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Case Report

Pathologic fracture of the thoracic spine in a male master ultra-marathoner due to the combination of a vertebral hemangioma and osteopenia

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ABSTRACT

Vertebral hemangiomas are the most common benign vertebral neoplasms and are generally asymptomatic. In the present study, we report the case of a 52-year-old male master ultra-marathoner suffering from a pathologic fracture of the thoracic spine due to a vertebral hemangioma. A further examination in the athlete revealed an accompanying osteopenia, which was most likely due to a deficiency in both vitamin D and testosterone. The treatment of the fracture consisted of percutaneous vertebroplasty. Shortly after the operation the athlete was able to continue running. The most likely reason for the pathologic fracture of the vertebral body was the combination of the vertebral hemangioma and osteopenia. The further treatment consisted of supplementation of both vitamin D and testosterone. Athletes and physicians should be aware that male master ultra-marathoners older than 50 years might suffer from osteopenia, where a deficiency in vitamin D and testosterone could be contributing factors for osteopenia development in general.

1. Introduction

Primary intraosseous hemangiomas are most frequently seen in the vertebrae or in the skull [1,2]. Vertebral hemangiomas are the most common benign vertebral neoplasms and incidentally detected due to their characteristic features on imaging for other reasons. They usually occur in the lower thoracic and upper lumbar region.

We present the case of a 52-year-old male master ultra-marathoner who suffered from a pathologic fracture of his thoracic spine while running. Radiological imagining revealed

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an intraosseous hemangioma as the main reason for the fracture. The most likely reason for the fracture of the vertebral body with the hemangioma was an accompanying osteopenia most likely due to a combination of low vitamin D and low testosterone.

2. Case presentation

The 52-year-old highly experienced male ultra-marathoner went with his 64-year-old run comrade on a routine long training run. The two athletes had planned to complete their usual running distance of 40 km. Shortly after takeoff, the athlete under examination felt a sharp pain between his shoulder blades. He ignored the pain and thought it was a muscular strain after he had changed from the sidewalk across the street. Over the kilometers the pain became stronger and by the time he could hardly breathe and had to let his colleague running alone to the turning point (~20 km). On the way back both ran slowly together to the starting point. Back home, breathing became more severe and the pain increased steadily. From a recent operation he had available some painkillers and could sleep for a while after high-dose ingestion.

Over the next few weeks the pain between the shoulder blades remained and each step caused pain. While the pain remained for several weeks, the athlete visited his primary care physician who performed a radiograph of the thoracic spine showing a compression fracture in the mid thoracic spine (Fig. 1). For a more precise clarification, a magnetic resonance imaging (MRI) scan (3.0 Tesla, Achieva, Release 3.2.3.2; Philips Medical System, Best, the Netherlands) of the thoracic spine was performed using coronal, sagittal, and axial T1-weighted and T2-weighted sequences. Since the suspicion had been expressed on osteopenia in the X-ray of the spine, a DEXA (Dual-Energy X-ray Absorptiometry) followed the MRI.

In the MRI of the thoracic spine (Figs. 2–4), the vertebral body 5 showed a reduction in height in the anterior part with an increase in signal in the whole vertebra in all sequences with a loss of signal in the caudal part of the vertebra in the T1 and T2 sequences so that a relatively fresh fracture of this vertebra was diagnosed with an underlying vertebral hemangioma. Apart from vertebral body 5, further hemangiomas were found in vertebral bodies 6 and 10 of the thoracic spine and in vertebral body 1 of the lumbar spine.

In the DEXA, bone density showed a transition to osteopenia (*T*-score of −1.0 and lower) at the left wrist (*T*-score of −1.0), at the left femoral neck (*T*-score of −1.1) and at the lumbar spine (*T*-score of −1.6). A laboratory examination showed a decreased total testosterone (7 nmol/L, reference 9.5–30 nmol/L), a decreased free testosterone (20.8 pmol/L, reference range 22.9–104.1 pmol/L), an increased estradiol (181 pmol/L, reference 40–162 pmol/L), a decreased vitamin D (42 nmol/L, reference 50–100 nmol/L), an increased bone specific alkaline phosphatase (35.5 μg/L, reference 5.7–32.9 μg/L), and a decreased phosphate (0.67 mmol/L, reference 0.87–1.45 mmol/L). Sex-hormone binding globulin (31.7 nmol/L, reference 13.1–49.4 nmol/L), calcium (2.49 mmol/L, reference 2.1–2.6 mmol/L), and parathyroid hormone (2.0 pmol/L, reference 1.0–6.8 pmol/L) were within the reference range. Immunoassays analyzed the concentrations of sex hormones. Serum concentrations of testosterone and sex hormone-binding globulin were obtained using radioimmunoassay.

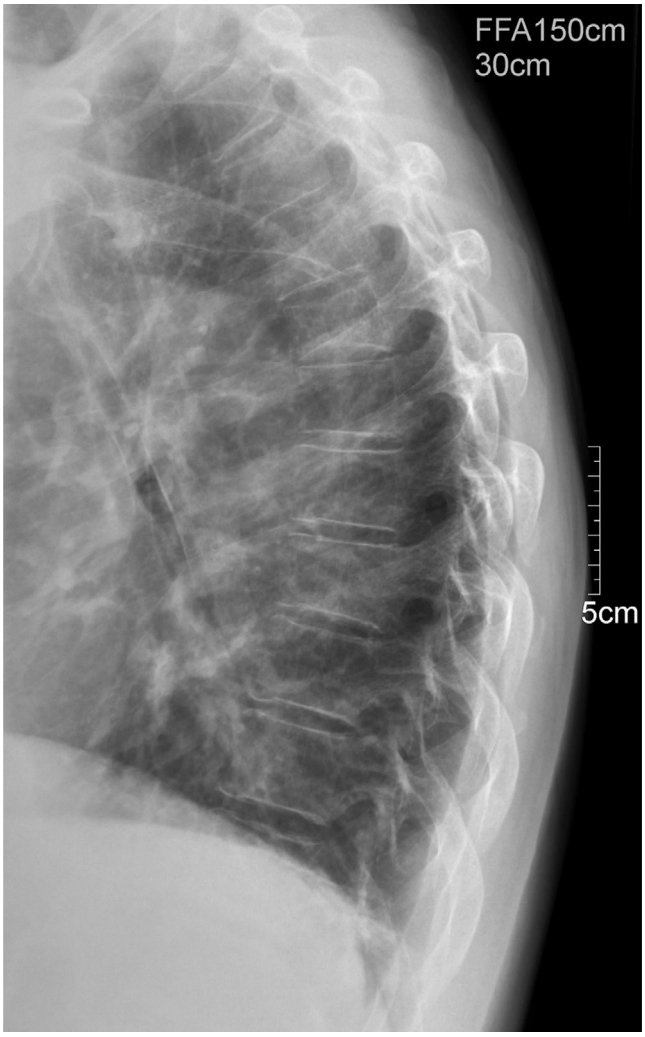

Fig. 1 – X-ray of the thoracic spine with compression fracture of vertebral body 5.

The fracture of the vertebral body was treated with vertebroplasty (Fig. 5) and the deficiency of testosterone and vitamin D was treated with parental supplementation of testosterone (Testosteronienantas, Testoviron®, 250 mg every 3 weeks) and vitamin D (Calciicarbonas et Cholecalciferolum, Calcimagon®-D_3/-Forte, 1 tablet per day with 1000 mg calcium and 800 I.U. cholecalciferol), respectively. A few days after the operation, the athlete started again his running training without pain or any other discomfort.

The family history of this athlete showed that his aunt (*i.e.*, sister of his father) and his great aunt (*i.e.*, sister of the mother of his father) suffered from severe osteoporosis with multiple fractures. His aunt had a fracture of the hip and the forearm, and his great aunt died of multiple fractures of the thoracic spine. An actual DEXA of the mother of the athlete showed also severe osteoporosis but without fractures.

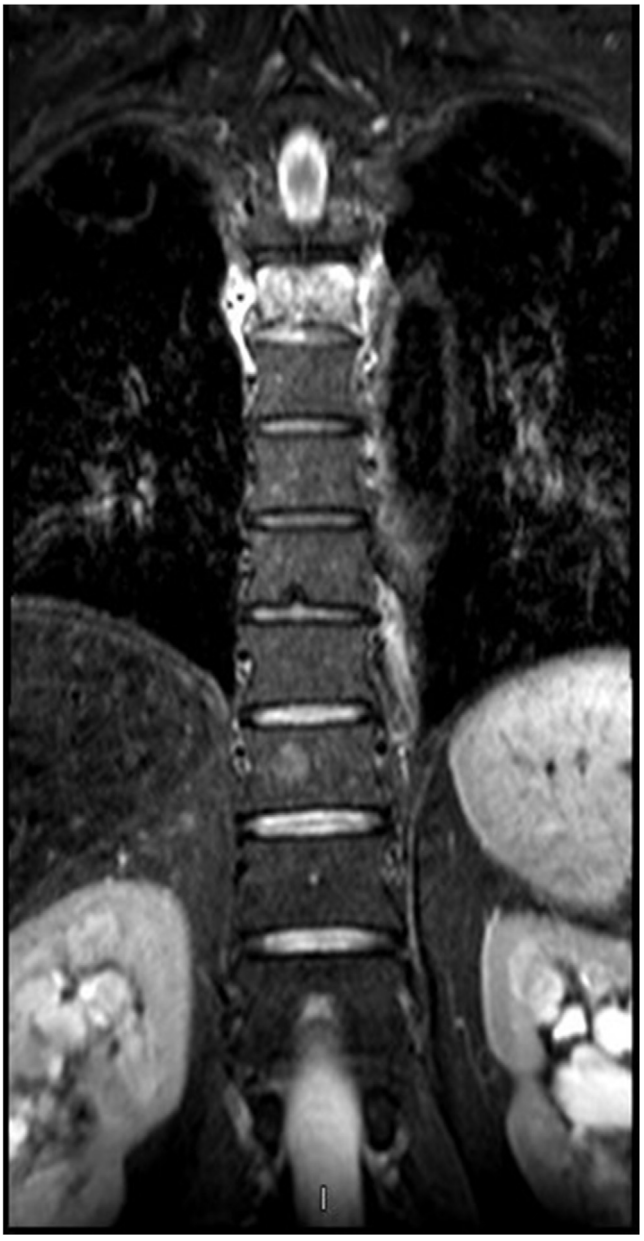

Fig. 2 – Coronal T2-weighted MRI of the thoracic spine.

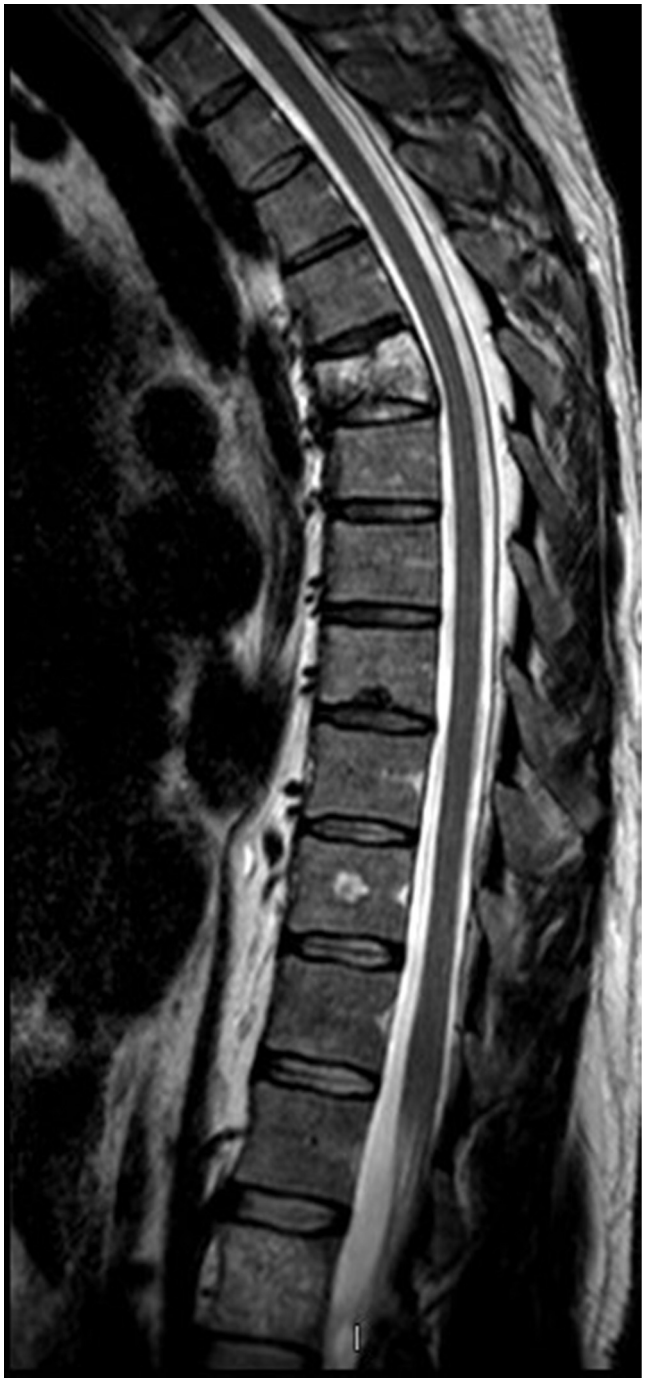

Fig. 3 – Sagittal T2-weighted MRI of the thoracic spine.

3. Discussion

The most likely reason for the pathologic fracture of the vertebral body in the athlete was the combination of the vertebral hemangioma and the osteopenia due to deficiency of both vitamin D and testosterone.

3.1. *Vertebral hemangiomas and potential symptoms*

The athlete suffered from a fracture of a vertebral hemangioma of the spine. The MRI of the spine showed four hemangiomas. Previous studies have shown that spinal hemangiomas are the most common benign spinal neoplasms, whereas vertebral hemangiomas are very frequent and often incidental findings on computed tomography and MRI of the spine [3,4]. The prevalence of vertebral hemangiomas is 26%, sex-independent, appearing in 28.6% of females and 23.5% of males, and age-dependent where the mean age of affected individuals (65.8 years) is higher than that of unaffected individuals (56.2 years) [4]. The size of vertebral hemangiomas is age-dependent. No vertebra seems significantly more prone to be affected by a hemangioma; however, T11 and T12 show the highest prevalence of vertebral hemangiomas (3.57% of vertebrae affected) [4]. Vertebral hemangiomas are found in similar percentages in the anterior

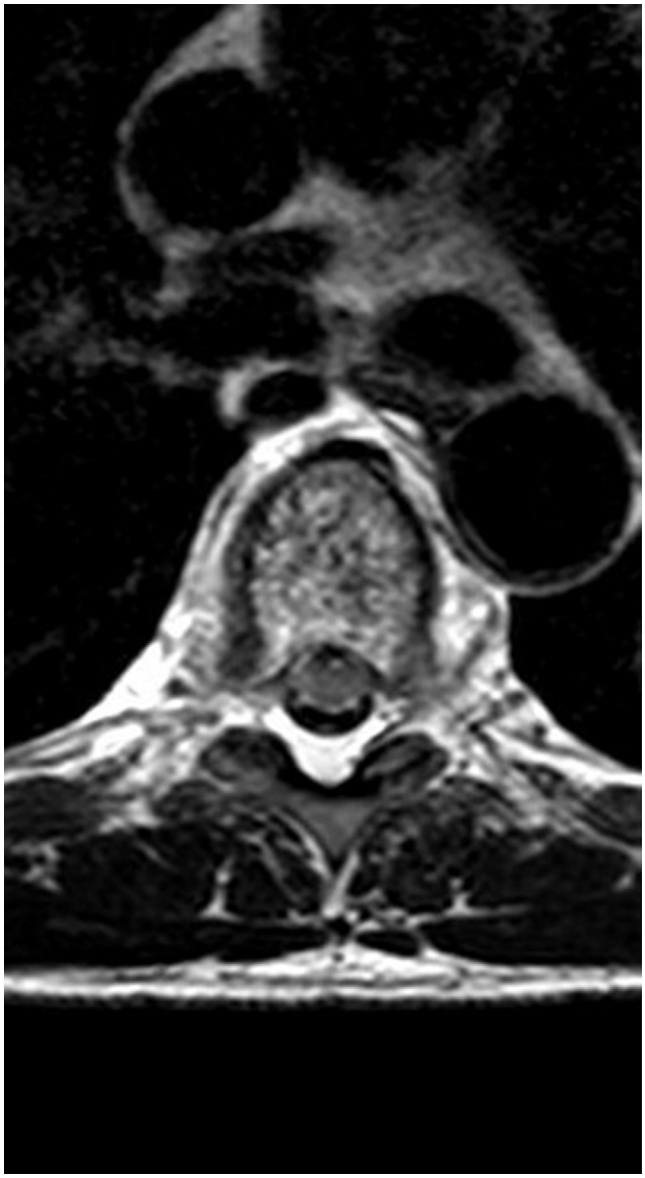

Fig. 4 – Transversal T2-weighted MRI of the thoracic spine.

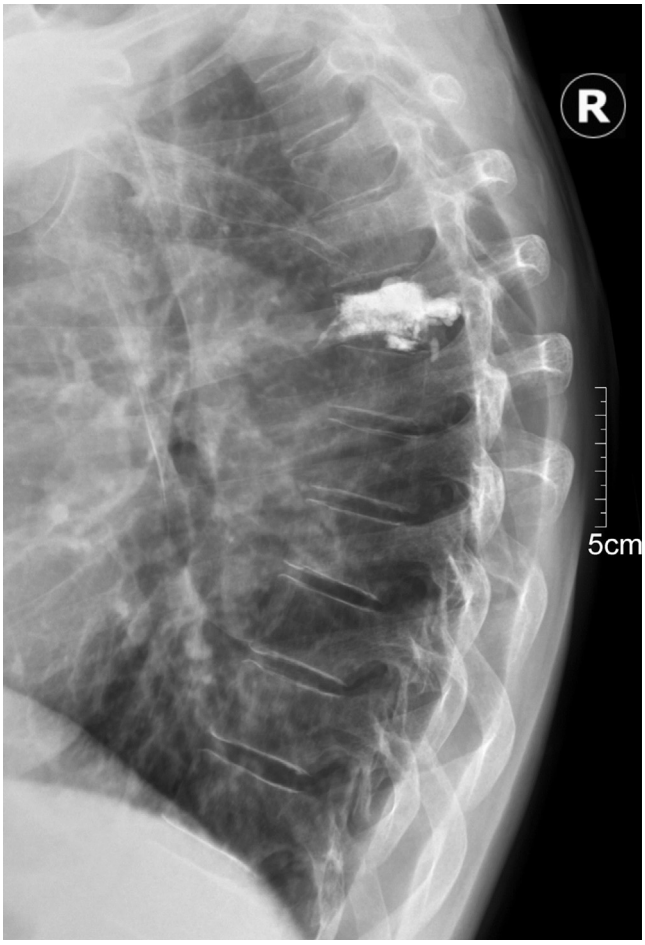

Fig. 5 – X-ray of the thoracic spine after vertebroplasty of vertebral body 5.

and posterior parts of the vertebral body, in its center and periphery [4]. Vertebral hemangiomas usually appear in the mid-height of the vertebral body or slightly higher [4].

Most hemangiomas of the spine are asymptomatic. Sixty percent of the spinal hemangiomas are asymptomatic without sex preference [5]. Symptomatic hemangiomas are more common in women and are accompanied with back pain, radicular pain, or spinal cord compression. Acute symptoms result from compression fracture, epidural extension and sudden mass effect, and bleeding. Common causes of pain due to a vertebral hemangioma could be a collapse of a vertebral body or encroachment into the neural canal [6]. An increase in physical activity can cause the vertebral hemangioma to become painful, such as starting to exercise or housework. This is most likely due to axial loading through the body of the vertebra. Symptomatic hemangiomas may cause vertebral compression fractures or vertebral collapses [7]. Generally, vertebral hemangiomas are not associated with osteopenia or osteoporosis [8,9] where only medium to large size vertebral hemangiomas are associated with osteoporosis [9].

In a symptomatic vertebral hemangioma, percutaneous cement vertebroplasty is an effective treatment technique, which is a valuable, minimally invasive and quick method that allows a complete and lasting resolution of painful vertebral symptoms [6,10]. Moreover, percutaneous vertebroplasty is the treatment of choice for osteoporotic fractures [11]. Therefore, in the present case, vertebroplasty was the treatment of choice for both the hemangioma and the osteoporotic fracture.

3.2. *Master athlete and osteopenia*

The athlete of the present case study was shortly after the age of 50 years. At that age, the prevalence of osteopenia and osteoporosis is increased not only in women, but also in men. For instance, in a sample of 346 elderly healthy men aged 50 years and above, 18.5% had osteoporosis, 55.5% had osteopenia and 26.0% had neither osteopenia nor osteoporosis [12].

Elderly men are at substantial risk for fractures, for which low bone mineral density is an important risk factor. Low bone mineral density is associated with an increased risk of

vertebral fractures in both men and women [13] and vertebral deformities in men are at least as common as in women suggesting that vertebral osteoporotic fractures are overlooked in men [14]. In middle-aged men, age, femoral and spine bone mineral density were significant predictors of the presence of a vertebral fracture [15]. The site of low bone mineral density seems to be important for the risk of an osteoporotic fracture in men. A study investigating bone mineral density in 402 men aged 45–92 years (mean 70 years) showed that low bone mineral density at the spine was associated with vertebral fracture [16]. Men with a vertebral fracture had lower bone mineral density than those men without fracture [17].

3.3. *Osteopenia and sex hormones*

Osteopenia in the present athlete might also be due to low testosterone due to his training. It has been shown that ultra-endurance performance leads to a suppression of testosterone [18,19]. The athlete competed since more than 20 years in endurance and ultra-endurance races and has a mean running training volume of about 330 kilometers per month, equal to more than 80 km per week. A study investigating high volume runners with 64–80 km running kilometer per week and very high volume runners running more than 95 km a week showed that long term distance running with training volumes less than 80 km a week had a positive effect on bone mineral density of the proximal femur. With running volumes greater than 64 km a week, training was inversely related to testosterone levels, but levels remained within the normal range [20]. A recent review summarized adverse effects of the extremes of physical loading as a function of duration of exposure [21]. After years of physical load the risk for bone demineralization and osteoporotic fractures is increased [21]. Endurance athletes are known to have decreased levels of sex hormones, which can cause physiologic changes leading to bone loss. This may result in relative osteoporosis despite the loading of the bone during exercise, which would normally increase bone mineral density. Premature osteoporosis may be irreversible, causing young athletes to become osteoporotic at an earlier age and have an increased risk of fracture later in life [22].

Testosterone and estradiol levels in this athlete were only marginally out of the reference range. These hormone levels were determined using immunoassays. However, this method is not the gold-standard [23] and most likely the values of testosterone and estradiol are not the real reason for osteopenia in this athlete.

Osteopenia can be found in long-distance runners but it is rather due to the high volume training in these athletes than to a reduction in testosterone [24]. In a study investigating runners older than 50 years with high volume training, no case of exercise-induced hypogonadotropic hypogonadism could be identified [25].

In this athlete, estradiol was marginally increased. Estradiol seems to be more important for bone mineral density in male athletes than testosterone. Indeed, an increased estradiol is rather protective for bone mineral density than the opposite. A study investigating the effect of testosterone and estradiol on bone mineral density in male athletes showed that free and total levels of estradiol, but not of testosterone, were important significant predictors of bone mineral density [26].

Sex hormone binding globin might be of more importance for premature fractures of vertebral bodies than testosterone [27,28]. It has been shown that a higher level of sex hormone binding globin, but not testosterone or estradiol, is an independent risk factor for vertebral fractures in older men [29–32]. In the present case, however, sex hormone binding globulin was within the reference but estradiol was elevated above the reference range. In men with idiopathic osteoporosis, estradiol may be decreased, but not increased [33,34].

3.4. *Osteopenia and age*

Also advanced age might be responsible for low testosterone and consecutive osteopenia in male athletes. In a study investigating 183 Caucasian male athletes older than 50 years, severe or mild testosterone deficiency was observed in 12% and 18%, respectively, of overall athletes, with the highest prevalence in athletes older than 70 years (27.5% and 25.0%, respectively). Interestingly, total testosterone did not correlate with training duration [35]. In 734 men aged 43–87 years, the prevalence of androgen deficiency was 24.1% based on the criterion of total testosterone level $<$300 ng/dL, and 16.6% based on the criterion of both total testosterone $<$300 ng/dL and free testosterone $<$5 ng/dL [36]. Low testosterone is not required for osteoporosis development leading to osteoporotic fractures in men. A study evaluating 38 men with primary osteoporosis and vertebral fractures showed that testosterone levels were similar between osteoporotic and control men [37].

A further aspect is the family history of this male runner. It has been reported that a parental history of fracture (*i.e.*, particularly a family history of hip fracture) confers an increased risk of fracture that is independent of bone mineral density [38].

4. Conclusions

In this athlete, the pathologic fracture of the thoracic spine is most likely due to the vertebral hemangioma but not due to the osteopenia which might be a contributing factor, but not the cause. A pathologic fracture of the spine due to osteopenia is very unlikely; the present case is most probably a very rare exception. Athletes and physicians should, however, be aware that master ultra-marathoners older than 50 years might suffer from osteopenia. For athletes and coaches, master ultra-marathoners older than 50 years and training and competing since decades should undergo determination of bone mineral density, testosterone and vitamin D in order to exclude osteopenia and/or osteoporosis and to establish a treatment, if necessary, to continue training and competing without a pathologic fracture. This is especially recommended when the athlete has a positive family history for osteopenia and/or osteoporosis.

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

B.K. and P.N. drafted the manuscript, T.R. revised the manuscript critically for important intellectual content, B.L. performed MRI, and C.B. carried out the operation. All the authors read and approved the final manuscript.

Consent to publish

Consent to publish has been obtained from the participant to report individual data.

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