CASE REPORT

Osteonecrosis of the femoral head following an electrical injury to the leg

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We report a case of osteonecrosis of the femoral head in a young man who is a carrier of the prothrombin gene mutation. We suggest that an electrical injury to his lower limb may have triggered intravascular thrombosis as a result of this mutation with subsequent osteonecrosis of the femoral head. No case of osteonecrosis of the femoral head secondary to a distant electrical injury has previously been reported.

Abnormalities of coagulation may be associated with osteonecrosis of the femoral head.\(^1,2\) We present a patient who was a carrier of the prothrombin gene mutation who developed osteonecrosis after being electrocuted.

Case report

A 36-year-old Caucasian man was working on a wet building site when his right lower leg and foot made contact with an exposed (live) 500 V electrical cable through his wet footwear. He sustained an electrocution with muscular contractions for approximately 30 seconds, but was subsequently able to pull himself away from the cable, thereby breaking the circuit. He did not seek medical advice.

Approximately 18 months later he presented with increasing pain in the right groin. He denied any trauma to the right hip. He drinks a moderate amount of alcohol and has never taken steroids.

Examination revealed shortening of the right leg of 2 cm. The range of movement in the right hip was restricted and painful. A full blood count, including mean corpuscular volume, erythrocyte sedimentation rate, C-reactive protein, liver function tests (bilirubin, alanine transaminase), bone profile (calcium, phosphate, alkaline phosphatase) lipid profile (cholesterol, triglycerides) and coagulation screen (prothrombin time, partial thromboplastin time and fibrinogen) were all within normal limits.

Radiographs of the hip showed advanced degenerative changes of the hip joint with collapse of the femoral head. A provisional diagnosis of osteonecrosis of the femoral head was made (Fig. 1).

A further detailed medical history focusing on drugs and diseases associated with osteonecrosis of the femoral head was negative.\(^3,4\) A haematological evaluation was carried out aimed at identifying any of the hypercoagulable disorders that have most recently become implicated with osteonecrosis of the femoral head.\(^5\) Tests for protein C and S deficiency, Factor V Leiden, activated protein C resistance, antithrombin III deficiency, hyperhomocystinaemia, antiphospholipid antibodies (IgG and IgM), lupus anticoagulant, lipoprotein (a), tissue plasminogen activator antigen levels, plasminogen activator inhibitor-1 antigen levels and plasminogen activator inhibitor activity were all found to be within their normal ranges. However, he tested positive (heterozygous) for the presence of the prothrombin gene mutation.

He underwent total hip replacement (THR) and the femoral head was submitted for histological examination, which showed features consistent with avascular necrosis (Fig. 2).\(^6,7\) More than five years after the injury the patient’s THR was functioning well and the opposite hip remained clinically and radiologically normal.

Discussion

Non-traumatic osteonecrosis of the femoral head is quite common; between 15 000 and 20 000 new cases are diagnosed annually in the United States, and this contributes to approximately 10% of the indications for primary THR. Young adults between 20 and 45 years of age are most frequently affected, and the condition is bilateral in approximately 50% of patients at presentation.\(^5\)
Despite the fact that electrical injuries are common, and that there is a wealth of information in relation to electrical soft-tissue injuries, there is a paucity of literature describing the bony changes. This may reflect the fact that electrical currents of low, commercially-available voltages are rarely the cause of bone pathology. The consequences of electrical injuries to bone may present immediately or after a delay of months to years; in addition, the bony injuries may exist near the entry point, or at a point distant from it.

Most reports to date relate to high-voltage injuries (i.e. currents over 1000 V).8-11 These currents take a direct path between entry and exit point. Blood vessels and nerves are severely damaged as are muscle and skin damage, resulting in amputation in over 50% of cases.8-11 The radiological features of the skeleton following this type of injury are varied, but in most cases are non-specific and can be interpreted only with knowledge of the injury (e.g. osteonecrosis, osteoporosis, growth disturbances). Only osteoschisis (longitudinal diaphyseal zigzag fractures) and ‘bone pearls’ are thought to be pathognomonic.12,13

Low-voltage currents (< 1000 V) behave differently and follow the path of least resistance along nerve and blood vessels. Bone itself is a poor conductor and does not carry a large enough current to sustain direct damage. Osteonecrosis observed at a site distant to the entry or exit point is most likely attributed to damage of the vascular wall followed by thrombosis and in ischaemia.14 Williams and Karl15 reported a case of a 16-month-old girl who sustained an isolated injury to the terminal ileum following low-voltage electrocution. It appears that the current passed down the superior mesenteric artery and dissipated in its terminal branches, resulting in infarction of a segment of bowel. A similar pathological process may have affected our patient, damaging the blood supply to the femoral head. This blood supply is known to be precarious because of the terminal nature of the subsynovial branches of the medial femoral circumflex artery, as shown by the high incidence of pathological processes secondary to vascular disturbances observed in this part of the skeleton.16 Indeed, this vulnerable blood supply may be even more marginal if anomalies of the femoral head circulation are present. In a study of the vascular anatomy of 99 femoral heads undergoing vascularised fibular grafting for osteonecrosis, 94% had an abnormal vascular pattern, in contrast with only 13% in a control group. Absence or hypoplasia of the superior capsular artery were the most common abnormalities.17

Govoni et al18 described a 52-year-old woman who received a 220 V electrical shock to the right hand, and who was subsequently shown to have osteonecrosis of the ipsilateral humeral head. The author questioned whether, despite a negative history of previous shoulder complaints, the electrical injury had not just merely drawn attention to a pre-existing osteonecrosis of the humeral head.

The paucity of reports and studies linking electrical injuries to osteonecrosis suggests that not only anatomical factors determine the outcome of this type of injury. A concept unifying traumatic and non-traumatic osteonecrosis has recently been proposed. Osteonecrosis appears to be the final common pathway of ischaemia; more specifically in the case of non-traumatic osteonecrosis, poorly-regulated coagulation, either genetic or acquired in origin, is activated by a variety of diseases or events resulting in thrombosis and ischaemia.19 The scientific literature has recently identified an increasing number of hereditary and acquired coagulation abnormalities with thrombotic potential. These studies increasingly demonstrate an association between the thrombophilia (an increased likelihood of

Fig. 1
Anteroposterior radiograph of the patient at presentation.

Fig. 2
Unstained sections of the femoral head, showing fibrocartilage on the articular surface with areas of granulation tissue replacing bone marrow. Sclerotic bone with foci of new bone formation were seen.
The prothrombin gene mutation (G20210A) that affected our patient was first described by Poort et al. and colleagues in 1996. They noted that 18% of patients with venous thrombosis and approximately 1% of a comparable group of healthy volunteers had a G to A mutation at nucleotide position 20210 of the prothrombin gene. This mutation increases prothrombin levels; heterozygous carriers were found to have approximately 30% higher prothrombin levels, and presumably this is the mechanism through which it exerts its effect. The prevalence of this mutation among healthy individuals varies from 0.7% to 4% among Caucasians; it is rare among Africans and Asians. Although well recognised as a hypercoagulable syndrome, the prothrombin gene was only recently demonstrated to be a risk factor for osteonecrosis of the femoral head in adults by Björkman et al. Zalavras et al also found a considerably higher incidence of this mutation in patients with osteonecrosis compared with controls, but attributed the lack of statistical significance to the insufficient power of their study. To our knowledge, the prevalence of osteonecrosis of the femoral head among carriers of the prothrombin gene mutation is unknown.

The risk factor for venous thrombosis for carriers of this mutation is relatively low, and most carriers will not experience a thrombotic event by the age of 50 years. More significantly this mutation requires an additional environmental event for the clinical expression of its thrombotic potential. Surgery, trauma, pregnancy and oral contraceptives are among the most commonly recognised ‘triggers’. Most coagulation defects have a fairly high prevalence in the general population, and it is, therefore, unlikely that the presence of one or several of these defects alone is sufficient to trigger thrombosis and subsequent osteonecrosis. It is currently thought that, through these coagulation defects, a predisposition exists in most patients with osteonecrosis which could be acquired (e.g. antiphospholipid antibodies) or genetic in nature (e.g. the prothrombin mutation). These subclinical coagulation defects could result in clinical disease when challenged by environmental factors, the so-called ‘second hit’ (e.g. trauma, alcoholism, steroids). Indeed, a recent study demonstrated that over 80% of patients with non-traumatic osteonecrosis of the femoral head had at least one coagulation abnormality detected on screening. The author did not find any significant difference in the prevalence of these coagulation abnormalities among the major causative groups recognised so far (steroids, alcohol, trauma, mixed). This suggests that osteonecrosis will occur with a given frequency in the presence of a thrombotic coagulation abnormality once a second hit is present, regardless of the latter’s nature. With this in mind, we believe that a low-voltage current could qualify as such a second hit, and selectively and indirectly damage the blood supply to the femoral head. It is probably self-evident that this essentially vascular process would express itself most obviously in an area where the microcirculation is most vulnerable, such as in the femoral head.

There is a strong causative link between alcohol consumption and osteonecrosis of the femoral head. Although self-declared levels alcohol use in a labourer may be somewhat suspicious, there was no clinical or biochemical evidence of chronic heavy alcohol use in this patient. Despite the emergence of newer, albeit not yet fully-established, techniques for screening and monitoring alcohol use/abuse, testing with mean corpuscular volume and alanine transaminase remains in widespread use because of their favourable test characteristics. Chronic alcohol abuse (defined as a consumption of more than 60 g per day over a period of two weeks) would cause the mean corpuscular volume to be elevated for several months, despite the individual ceasing drinking. Elevated alanine transaminase levels are a non-specific indicator of liver disease. When otherwise healthy people drink large amounts of alcohol, alanine transaminase levels in the blood increase. Both mean corpuscular volume and alanine transaminase results were within normal limits in our patient. ‘Excessive’ alcohol intake remains difficult to define, particularly in relation to osteonecrosis. One prospective study suggested that a weekly intake of more than 400 g of alcohol (approximately 60 g per day) would increase the relative risk of osteonecrosis tenfold. This level of exposure to alcohol in our patient was firmly refuted by his normal biomarkers and he was not ‘at risk’ in relation to his alcohol use.

The clinical fact that this commonly bilateral disease remains unilateral in this patient after more than five years suggests that a unilateral second hit occurred. If a systemic condition is to affect both hips, it will usually become apparent in the second hip within two years of diagnosis in the first. Bradway and Morrey studied a group of 15 patients who had undergone unilateral THR for non-traumatic ischaemic necrosis of the femoral head triggered by alcohol abuse, steroid use or idiopathic factors. The contralateral ‘silent’ hip was asymptomatic and radiologically normal at the time of entry into the study, and was at no time subjected to invasive investigations or prophylactic treatment. The authors observed that all the hips studied progressed rapidly to collapse within five and a half years, with over three-quarters collapsing within three years.

The time delay of 18 months between the electrical injury and the clinical presentation of osteonecrosis is in keeping with our suggested association: when there is a known and isolated predisposing event, the condition may be identified as early as three months and as late as five years following that event.

We believe a relatively low-voltage electrical current was the cause of the osteonecrosis in our patient. Our patient was genetically primed for osteonecrosis by a thrombotic coagulation abnormality which under normal conditions would probably have remained without effect during his lifetime. However, the low-voltage current probably sufficed to cause enough local damage to over-
whelm the abnormal clotting cascade, resulting in ischaemia and osteonecrosis. Whether this electrical current damaged the arterial or venous vasculature remains conjectural.

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References