Expert's comment concerning Grand Rounds case entitled “Kümmell's disease: delayed post-traumatic osteonecrosis of the vertebral body” (by R. Ma, R. Chow, F. H. Shen)

Jean-Denis Laredo

Received: 21 October 2009 / Published online: 24 November 2009
© Springer-Verlag 2009

In the present issue, R. Ma et al. report a case of Kümmell disease in a 75-year-old woman who experienced a delayed T12 vertebral collapse, after a ground level fall [1]. Kümmell described, in 1891, a “postraumatic osteitis” characterized by a painful kyphosis developing several months or years following an injury, after a symptom-free period [2]. With the discovery of radiographs, it appeared that in Kümmell disease, radiographs taken early after the trauma should be normal, while delayed radiographs show a vertebral collapse. It then appeared in the numerous reports of this entity published in the last two decades that the above sequence of events was often associated with the radiographic appearance of a vertebral “vacuum phenomenon” characterized by the presence on radiographs of a

gas collection within a benign collapsed vertebra [3]. Because the vacuum phenomenon was attributed to ischemic osteonecrosis [3], it was, therefore, proposed by most subsequent authors that Kümmell disease was also secondary to an ischemic necrosis of the vertebral body and that the vascular injury was responsible for an impaired bone healing as mentioned by the authors of the Grand Round case published in the present issue.

Arguments that support the ischemic pathogenesis for the Kümmell disease include its association with the vacuum phenomenon, the documentation in one case of Kümmell disease of vascular occlusions by spinal arteriography [4], and the uncommon association of delayed vertebral collapse with conditions with vascular occlusions and osteonecrosis such as pancreatitis and Gaucher's disease [5].

Kümmell disease is an uncommon condition, which occurs in adults more than 50 and elderly, mostly women. The thoracolumbar junction, especially the T12 vertebra, is the most common affected vertebral segment [6]. To our knowledge, the disease is not commonly associated with conditions causing ischemic osteonecrosis such as sickle cell disease, alcoholism, longstanding steroid therapy and myeloproliferative disorders. It is not either commonly associated with typical avascular osteonecrosis of the femoral heads. Conversely, it often occurs in patients presenting chronic benign vertebral fractures, as illustrated by the case reported in the present issue by R. Ma et al.

These findings suggest that bone ischemia is probably not the primary event in Kümmell disease and in the vertebral vacuum phenomenon as well. Observations are more consistent with an osteoporotic collapse, secondarily complicated by impaired bone healing and osteonecrosis. Osteonecrosis is a non-specific finding that is found at the margins of all fractures with a variable extent. Osteonecrosis

J.-D. Laredo (✉)
Service de Radiologie ostéo-articulaire, Hôpital Lariboisière,
Assistance Publique des Hôpitaux de Paris et Faculté de
Médecine Diderot, Université Paris 7, Paris, France
e-mail: jean-denis.laredo@lrb.aphp.fr

may result not only from ischemia, but also from impaired mechanical conditions. For example, although femoral head osteonecrosis usually results from ischemia and is associated with diseases known to induce or favor the onset of vascular occlusion and bone ischemic injury, the so-called osteonecrosis-like syndrome of the femoral condyle at the knee is not associated with such conditions, but rather with overloading and manifests at MRI at an early stage as a subchondral fracture. Therefore, an osteoporotic fracture complicated by impaired healing and secondary osteonecrosis is a more likely explanation to both the Kümmell disease and the vertebral vacuum phenomenon [7]. In the Grand Rounds case report of Ma et al., the delayed post-traumatic vertebral fracture obviously belongs to a cascade of osteoporotic vertebral collapses [1]. Interestingly, Ma et al. cautiously called their case “delayed post-traumatic osteonecrosis” without adopting the term of “avascular osteonecrosis” quoted by most authors [1].

Another challenge raised by the Kümmell disease is the understanding of the sequence of events: trauma/ normal initial radiographs/persistent back pain/delayed vertebral collapse occurring after a time interval of several weeks or months. Osteonecrosis may account for this sequence of events, similarly to what happens in femoral head osteonecrosis: the development of osteonecrosis by itself is asymptomatic and symptoms appear only at the time of delayed bone collapse. However, putting the Kümmell disease in the group of the post-traumatic osteolyses occurring in the elderly, together with the post-traumatic osteolysis of the pubic bone [8] and the recently described osteolytic vertebral collapses simulating a malignancy [9], better explains the observed sequence of events. A missing link for this hypothesis would be the observation at MRI, in a Kümmell disease at an early stage, of a fracture line without collapse of the vertebral body. Finding of an undisplaced fracture line in a vertebral body adjacent to an acute osteoporotic collapse is not uncommon. Therefore, a pathophysiological hypothesis for the Kümmell disease is the following: after a trauma, an undisplaced fracture line involving a vertebral body (in a patient with a yet unidentified abnormal fracture reparation process) sets a

vicious circle with exceeding osteolysis and possible abnormal callus formation or mineralization. Although in a normal fracture reparation process, osteolysis is a very transitory step preliminary to callus formation, post-traumatic osteolyses may be characterized by durable osteolysis and abnormal coupling between osteolysis, and reparation occurring in an osteoporotic or senescent bone.

Finally, there is no mean to predict the end of the destructive process, which characterizes the Kümmell disease. Therefore, the indication of a vertebroplasty should be taken with caution in such cases, since the destructive process may further progress after the vertebroplasty, with a theoretical risk of secondary displacement of the cement.

References

1. Ma R, Chow R, Shen FH Kummell's disease: delayed post-traumatic osteonecrosis of the vertebral body. *Eur Spine J*. doi: [10.1007/s00586-009-1205-4](https://doi.org/10.1007/s00586-009-1205-4)
2. Kümmell H (1895) Die rarefizierende Ostitis der Wirbelkörper. *Deutsche Med* 21:180–181
3. Maldague BE, Noel HM, Malghem JJ (1978) The intravertebral vacuum cleft: a sign of ischemic vertebral collapse. *Radiology* 129:23–29
4. Stojanovic J, Kovac V (1981) Diagnosis of ischemic vertebral collapse using selective spinal angiography. *Rofo* 135:326–329
5. Resnick D (2002) Osteonecrosis: pathogenesis, diagnostic techniques, specific situations, and complications. In: Resnick D (ed) *Diagnosis of bone and joint disorders*, 4 edn. W.B. Saunders, Philadelphia, 3646–3650
6. Young WF, Brown D, Kendler A, Clements D (2002) Delayed post-traumatic osteonecrosis of a vertebral body (Kümmell's disease). *Acta Orthop Belg* 68(1):13–19
7. Laredo JD, Goumont V, Cywiner-Golenzer C, Bard M, Roucoules J, Ryckewaert A (1985) Vertebral compression with intrasomatic vacuum images: a proposal of 2 cases with histological control by trocar biopsy. *Rev Rhum Mal Osteoartic* 52(6):373–379
8. Goergen TG, Resnick D, Riley RR (1978) Post-traumatic abnormalities of the pubic bone simulating malignancy. *Radiology* 126:85–87
9. Sattari A, Quillard A, Laredo JD (2008) Benign nontraumatic osteolytic vertebral collapse simulating malignancy. *Eur Radiol* 18(3):631–638