via CT-guided biopsy provided the definite diagnosis and indicated the targeted therapy. Successful recovery was obtained with conservative treatment. (www.actabiomedica.it)

Key words: cervical osteoradionecrosis, spondylodiscitis, hypopharyngeal cancer

Introduction

Osteoradionecrosis involving the cervical spine after surgery and radiotherapy for head and neck cancer is an extremely rare condition. It has been hypothesized that the effect of irradiation and surgery-related devascularization provide a poor environment to fight infection or heal wounds. As a consequence, osteoradionecrosis represents in the spinal area a possible nidus for spondylodiscitis (1). Diagnosis is usually difficult and delayed because these are very rare diseases and initial signs and symptoms are non-specific (2). Severe complications such as neurological deficit, paravertebral abscess, vertebral instability and progressive vertebral deformity may arise from a cervical osteoradionecrosis and spondylodiscitis. We describe a rare case of osteoradionecrosis and spondylodiscitis of the cervico-thoracic spine after surgery and radiation for hypopharyngeal cancer.

Case report

A 68-year-old man was treated at our department for a squamous cell carcinoma of the right-sided pyriform sinus' lateral wall (cT2N2aM0) with partial pharyngectomy through a lateral pharyngotomy approach and ipsilateral modified radical neck dissection with sacrifice of the internal jugular vein. The pathologist reported clear surgical margins and the clinical staging was confirmed. Post-operative course was uneventful.

The patient subsequently received 3D-conformal radiotherapy by two lateral fields from skull base to the clavicle, with conventional fraction of 2 Gy/die for 5 days a week, until a cumulative dose of 45 Gy; tumour bed and metastatic neck regions recieved a total dose of 58 Gy during 43 days.

Four months later, a follow-up CT evaluation of the neck showed a large, irregular and contrast enhanced mass wrapping the cervical bodies from C5 to

T4, diffuse demineralization of vertebral bodies involved, osteolytic lesion of C5, collapse with cuneiform deformation of C6, gas bubbles in the prevertebral space and anterior epidural space. Tumour recurrence associated with an inflammatory-necrotic process was suspected, thus the patient was re-admitted at our department. Clinical examination was unremarkable and the patient complained only of a mild reduction of neck movements. He was apyretic. Neurological evaluation found no signs of spine or neural compression. He had no history of recent trauma. Broad spectrum antibiotic treatment was administered. Magnetic resonance (MR) confirmed the presence of a contrast enhanced mass involving the retropharyngeal and prevertebral space with involvement of vertebral bodies and intervening disks from C5 to T4. Neural foramina appeared also involved. There was a slight compression of the dural sac at the level of T1 (Figure 1). Positron emission tomography (PET) scan showed the lesion to be hypermetabolic. Haematochemical tests and inflammation indicators revealed alterations of ESR and CRP (56 mm/h and 114 mg/L respectively) while white blood cell count was normal.

A CT-guided biopsy performed at the level of T3-T4 showed fibroconnective tissue, marrow fibrosis with focal necrosis and plasma cell infiltrate consistent with an inflammatory process. No neoplastic cells were found. Microbiological work-up was positive for Streptococcus mitis. Therefore osteoradionecrosis with pyogenic spondylodiscitis was diagnosed.

A targeted intravenous antibiotic treatment with linezolid 600 mg x 2 was administered. Inflammatory indicators gradually decreased until normalization and the patient was discharged with oral amoxicillina clavulanate (1 g x 3 for 3 months) and cervical immobilization after 7 days of hospitalization. A follow-up CT scan performed 3 months later demonstrated a nearly complete disappearance of the inflammatory disease and vertebral healing process toward fusion (Figure 2). Blood tests revealed normal inflammatory indicators. Cervical immobilization was intermittently used for further 2 months. The patient recovered with a certain degree of reduction of cervical movements, no residual pain and no neurological deficit.

Discussion

Spondylodiscitis is an infectious process involving at least two vertebral bodies and the intervening

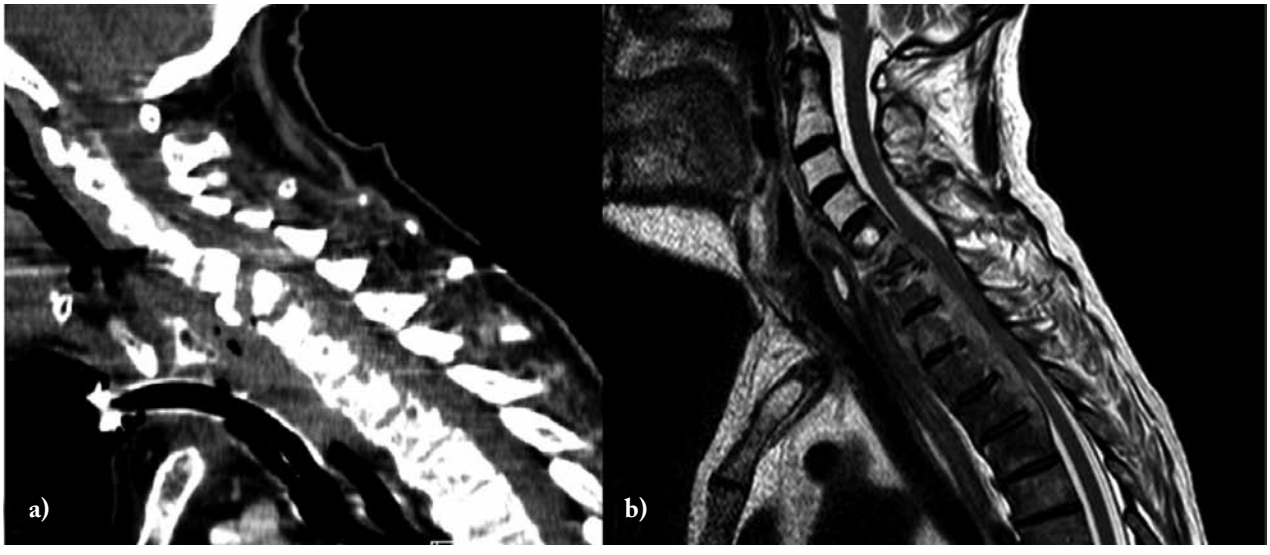

Figure 1. Sagittal CT (**a**) and T2-weighted MR imaging (**b**) showing a large, irregular and contrast enhanced mass wrapping the vertebral bodies from C5 to T4 with involvement of the intervening disks; diffuse demineralization of vertebral bodies involved; osteolytic lesion of C5; collapse with cuneiform deformation of C6; gas bubbles in the prevertebral space and anterior epidural space; slight compression of the dural sac at the level of T1

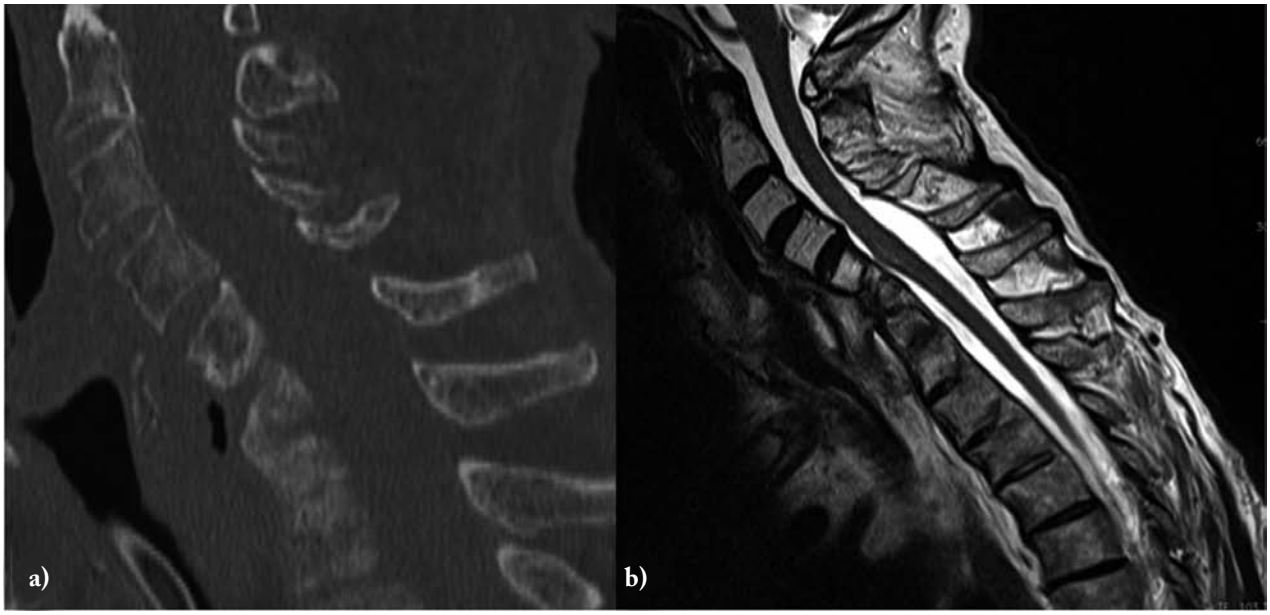

Figure 2. CT (bone algorithm) (**a**) showing vertebral fusion between C6 and C7 and sagittal T2-weighted MRI (**b**) showing nearly complete regression of the inflammatory mass

articular disk, usually caused by hematogenous spread of bacteria form a distant focus (3). Less frequently, spondylodiscitis may occur through a direct spread (4) of infection from a nearby focus after surgical procedures involving the head and neck, after removal of a foreign body from the hypopharynx or the esophagus, or after traumatic endoscopic procedures (5).

Although osteoradionecrosis represents a well described complication affecting bone tissue after radiotherapy for head and neck malignancies (6, 7), cervical spine involvement is extremely rare and scarcely reported in literature.

The relationship between osteoradionecrosis and spondylodiscitis remains unclear. While infection has been generally considered to be a minor secondary phenomenon, recent data suggest that spondylodiscitis may play a contributive role in the development of osteoradionecrosis.

Differential diagnosis of a cervical spine osteolytic lesion in a patient with history of head and neck cancer includes: locoregional tumour recurrence, bone metastasis, radiation-induced neoplasm and radiation-induced myelopathy.

Diagnosis of cervical spondylodiscitis complicated with spinal osteoradionecrosis is clinical, radiological and histological.

Clinical symptoms are often aspecific and treacherous as in the patient we described who complained only of a mild reduction of neck movements. The most common presenting symptom is neck pain but also headache and cervicobrachial pain may occur (8).

On plain films and CT (9), cervical spondylodiscitis and osteoradionecrosis appears as a zone of focal osteolysis involving the vertebral body cortical and cancellous bone. On MR imaging, it shows low signal intensity on T1 and contrast enhancement. The T2 signal intensity has been found to be variable. The case presented herein showed typical imaging features of vertebral osteoradionecrosis together with a florid soft tissue mass consistent with inflammatory collection. Involvement of the intervening vertebral disks suggests an infectious process whereas tumours more commonly spare the disks. PET has proven able to distinguish neoplasm from radiation-induced changes in approximately 85% of cases, where the latter show a weaker uptake of radionuclide. In our case, the lesion resulted hypermetabolic probably because of the intense inflammatory reaction associated.

Also when spondylodiscitis superimposes, most laboratory tests results are normal and only 50% of patients have fever (10). Anorexia, lethargy, weight loss and vomiting may occur (11).

To obtain a definite diagnosis of spondylodiscitis it is necessary to isolate a pathogen via blood culture or biopsy.

Percutaneous CT-guided biopsy has a high accuracy rate, although bacterial growth is obtained in only 6% to 17% of patients.

Conservative treatment of osteoradionecrosis and spondylodiscitis includes prolonged antibiotic therapy and cervical immobilization. Clodronate, corticosteroids, combination of pentoxifylline and tocopherol and hyperbaric oxygen have also been suggested. Neurological deficit, vertebral instability, progressive vertebral deformity and paravertebral abscess require surgical treatment. It consists in debridement of necrotic and infected tissue, decompression of neural structures and reestablishment of spinal alignment and stability by using a structural graft.

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Accepted: 19th April 2013
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