Rhabdomyolysis after heterotopic ossification: an unusual complication in a spinal cord injured patient

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Abstract

Purpose Loss of joint mobility, resulting loss of function peripheral nerve entrapment and pressure sores are the known complications of heterotopic ossification.

Methods We reported about a 32-year-old male patient, referred to our clinic presenting with incomplete tetraplegia after a motorcycle accident. The patient developed a HO in both hips 19 days after injury. Single-dose radiation therapy with 7 Gy with an electrode voltage of 15 MeV was performed.

Results One month after radiation therapy, the patient developed a fever of 104°F and blood tests showed increased C-reactive protein (CRP), creatinine kinase (CK), serum myoglobin and dark-colored urine due to myoglobinuria. Further diagnostic measures showed only massive recurrent HO. With the suspicion of a possible case of rhabdomyolysis due to the severe muscular distension caused by the recurrent HO, the patient was transferred to our intermediate care unit for further treatment. Forced diureses were applied to reduce the high levels of CK in blood. However, laboratory monitoring

showed a further increase in CRP, CK (15,000 U/l) and myoglobin. A repeated radiation therapy was undertaken in a time interval of 14 days. After radiation therapy, a control MRI was performed and no signs of florid HO were visualized, CRP, CK, myoglobin, as well as body temperature and urine colour, had returned to normal and the patient was feeling well.

Conclusions We report the case of our patient to increase awareness among physicians and training staff working with patients with a spinal cord injury to expedite the diagnosis of rhabdomyolysis in the setting of HO and SCI.

Keywords Spinal cord injury · Heterotopic ossification · Radiation therapy · Rhabdomyolysis · Complication

Introduction

Heterotopic ossification (HO), also known as heterotopic bone formation, is the presence of bone in the soft tissue surrounding large proximal joints, where bone does not normally exist. HO was first described in 1883 by Reidel and, in 1918 Dejerne et al. [1] reported that HO frequently occurred among soldiers with spinal cord injury. Nowadays, it is known that spinal cord injured patients have a high risk of developing HO, with an incidence varying from 1 to 50%, and that it usually affects the large proximal joints [2].

Loss of joint mobility and resulting loss of function are the main complications of HO [3–5], while other complications like peripheral nerve entrapment or pressure sores can occur [6, 7]. In this context, we present an unusual case of rhabdomyolysis after HO in a tetraplegic patient. The patient was informed that data concerning the case would be submitted for publication, and he consented.

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Case report

We report about a 32-year-old male patient, referred to our clinic from a regional hospital where he had been admitted 4 days earlier, presenting with tetraplegia after a motor-cycle accident. He had suffered cervical spine fractures (C6 and C7) and also a right eighth rib fracture. Due to the cervical spine fractures, decompression of the spinal canal following by ventral fusion of the cervical spine with plate osteosynthesis and bone graft was performed in the referring hospital. Postoperative immobilization was achieved in a cervical collar for 12 weeks. After postoperative stabilization, the patient was referred to our hospital for further treatment. The clinical examination on admission showed a cardiopulmonary stable patient with an incomplete tetraplegia (ASIA grade B). No systemic illnesses, such as diabetes, cancer or peripheral artery occlusive disease (PAOD) were noted. The patient was a non-smoker. He was admitted to our spinal cord injury ward and was trained in early mobilization with the wheelchair by our specialized physiotherapists.

Routinely performed ultrasound (US) examination showed a suspicion for HO in both hips, 19 days after injury. This was confirmed by magnetic resonance imaging (MRI), which showed a highly florid bilateral HO of the soft tissues of the pelvic and thigh, left emphasized, with clear swelling of the M. obturator internus and participation of bilateral Mm. gemelli (Fig. 1). Furthermore, extensive edematous changes in the left M. gluteus medius with large dorsal ossification of the acetabulum were seen in the MRI. The benefits and risks of single-dose radiation therapy were explained to the patient. Three days later, after the patient consented, single-dose radiation therapy with 7 Gy with an electrode voltage of 15 MeV was performed in the radiation therapy clinic. After radiation therapy, no side-effects were noted and the patient was mobilized in the

wheelchair. US examination was repeated 2 weeks after radiation therapy and no signs of HO progression were identified.

However, 1 month after radiation therapy, the patient developed a fever of 104°F. Blood tests showed increased C-reactive protein (CRP) with 17.8 mg/dl, creatinine kinase (CK) with 5,021 U/l, elevated serum myoglobin with 1369 ng/l. Other parameters were within normal limits, including the procalcitonin level. Further diagnostic measures included urine analyses and blood cultures. Dark-colored urine due to myoglobinuria (303 ng/l) was also conspicuous. No signs of potential infection were noted at the time. Computed tomography (CT) of the thorax, abdomen and pelvic region was performed. The pelvic CT showed massive edematous swelling of the left gluteal muscle, obturator muscles, adductor group and quadriceps muscles, indicative of recurrent HO (Fig. 2). With the suspicion of rhabdomyolysis due to the severe muscular distension caused by the recurrent HO, the patient was transferred to our intermediate care unit for further treatment. Forced diureses were applied to reduce the high levels of CK in blood. However, laboratory monitoring showed a further increase in CRP (31.2 mg/dl), CK (15,000 U/l) and myoglobin. After discussions with the radiation therapists and the patient, we decided to undertake an additional round of radiation therapy. Seven sessions were performed in a time interval of 14 days. In each session, 0.5 Gy with an electrode voltage of 15 MeV was applied. After radiation therapy, a control MRI was performed and no signs of florid HO were visualized (Fig. 3); CRP, CK, myoglobin, as well as body temperature and urine colour had returned to normal and the patient was feeling well. The patient was discharged 3 months after admission in good overall condition. After 19 months of follow-up, the patient had no signs of infection or any further recurrence of the HO was reported. The patient had

Fig. 1 MRI images showing bilateral heterotopic ossification around the pelvic and thigh soft tissues

Fig. 2 CT of the pelvic region showing the recurrence of heterotopic ossification, with massive edematous swelling of the left thigh muscles, indicative of rhabdomyolysis

achieved good passive range of motion of the hip joint, without any restrictions.

Discussion

Spinal cord injured patients have a high risk of developing HO with an incidence varying from 1 to 50% [2]. The exact etiopathogenesis is still unknown, but several factors might influence the development of HO. For example, local micro-trauma, unbalance of calcitonin and parathyroid hormone levels, genetic predisposition, immobilization and venous stasis are all known to be the potential risk factors [2, 8–10]. Early detection of HO plays an important role in the treatment choice and the outcome, while ultrasound

screening examinations are the most cost-effective option, with overall good sensitivity [11, 12]. Therapeutic options include the use of nonsteroidal anti-inflammatory drugs (NSAIDs), such as indomethacin, the use of single radiation therapy, and, finally, surgical intervention if ankylosis has already occurred [13–22].

Eighty percent or more of HO cases run a relatively benign course without any severe complications, while the remaining 10–20% of cases can develop loss of motion, with ankylosis occurring in up to 10% [23]. Typically described symptoms of HO are pain (if sensation is intact), fever, swelling, erythema and decreased joint mobility, which were also present in our patient. However, a furthermore severe complication presented in this patient, with rhabdomyolysis developing after a massive recurrence of HO about the hip joint musculature. Clinical diagnosis of rhabdomyolysis was made due to the severe muscular distension observed in the MRI images, dark-colored urine due to myoglobinuria, highly elevated CK and myoglobin levels [24]. Although higher than normal levels of CK have been associated with HO [25], serum CK measurement over five times the normal limit has been traditionally used as the cutoff for diagnosis of rhabdomyolysis [26]. The patient required forced diuresis and seven sessions of radiation therapy to treat the recurrent HO. After treatment CK and myoglobin levels went down back to normal and MRI imaging was negative.

To this date, the only other case describing an association between HO and rhabdomyolysis was reported by Béthoux et al. [27] following a suicide attempt. However, the etiopathology was different in their case, as their patient developed heterotopic ossification in the left hip 2 months after rhabdomyolysis of the left upper and lower limb.

We report the case of our patient to increase awareness among physicians and training staff working with patients with a spinal cord injury in order to expedite the diagnosis of rhabdomyolysis in the setting of HO and spinal cord injury.

Fig. 3 Follow-up MRI of the pelvis before discharge without any signs of fluid HO

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

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