Cervical vertebral osteoradionecrosis: surgical management, complications and flap coverage—a case report and brief review of the literature

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Received: 29 May 2008 / Revised: 21 January 2009 / Accepted: 12 March 2009 / Published online: 2 April 2009
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Abstract A case description and a review of the literature. To report a case of deformity secondary to cervical vertebral osteoradionecrosis (ORN) associated with severe wound complications and review the pertinent medical literature. The incidence of deformity after ORN is rare and its association with extensive damage of soft tissues makes surgical treatment difficult. The spine surgeon should be aware of this to adapt the evaluation and surgery and be prepared to manage the numerous potential complications. A case of post-irradiation symptomatic kyphosis involving ORN of C5–C6 is reported. Failure of the anterior approach surgery was observed, and the secondary course was marked by the development of substantial cutaneous

necrosis associated with severe and extensive post-irradiation cutaneous and muscular atrophy. Failure of the anterior approach surgery justified the use of posterior stabilization. Secondary destabilization of the posterior fixation at the cervico-thoracic junction required extension of the osteosynthesis to the middle thoracic region. Extensive posterior stabilization permitted obtaining reliable mechanical control of the radio-induced kyphosis with a 3-year follow-up. Wound freshening and covering with well-vascularized tissue was used to fill dead spaces and helped prevent soft-tissue complications after revision surgery. Radio-induced kyphotic deformity is an important entity. Surgeons should be aware of the complications that can lead to further deformity. Corrective procedures are also at high risk for mechanical, atrophic and infectious complications. Surgical repair strategies should be based on thorough comprehension and work-up of the disorder.

Keywords Cervical vertebrae · Radionecrosis · Kyphosis · Surgical flaps

Introduction

Radiotherapy has become a primary arm in the management of cancers in children or adults, and in vascular malformations. Although the advantages have been well established, it is important not to forget the risks of radio-induced neoplasms and other known disadvantages including damage to soft tissue of the head and neck (cutaneous and muscular) and bone (osteoradionecrosis), which can be sources of mechanical complications (instability, deformity and neurological compromise) and infectious complications (soft tissue infections and osteomyelitis).

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Case report

We report the case of a 70-year-old patient, who was sent to our unit in January 2000 for post-irradiation symptomatic kyphosis involving osteoradionecrosis (ORN) of C5–C6. She had been treated in 1952, i.e., 48 years earlier for thyroid cancer, treated by surgery and radiation therapy. In 1994, she began to experience moderate isolated neck pain. The static and dynamic bending films then performed showed degenerative disc disease of C5–C6 with no sign of instability. In January 2000, the patient consulted for incapacitating cervicobrachial pain [World Health Organization (WHO) grade 2, Visual Analog Scale (VAS) at 6/10] without signs of myelopathy [Nurick grade 0; JOA score at 14/17 (2, 3, 2, 2, 2, 3)]. Plain films showed an exacerbation of the degenerative disc disease and loss of vertebral body height resulting in severe kyphosis (Fig. 1). The dynamic bending films revealed instability. A computed tomography (CT) guided biopsy was performed, which eliminated the hypothesis of a recurrent or radio-induced neoplasm. We concluded that the patient had necrosis and fibrous lesions compatible with post-irradiation fibrous scarring. She was operated for this in January

Fig. 1 X-rays made in 1994 and 1998 showing the exacerbation of degenerative disc and bone lesions leading to kyphosis and substantial offset of the facet joints in flexion

2000. Because of the soft tissue damage already present on the posterior side of the neck, an anterior approach was chosen (Fig. 2).

The operation consisted in C5 and C6 corpectomy through a right anterior cervicotomy. The vertebral bodies of C5 and C6 had become quite hard and there was substantial loss of the C5–C6 disc height. The C4–C5 and C6–C7 discs were removed and anterior cervical fusion was performed using autogenous iliac crest tricortical graft and rigid plate fixation correcting the preoperative cervical kyphosis. No primary or additional posterior approach was carried out because of the severe post-irradiation skin and soft-tissue atrophy on the posterior side of the neck.

Because of persistence and worsening of the painful cervicobrachial symptoms, magnetic resonance imaging (MRI) was requested 1 month postoperatively, showing lysis of the lower endplate of C3. The zone around the bone graft was enhanced by gadolinium and there was evidence of spinal epidural space inflammation. Surgical exploration through a standard anterior approach found infection of the arthrodesis with suppuration around the osteosynthesis material. Infected tissues were resected, screws and plate were changed, and the wound was abundantly washed. Specimens were positive for methicillin-resistant *Staphylococcus epidermidis* and those of the hardware swabbing for methicillin-resistant *Staphylococcus capitis*. A targeted antibiotic treatment (ofloxacin 200 mg three times daily and rifampin daily) was initiated and continued for 6 months. The patient wore a Philadelphia Minerva collar throughout this 6-month period (Fig. 3).

The secondary course was marked by the development of substantial cutaneous necrosis associated with severe post-irradiation cutaneous and muscular atrophy of the right lateral aspect of the neck, extending in depth and laterally beyond the posterior midline of the neck (Fig. 4).

In order to correct the residual kyphosis, surgical revision was performed through a posterior approach. The first stage was performed in April 2002 to cover the right lateral and posterior aspect of the neck with a musculocutaneous pedicle flap using the ipsilateral latissimus dorsi. Because of necrosis of the summit, the fragment did not cover the posterior median region. A pedicle flap from the lower posterior portion of the right trapezius was used to cover the posterior midline of the neck in November 2002. This coverage was successful. The use of the Minerva collar was still necessary (to keep her head upright). Cervical kyphosis remained severe and measured to -28° . The average physiological value of cervical lordosis has been estimated to be $40^\circ (\pm 9.7^\circ)$ [5]. The neck was bent anteriorly and towards the right-hand side.

The second stage of revision (stabilization by occipito-cervical fixation) was performed in March 2003. After installation in a Mayfield frame, and median posterior

Fig. 2 Photograph showing important extension of severe post-irradiation cutaneous and muscular atrophy already present during the first surgery

Fig. 3 Radiographic verification at the end of the antibiotic treatment showing the absence of fusion in C6–C7 and C4–C5, and the persistence of kyphosis

exposure from the back of the head to T1, a Euros frame was used for occipitocervical fixation. Sublaminar wires were used from C2 to C7, and the frame was secured to the occipital bone by four screws. Bone for grafting was harvested from the iliac crest and placed posterolaterally. A sternal occipital mandibular brace was worn for postoperative immobilization.

Postoperative sequelae included weakness in raising the arms corresponding to the displacement of the muscles used for the flap coverage. There was substantial muscular atrophy making it difficult for the patient to hold her head in the neutral position. Pain persisted, as well (WHO grade 2 and VAS at 4/10). Secondarily, the distal portion

of the occipitocervical frame came loose and protruded under the skin at C6 (Fig. 5). Because of this secondary destabilization of the posterior fixation at the cervico-thoracic junction and because a menace to the cutaneous integrity at the distal portion of the scar appeared eminent, revision surgery was decided to extend the osteosynthesis to the middle thoracic region.

Surgical revision was performed in October 2003. The posterior exposure was extended down to T7. Both the frame between C6 and C7 and the wires under C7 were broken. Titanium 220 rods were proximally linked by dominoes to the frame and attached from T1 to T6 with sublaminar wiring. Correction of the deformity in the coronal and sagittal planes was obtained. Substantial bone graft harvested from the thoracic posterior arches was applied at the cervico-thoracic junction.

There were no further postoperative complications. After 3 years of follow-up the good clinical and radiographic result has been maintained (Figs. 6, 7). The patient is no longer required to wear a cervical collar, despite persistence of marked atrophy of the neck muscles, especially on the right contralateral to the irradiated zone. This atrophy has prompted muscle strengthening sessions in a rehabilitation center.

Discussion

Osteoradionecrosis subsequent to radiotherapy for head and neck tumors is relatively rare. When it does occur, it is more typically located in the upper or lower jaw [7]. The

Fig. 4 **a** CT scan showing pseudarthrosis and subsequent kyphosis; **b** T2-weighted MRI showing severe posterior soft-tissue atrophy without damage of the epidural canal or spinal cord

Fig. 5 Subsequent destabilization at the caudal extremity of the frame

involvement of cervical vertebrae appears to be rare and the existing reports are succinct [2, 7, 11].

Criteria used to define ORN have changed over time [3, 8, 16]. Nonetheless, the description of three types proposed by Marx is still useful. Type 1 develops quickly after radiation therapy. Type 2 occurs within a few years after the radiation and secondary to progressive endarteritis and localized vascular thrombosis. The latter type of ORN always follows a traumatic event and occurs in most cases between 2 and 6 years after irradiation [3]. Type 3 occurs

spontaneously without traumatic triggering factors between 6 months and 3 years after radiation therapy. This type is associated with total doses of radiation >7,000 cGy or with doses >200 cGy per day, causing immediate radio-induced cell death. Time to onset of the latter two types of ORN is relatively long. As for most extra-osseous complications of radiation therapy, their development is dose-dependent (Table 1) [12].

The effect of the radiation on osteoblasts and osteoclasts associated with the vascular lesions leads to this loss of

Fig. 6 Extension of the posterior osteosynthesis to T7

cellular and metabolic balance, which notably results in osteolysis [7]. The often reported neurological symptoms may also involve several factors [14]. Osseous or ligamentous lesions [11], a potential source of deformity and instability can also lead to neurological decompensation caused by compression. In the present case, ORN was revealed by cervicobrachial pain with no accompanying myelopathy. At these levels of the cervical spine, the most frequent symptom appears to be pain [15].

On plain films and computed tomography, the ORN was visible as a zone of focal osteolysis showing involvement of the vertebral body cortical and cancellous bone extending to include the C5–C6 intervertebral disc. Dynamic bending films remain important to demonstrate instability caused by the lesion as in the present case. MRI confirms cancellous bone and disc changes, as well

as cortical destruction, which can mimic recurrence of a tumor or radio-induced neoplasms in some cases [12, 13].

Because of the lack of specificity of imaging studies in the differential diagnosis with a recurrent tumor, positron emission tomography (PET) scan can be useful. High-grade neoplasms, in particular, exhibit hyperactivity on PET scans, contrasting with the weak uptake of radio-induced lesions. The specificity of this examination in distinguishing the aetiology is approximately 85% [12]. If doubt persists, a CT-guided biopsy is indicated.

Kyphosis and instability in this patient were related to disc and vertebral body changes. The adjacent C5–C6 disc was involved and probably contributed to the painful symptoms as well as the focal kyphosis. Consequently, it was reasoned that an isolated anterior approach would permit us to address all the mechanical aspects of this patient's disorder with relatively little soft tissue sacrifice. However, in reference to a series [2], albeit small, that showed a strong tendency for infection and failure to consolidate if surgical management is employed, one should consider long isolated posterior or circumferential stabilization as the primary procedure to use in this type of patient. Although controversial, some physicians advocate associating hyperbaric oxygen therapy with surgery to stimulate tissue vascularization and wound healing [7, 9].

The most common change in superficial soft tissue in the acute phase is edema of ORN. Secondarily, fibrous scarring is typically seen along with muscle atrophy [4]. The risk of superficial and deep wound dehiscence in patients with ORN is high exposing patients to this complication if revision is necessary. Wound freshening and covering with well-vascularized tissue can be used to fill dead spaces and helps prevent this type of complication after revision surgery. The filling of a large zone of surface repair requires a reliable high quality pedicle flap of sufficient size.

The three muscular and musculocutaneous pedicle flaps most commonly used in this region are portions of the latissimus dorsi, the paraspinal muscles and the trapezius, by rotation-transposition, reverse-flap or free-flap coverage [6, 10]. The ascending pedicle flap of lower trapezius is the most frequently chosen among them and the most reliable for coverage of the upper thoracic and cervical spine [1]. Its anatomy is notably ideal to manage posterior cervical tissue gaps. Dissection of the trapezius and its transposition beyond the midline are straightforward providing reliable coverage of posterior spinal hardware. The pedicle to the lower portion of the trapezius is a branch of the trapezius artery, a descending branch of the transverse cervical artery. The possibilities of lower and superior rotation are considerable for adjustments in covering the region being reconstructed.

Fig. 7 Most recent clinical follow-up (2006), posterior and right lateral views

Table 1 Pathological changes: approximate periods of latency and doses of irradiation [16]

Radio-induced changes	Dose threshold (cGy)	Latency period (months)
Bone		
Fatty marrow replacement	800	>0.5
Osteoradionecrosis	6,000 (adults) 2,000(children)	12–36
Neck		
Superficial soft tissues	5,500	3–4
Deep soft tissues (pharynx, larynx)	6,000	2–6
Salivary glands	4,500	>7
Central nervous system		
Spinal cord focal necrosis	4,100	20
Diffuse white matter injury	2,000 (ctx+) 3,000 (ctx–)	3–120
Central nervous system atrophy	2,000	>6
Mineralizing microangiopathy	1,500 (ctx+) 2,000 (ctx–)	6–120
Telangiectasia	1,800–7,800	5–264
Large vessels vasculopathy	2,000	4–240
Neoplasms		
Meningioma	1,000–2,400	228–420
Osteosarcoma	1,660–11,500	36–396
Osteochondroma	1,660–6,425	96

ctx Chemotherapy

Conclusion

Radio-induced lesions can have severe consequences. Vertebral bony complications of these lesions require

surgical management with a high risk of mechanical and infectious complications. In addition to the importance of early detection of ORN, permitting one to avoid surgery in some cases [16], it is important to eliminate a possible

recurrent tumor or radio-induced cancer and to thoroughly consider surgical strategy, based upon understanding of this disorder.

Conflict of interest statement None of the authors has any potential conflict of interest.

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