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Posttraumatic vertebral body necrosis (Kümmell-Verneuil disease)

Case report of a rare orthopaedic disease with histopathological correlation

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Nécrose post-traumatique du corps vertébral (Kümmell-Verneuil). A propos d'un cas de cette affection rare, avec corrélation anatomopathologique

Résumé. Nous rapportons le cas d'une nécrose post-traumatique partielle et tardive, un an après un traumatisme rachidien. Le traitement chirurgical devient nécessaire en raison de l'installation d'une paraplégie débutante. Cette affection également connue sous le nom de maladie de Kümmell-Verneuil, peut se présenter comme une complication rare mais sérieuse pouvant survenir plusieurs mois ou années après un traumatisme du rachis. Radiologiquement la nécrose post-traumatique du corps vertébral se traduit par un vide intravertébral. Les raisons de cette nécrose proposées dans la littérature semblent être à l'origine, un microtraumatisme répété, un trouble métabolique ou une fracture tassement initialement négligée du corps vertébral. Histologiquement elle ressemble à une pseudarthrose avec une reconstruction osseuse très pauvre, comme dans l'ostéonécrose de la tête fémorale.

Mots-clés: Ostéonécrose – Fracture du rachis – Nécrose post-traumatique – Vide intravertébral

Summary. We report the case of a delayed partial posttraumatic vertebral body necrosis (PVBN) 1 year after a spinal trauma. Operative treatment became necessary due to incipient paraplegia. PVBN, also known as Kümmell-Verneuil's disease, can occur as a rare but serious complication several months or even years after a spinal trauma. Radiologically, it often presents with an intravertebral vacuum phenomenon. The underlying reasons for the necrosis as mentioned in the literature seem to be an initial microtrauma, metabolic disturbances and primarily overlooked compression fractures. Histologically, it resembles a pseudarthrosis with only little bony reparation, as in an avascular femoral head necrosis.

Key words: Bone necrosis – Spine fracture – Posttraumatic necrosis – Intravertebral vacuum sign

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The PVBN as a pathomorphologic entity was first described by H. Kümmell in 1891. He reported 6 cases of a relatively insignificant initial spinal trauma with a longer symptom-free interval followed by serious back pain, partly with neurological deficits. Clinically, a gibbus resulted which Kümmell himself related to a "rarefying process of the vertebral body" [19]. In 1892 Verneuil mentioned 3 similar cases of PVBN [28]. He was followed by Baker and Arbor [1], Cardis et al. [4] and Blaine [2]. In 1923 Jones summarized the work of more than 30 authors dealing with this subject. In most cases the vertebral bodies of the dorsolumbar area were seized up by the disease [16]. Recently, only a few descriptions of the phenomenon were published mentioning it as Kümmell's disease in the German and English literature and as Kümmell-Verneuil syndrome or delayed posttraumatic spondylitis in the French journals [3, 6, 7, 14, 18, 24].

Case report

A 60-year-old male patient sustained a compression fracture of T12 and L3 in 1987 (Fig. 1) that consolidated spontaneously without treatment. One year later he was referred to our department for acute back pain. No further trauma had taken place in the meantime. X-radiographs now additionally revealed a compression fracture of T11 with an intravertebral vacuum sign (Fig. 2). After conservative treatment with a high laced corset brace and balneophysical therapy, the pain diminished, but it reoccurred after 3 months accompanied by increasing neurological symptoms, first with a foot clonus and later on with a paraparesis predominantly of the right leg. Myelography showed a complete stop of contrast medium passage at T11 (Fig. 3).

The anamnesis of the patient contained three myocardial infarctions, a fracture of the ankle joint in 1984, a femoral neck fracture in 1986, and a depressive psychiatric disorder with a possible abuse of alcohol and tranquilizers. The levels of liver transaminases, however, were not significantly elevated.

In August 1990 the patient underwent anterior decompression with resection of the vertebral body of T11 and homologous replacement using a femoral head graft and anterior interbody fusion T10-L1 utilizing anterolateral USIS instrumentation (Fig. 4). The paraplegia subsequently subsided, and the patient is today able to walk again.

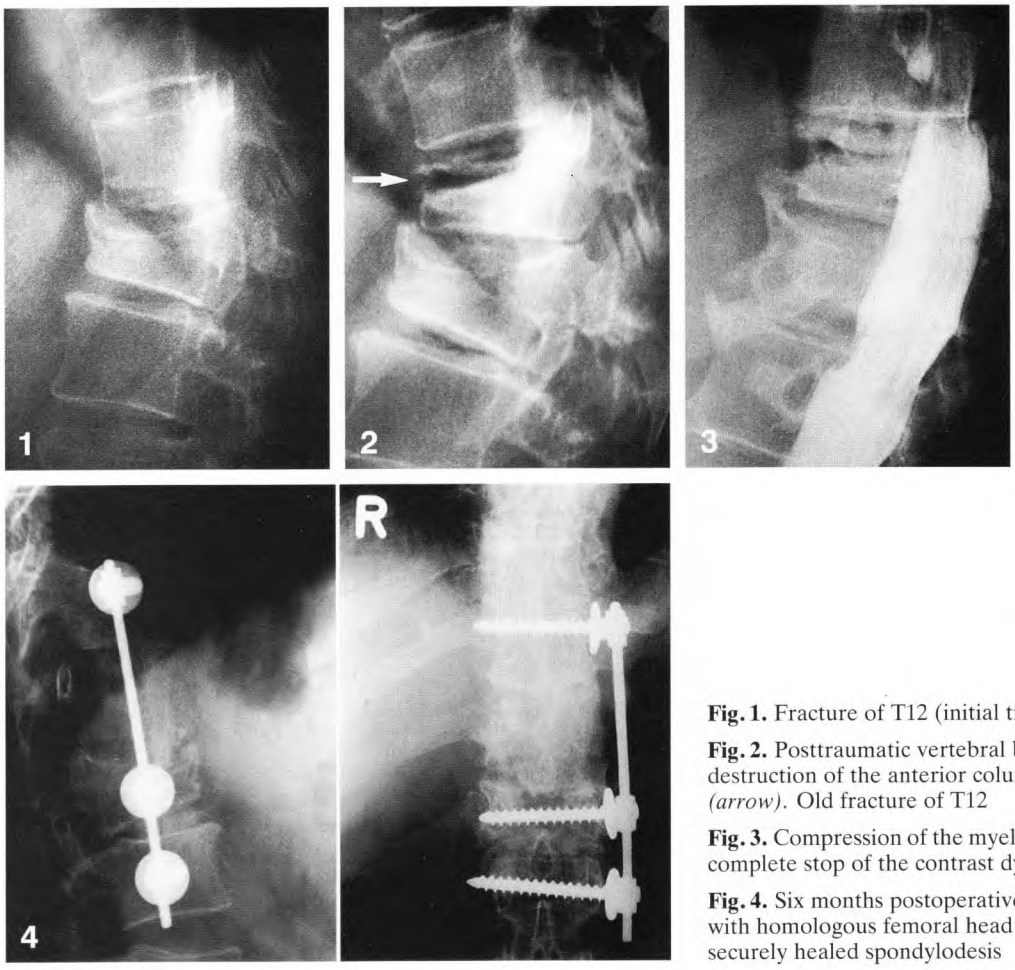


Fig. 1. Fracture of T12 (initial trauma)

Fig. 2. Posttraumatic vertebral body necrosis (PVBN) of T11 with destruction of the anterior column and intravertebral vacuum sign *(arrow)*. Old fracture of T12

Fig. 3. Compression of the myelon at the level of the necrosis with complete stop of the contrast dye passage

Fig. 4. Six months postoperatively, vertebral body replacement with homologous femoral head graft. Good bony ingrowth and securely healed spondylodesis

Histology

The resected vertebral body was embedded in polymethylmethacrylate (PMMA), prepared for undecalcified, thin-ground sections and stained with toluidine blue. The light microscopic examination of these sections allowed 6 zones to be delineated, as shown in Fig. 5.

In zones 1 and 6 parts of the aged intervertebral discs above and below with regressive and degenerative changes of the nucleus pulposus and the annulus fibrosus are visualized. The cartilage zone of the end-plates showed a normal cellular architecture. Its chondrocytes were normal in structure and number.

Zone 2 consists of the vital bone of the upper bony end-plate of T11. The cancellous trabeculae that normally originate from here seem to have broken off long before; the base stumps look rounded and abraded. Between these stumps lie condensed cell and tissue fragments as well as necrotic cancellous bone debris without vital cellular reaction.

Zone 3 represents the empty area corresponding to the radiological vacuum sign.

Zone 4 shows a variety of hyaline connective tissue areas, sections of active fibroblasts and cell clusters with chondroid metaplasia and metachromatic ground substance. Within this "zone of pseudarthrosis" numerous necrotic fragments of cancellous bone are embedded, seamed by osteoclasts and their lacunae of resorption. Sparse, new, woven bone indicates areas of bony reparation as would be expected to a greater degree in a normal vertebral fracture or a healing pseudarthrosis.

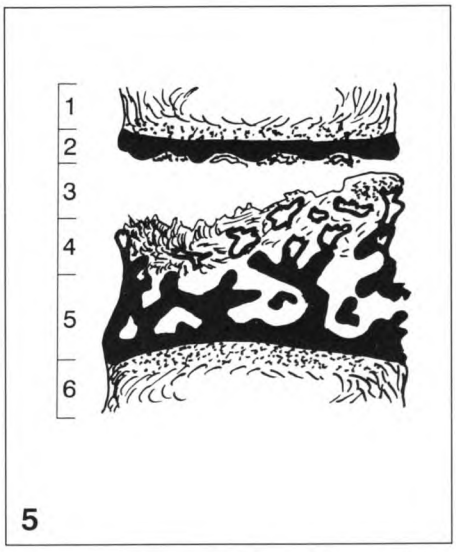
There is no distinct border to zone 5. Its trabeculae reach into zone 4 and are partly seamed by osteoclasts. Local necrobiosis is indicated by still empty osteocyte lacunae. The connective tissue of zone 4 merges into the normal fat and bone marrow of this vital part of the vertebral body remnant.

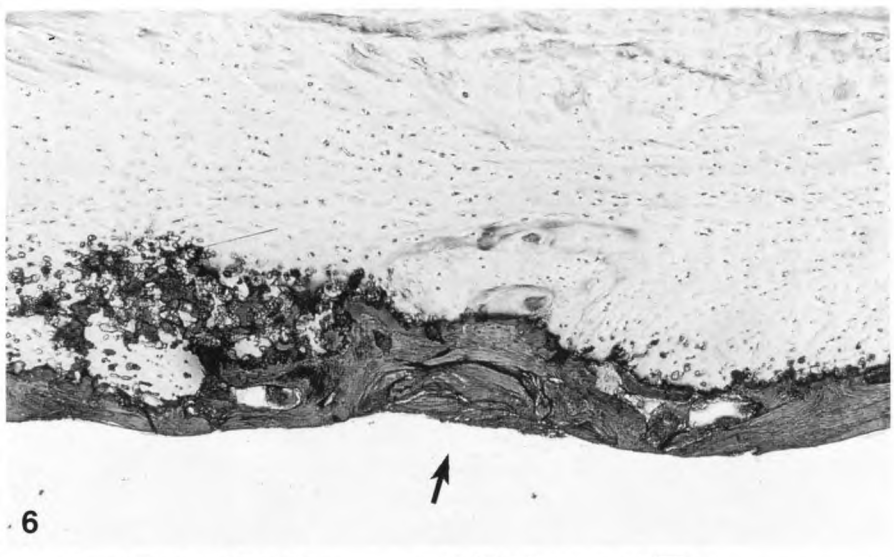
The summarized pathoanatomical diagnosis so far is partial necrosis of T11 with fissured collapse, osteoclastic resorption and intravertebral pseudarthrosis without significant bony regeneration. These changes are depicted in detail in Figs. 6–9.

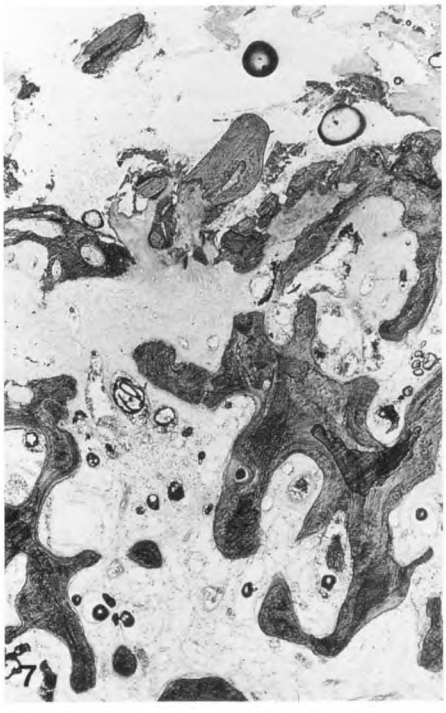
Discussion

The sequential course of PVBN was divided by Kümmell [19] into three stages:

1. The initial stadium with a bland trauma, sometimes not even remembered by the patient. Nevertheless, more severe accidents were mentioned, too, including fractures of adjacent vertebral bodies.







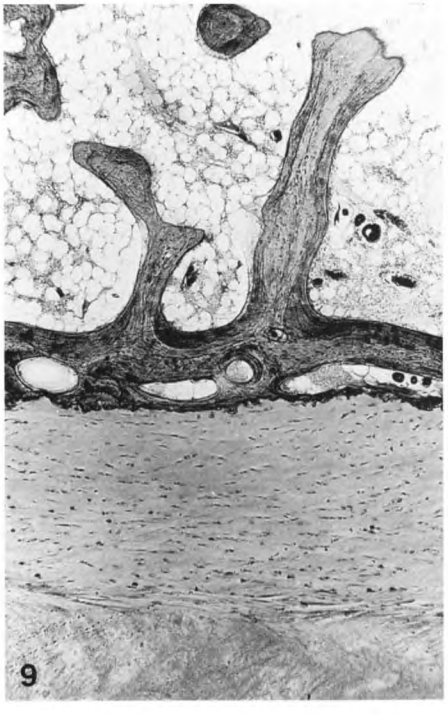


Fig. 5. Sagittal view of the vertebral body, schematic drawing analogous to the histologic sections in Figs. 6–9: (1) intervertebral disc T10/11; (2) vital upper end-plate of T11; (3) vertebral gap matching the radiological vacuum phenomenon; (4) broad necrotic zone with pseudarthrosis; (5) vital vertebral body remnant; (6) intervertebral disc T11/12

Fig. 6. Section of zones 1, 2 and 3. On *top* (zone 1), nucleus pulposus and fibrocartilaginous end-plate of the intervertebral disc T10/11. On the *left*, regressive metaplastic calcifications. In the *middle* (zone 2), vital remnants of the cranial end-plate of D11, superficially abraded. *Arrow* shows a hollow filled with trabecular debris. *Below*, empty area of the radiologic vacuum sign (zone 3).

Fig. 7. Overview of zones 4 and 5: pseudarthrosis in the upper part of the specimen merging with vital trabecular bone and normal bone marrow in the lower part of the specimen.

Fig. 8. Higher magnification of the entire pseudarthrosis (zone 4): numerous, necrotic, trabecular fragments in partly highly vascularized (*lower part* of the section), partly hyaline connective tissue, metaplastic calcifications, sparse desmoid bone proliferation *(Star)* and focal proliferation of chondrocyte clusters *(double stars)*

Fig. 9. Section of zone 6. On *top*, vital cancellous bone with normal bone marrow. In the *middle*, vital bony caudal end-plate of T11. On *bottom*, end-plate cartilage and nucleus pulposus of the intervertebral disc T11/12 without significant pathological changes

2. This painful stadium is followed by a symptom-free interval. It can last for several weeks or even a few years [1, 15].
3. In the third phase, all of a sudden and without further trauma, the back pain reoccurs and can be accompanied by paresis. On the X-radiographs a more or less pronounced gibbus and an intravertebral vacuum cleft can be diagnosed.

In the past different theories were discussed concerning the underlying cause of PVBN: Kümmell himself believed in an initial contusion of the vertebral body which

is not strong enough for a fracture but can provoke a disturbance of its nutrition, which again leads to a "softening and resorption or atrophy of the contact areas of the vertebral bodies" [19].

Verneuil [28] postulated multiple, intravertebral, trabecular microfractures without involvement of the cortical bone, later on causing a sintering and sinkage of the vertebral body.

Cardis et al. [4] discussed two different mechanisms. On the one hand, they assumed a local vascular disruption with haemorrhage and thrombosis, while on the other, they considered an initial nerve lesion or focal spinal cord trauma to be responsible for a lack of nutrition.

Recently, neurovegetative, vasomotoric disturbances and regional ischaemia due to a (micro)trauma as well as fatigue fractures were taken into account [3, 6, 27].

Besides the delayed posttraumatic form, there are also case reports of spontaneous, avascular, vertebral body necrosis. Almost all of these cases show a coincidence with long-term corticosteroid application, rheumatoid arthritis, diabetes mellitus, liver diseases and alcohol abuse [5, 8, 21, 23]. Additionally, they mainly concern elderly patients.

Both forms of vertebral necrosis have in common the intravertebral vacuum sign [17, 20, 22]. It must not be confused with the intradiscal vacuum phenomenon that can be observed in 1%–3% of all X-radiogoraphs of the vertebral column [25]. In the senium this rate increases up to 20%. The term "vacuum sign" is misleading: Ford et al. [9] measured a content of 90%–92% nitrogen in intradiscal defects. This can be explained by the reduced intradiscal pressure caused by extension and bulging of the internally disrupted disc. Thereby the solubility of nitrogen in the interstitial fluid is diminished, and the gas is set free ("soda water effect").

The intravertebral vacuum phenomenon is very seldom found and correlates with vertebral metastases [26], plasmocytoma [10] and aseptic vertebral body necrosis due to metabolic diseases [11, 21, 23]. Intravertebral gas accumulation was also proven by Brower and Downey in a case of PVBN [3].

Descriptions of histological findings with PVBN are rare and not very detailed [4, 27]. The results of our evaluation, analogous to the discussion in the literature, support the theory of trauma with subsequent malnutrition and intravertebral instability. The missing reparative process is particularly characteristic for PVBN [27].

One is inclined to compare PVBN with the spontaneous avascular (or aseptic) femoral head necrosis. Besides iatrogenic reasons for the latter, traumatic and metabolic mechanisms are also considered. The intact intervertebral disc cephalad to the pseudarthrotic vertebral body could be equivalent to the intact articular chondral layer of the femoral head. Both remain intact relatively long and are supplied and nourished from above. The still vital cranial end-plate may correspond to the bony "eggshell fracture" of the hip, caudally followed in both diseases by a gap with a possible vacuum sign. Below that area adjoins the necrosis literally speaking under which again the healthy remnant trabecular bone is situated.

In both diseases the vacuum phenomenon is related to a partial necrosis with intact neighbouring bone margins. In the vertebral body the necrosis leads step by step to a vertebral collapse, with hypermobility and "pseudarthrosis" of the necrotic area [20]. Only this instability can explain both the vacuum sign and the compression of the myelon. The therapy can only consist in the removal of the necrotic area and correction of the instability by anterior decompression of the myelon, vertebral replacement and spondylodesis in one single operation [12, 13]. Earlier therapeutic concepts are obsolete whenever the myelon is involved.

Our case report cannot definitely answer the question of whether PVBN is a genuine separate disease or not. We agree with the hypothesis of Schmorl and Junghans [27] that an initially overlooked vertebral compression without subsequent immobilization can lead to a partial necrosis of the fracture zone with consequent pseudarthrosis. We nevertheless know that such a necrosis can arise in spite of sufficient posttraumatic immobilization treatment, especially after vertebral plating with or without posterior fusion. The roentgen signs and pathohistological characteristics of PVBN closely resemble femoral head necrosis, which is less related to a direct trauma.

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