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Case Study

Cervical vertebral erosion caused by bilateral vertebral artery tortuosity, predisposing to spinal, sprain: A medieval case study

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ABSTRACT

Bone resorption within the cervical spine due to vertebral arterial tortuosities is rarely observed in medical practice because the condition often lacks clinical symptoms. Traumatic complications involving the vertebral arteries are relatively common and occasionally very serious, but very few affect bone, appearing only when survival has been sufficiently long for a pseudoaneurysm to form. CT scans and MRI screening, practised increasingly today following traffic and sports accidents, incidentally show that arterial tortuosities that had stimulated bone resorption are relatively frequent. Only rarely do such tortuosities cause nerve compression or trigger orthopaedic problems, while large pseudoaneurysms and congenital absence of a vertebral pedicle may require surgery to stabilize the spine. There are few publications by palaeopathologists reporting such conditions of the cervical vertebrae. This contribution reports a case of a tiered bilateral tortuosity of the vertebral artery dating from the Early Middle Ages; it provides a basis by which to recognize this type of lesion in osteoarchaeology, and it attests to the fact that multiple tortuosities may lead to spinal instability in the form of spine sprain.

1. Introduction

Vascular grooves and impressions on bone are observed at many skeletal locations, including the sulcus of the vertebral artery on the atlas and the aortic impression on the left side of the body of the fourth thoracic vertebra (Paturet, 1958). Arterial imprint on bone linked to an anomaly or pathology is infrequent, however. This contribution presents a unique case of bilateral multi-level bone resorption of the cervical vertebrae due to vertebral arterial tortuosities, with orthopaedic complications. Dating to the Early Middle Ages, this case provides an opportunity to review vascular diseases with associated bone resorption and to define the aetiology and complications of such lesions to inform a differential diagnosis.

1.1. Pathological data

In modern medical practice, arterial pathology is most frequently encountered in the lumbar region in cases of 'chronic contained rupture of aortic abdominal aneurysms' – a concept introduced by Jones et al. (1986). These are observed in only 2.7% cases of ruptured aneurysms, but they are accompanied by bone damage in 25% of their cases (Ando et al., 2003). Bony changes are seldom observed in cases of ruptured anastomotic aneurysms (Diekerhof et al., 2002; Mestres et al., 2006). Certain other forms of bone resorption of aortic origin are more rarely seen today, such as those due to syphilitic (Sheeran and Sclafani, 2000; Bodhey et al., 2005) or tubercular (Takahashi et al., 2007) thoracic aneurysms, or aneurysmal dilatation of the left atrium (Ashworth and Morgan Jones, 1946), vascular damage due to neurofibromatosis type I (Peyre et al., 2007), Behçet's disease (Roeyen et al., 1997; Barros et al., 2004), or collateral lumbar vein dilation after thrombosis of the inferior vena cava (Singson et al., 1984).

The other common site is the cervical spine. The vertebral artery is classically divided into four segments (Hong et al., 2008). The first segment (V1) starts from its origin on the subclavian artery to the C6 transverse process, the second (V2) from C6 to C2 transverse process, the third (V3) from C2 to the foramen magnum, and the fourth (V4) from the foramen magnum dura to the vertebrobasilar junction. Transverse foramina provide bony protection for the vertebral artery. Conversely, old vascular lesions in V2, due to closed trauma or wounds may trigger pulsating blood flow with a sufficient survival period for bone resorption to occur, as in pseudoaneurysms (Schittek, 1999; Inamasu and Guiot, 2006), which are sometimes associated with arteriovenous fistulas (Madoz et al., 2006). The first post-traumatic case with bone erosion was reported by Anand et al. (1993).

Extracranial atheromatous vertebral aneurysms are very rare, as are spontaneous arteriovenous fistulas, both of which infrequently produce bone lesions (Rifkinson-Mann et al., 1986; Shin et al., 1996). One case of diffuse arterial dysplasia of unspecified

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nature presented in a 22-year-old patient with several aneurysms of the cervical blood vessels, the left vertebral artery measuring 1.5 cm in the C5–C6 intervertebral foramen, produced by concentric widening (Schimmel et al., 1976). Neurofibromatosis type I can produce bone resorption in vertebral arterial aneurysms (Peyre et al., 2007), caused by alteration of the normal process of vascular maintenance and repair regulated by neurofibromin produced by the NF1 gene.

Angiographic tests currently practised more frequently in preoperative investigations (Peng et al., 2009) and for trauma of the cervical spine due to traffic and skiing accidents (Biffl et al., 2000, 2002; Cothren et al., 2003, 2006) reveal the occasional presence of tortuosities of the vertebral arteries; these are asymptomatic but are sometimes accompanied by bone resorption. In a study of 222 vertebral columns from the Hamann–Todd skeleton collection, Curylo et al. (2000) found a 2.7% prevalence of vertebral artery tortuosities, especially at the C3 and C4 level. Tortuosity is assumed when the distance between the medial margin of the uncovertebral joint and the medial border of the tranverse foramen is less than 1.5 mm. From their study of 500 vertebral arteries by magnetic resonance imaging or computed tomographic scans, Bruneau et al. (2006) identified two categories of medial loops for the segment of arteries between C6 and C2 (segment V2). In 2%, the vertebral artery formed a medial loop, either with a transverse foramen abnormally widened medially at the expense of the uncinate process (1.2%), or in the proximal part of the intervertebral foramen (0.8%). Lateral loops may occur in cases of osteoarthritis (George and Cornelius, 2001). Bilateral forms of bone resorption have been reported on the same vertebra (Babin and Haller, 1974; Hyyppä et al., 1974; Holden et al., 1996) and in a number of separate vertebrae (Royo Salvador et al., 1981).

Connective tissue disorders do not seem to be involved in this form of bone resorption, perhaps due to early death, although they are accompanied by large tortuosities of the vertebral arteries, as in Marfan syndrome and especially Loeys–Dietz syndrome. The latter which is due to a mutation of the TGFBR (transforming growth factor-β receptor) presents a 90% rate of tortuosities in the vertebral arteries (Kono et al., 2010).

1.2. Pathophysiology

The pathophysiology of vascular erosions highlights the preponderant role of pulsatile movements transmitted to bone. This accepted cause of aortic pathologies (Blakemore, 1947; Katz et al., 1962), is sometimes accompanied by debilitating bone ischaemia (Husson et al., 1996), as well as in repeated episodes of infection that occur in posterior ruptured aneurysms (Galessiere et al., 1994).

With regard to the cervical spine, it is well established that the embryological formation of transverse foramina is influenced by vertebral vessels (Taitz et al., 1978; Bruneau et al., 2006; Hong et al., 2008). In the limited space of the transverse foramen between C6 and C2 (segment V2 of the vertebral artery), there is very clear contiguity and adherence between the artery and the periosteum of the vertebral body, particularly near the uncovertebral joint (Kehr, 1974; Argenson et al., 1979). In these conditions, the bone is clearly exposed to blood pulsations if the vascular wall is injured, arteriovenous fistula is formed, or abnormal curvature of the artery concentrates flow on a limited area of the bone. Initial injury to the vertebral artery may be linked to wounds or to arterial stretching (Nibu et al., 1997; Lisanti and Hartness, 2009), especially in cases of unilateral dislocation of the spine or when there is fracture of the foramen transversarium (Mueller et al., 2011). Loop formation remains unclear; it has been attributed to congenital factors (Babin and Haller, 1974), arterial hypertension (Danziger and Bloch, 1975) or atheroma (Hadley, 1958; Zimmerman and Farrell, 1970). It has also been assumed that arterial loops could result from relative elongation of the vertebral artery following reduction of intervertebral disc space (Sakaida et al., 2001). Similarly, loop size could be correlated with the extent of cervical osteoarthritis (Oga et al., 1996). A study of 64 cadavers revealed that 13.4% of vertebral artery tortuosities occurred in the intervertebral space, especially C3 to C4, and that tortuosity significantly correlated with disc degeneration (Nourbakhsh et al., 2010). A peculiar case was reported by Danziger and Bloch (1975) wherein due to an anomaly in the insertion of the left vertebral artery on the aortic arch near an aortic coarctation, lesions at several levels of the cervical column were observed, similar to the costal resorption near the osteochondral junction typical of this condition.

1.3. Palaeopathology

Resorptive defects of arterial origin are rarely observed in bones from anatomical collections or archaeological samples. Most references concern the aorta (Ortner, 2003; Kelley, 1979; Walker, 1983; Waldron, 1993; Brothwell and Browne, 1994), sometimes the aneurismal popliteal artery (Wakely and Smith, 1998).

Equally exceptional are references to vertebral arterial bony defects in the published literature. An initial observation was reported by Henry Arthur Waldron (Vaswani and Waldron, 1997), considered to be of aneurysmal origin. Tony Waldron and Antoine (2002) saw it as the consequence of a tortuosity, perhaps hereditary since their case is from the same site as the previous observation. They presented two cases that they believe to be linked, one to a tortuosity, and the other to an aneurysm. After reviewing the literature, they suggested diagnostic criteria to differentiate these two aetiologies. Resorptive lesions have the same appearance, with a layer of compact bone on the bottom and sharp sclerotic edges. They often involve the pedicle where resorption may lead to widening of the intervertebral foramen or the transverse foramen. The first difference is the site where they develop, C4 and on the left-hand side for the tortuosity, at a higher level than is the case for aneurysms. Tortuosities are more common after age 50 and may be present at several levels, sometimes bilaterally. But most important, an aneurysm is larger than an arterial loop and can affect several vertebrae and posterior elements of the transverse foramen on the anterior surface of the lamina.

Recently, Billard and Fantino (2011) reported an archaeological case of a woman over 60 years old from Haute-Savoie (France), dating to the 5–6th centuries. They observed left lateral posterior resorption of the C3–C4–C5 bodies and the pedicle of C3. The authors attribute these lesions to arterial loops (megadolichoartery).

Appreciably different is the skeleton described by Knüsel and Bowman (1996), affected by severe lesions ascribed to neurofibromatosis. The right transverse and spinous processes of C3–C7 have been subject to an osteolytic process, most likely due to an altered vascularization.

2. Materials and methods

The fragment was excavated from a site on the future runway of the Metz–Nancy–Lorraine Regional Airport in eastern France. In the hamlet of Larry (commune of Liéhon, Moselle), a Merovingian cemetery was identified near a large agricultural courtyard of a Gallo-Roman villa, in ruins since the 5th century AD. The rescue excavation was undertaken by INRAP (Institut national de recherches archéologiques preventives) in 2003. Direct observation of lesions and plain radiographs (X-rays) were employed to explore this pathology.