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CASE REPORT

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Bilateral absence of the pars interarticularis of C2: developmental or post-traumatic abnormality?

Abstract A 19-year-old male was diagnosed with bilateral absence of C2 pars interarticularis incidentally after a motor vehicle accident. Plain radiography, cross-sectional CT, and 3D CT findings of this case are presented. The differential diagnosis and possible etiologies including remote child abuse are discussed in detail.

Key words Hangman's fracture – Pars interarticularis – Trauma – Cervical spine – Child abuse – Computed tomography

Case report

A 19-year-old male presented with neck pain to the emergency room after a motor vehicle accident. The neurological examination upon arrival was normal. The patient's medical history was noncontributory, although he stated that he had had mild cervical pain in the last few years. The initial lateral radiographs of the cervical spine (not shown) revealed mild anterior offset of C2 over C3 and possible fractures involving the C2 pars interarticularis bilaterally. Computed tomography of C1 through C4 with additional 3D reconstructions was obtained to verify bilateral C2 pars interarticularis fractures and to determine whether there were any additional post-traumatic abnormalities. This showed bilateral absence of the C2 pars interarticularis without evidence of acute fracture or significant soft tissue abnormalities (Figs. 1, 2). Subsequently, lateral plain-film views of the cervical spine in flexion (Fig. 3) and extension were obtained to determine whether there was instability of C2, which more clearly demonstrated the absence of the pars interarticularis of C2 without significant instability. Although the diagnostic consideration was a developmental absence of C2 pars interarticularis, the patient was admitted to the hospital for observation; he was discharged home the following day.

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Discussion

Absence of a pars interarticularis in the cervical spine, either unilateral or bilateral, is an unusual entity and may cause a considerable diagnostic dilemma when encountered in the setting of acute trauma. It is a complex of abnormalities in posterior arch development with potentially confusing clinical and radiological manifestations and may easily lead to a mistaken diagnosis of acute fracture or dislocation.

There are three phases in vertebral development: the membranous or blastema stage, chondrification, and ossification. After the membranous stage, six centers of chondrification appear on each vertebral blastema: two for the vertebral bodies; two for the pars interarticularis, lateral masses, and dorsal transverse processes; and two for the lamina and spinous processes. The embryologic basis for congenital absence of a pars interarticularis is lack of development of the vertebral chondrification center of the posterior arch which would form the pars interarticularis, ventral one-half of the lateral mass, and the dorsal part of the transverse process [1, 2, 3, 4].

The typical radiologic triad of absence of a pars interarticularis includes the false appearance of an enlarged neural foramen, dorsally displaced dysplastic ipsilateral articular pillar and lamina, and a dysplastic ipsilateral transverse process. The majority of cases reported in the literature describe unilateral absence of the pars interarticularis, most commonly seen at the C6 level and next most often at the C5 level. The most common clinical manifestation is cervical pain, often presenting after recent trauma. The differential diagnosis for enlarge-

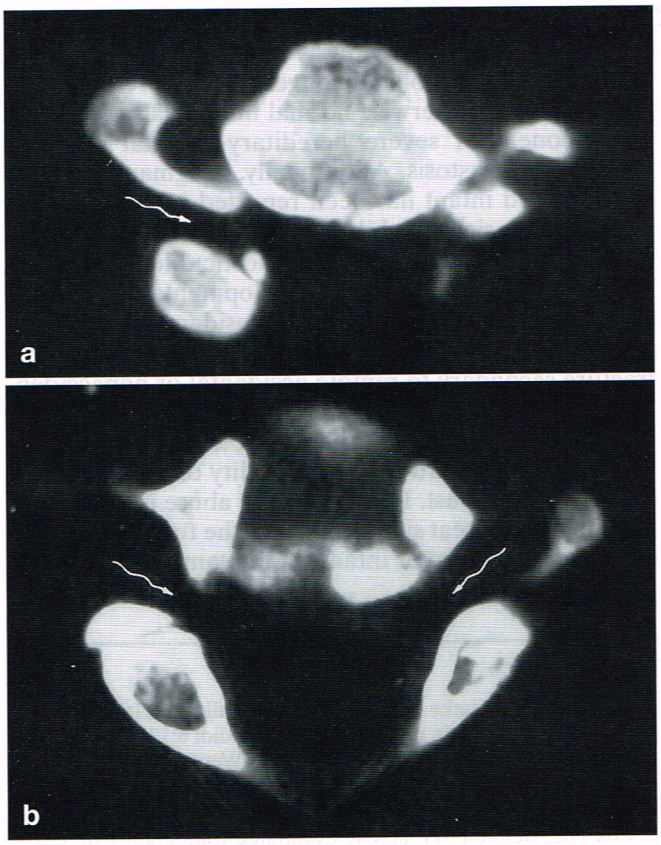

Fig. 1 Two contiguous axial CT images show bilateral absence of the C2 pedicles *(arrows)* with smooth sclerotic margins, suggestive of congenital absence of the pedicles or sequela of remote trauma (accidental or nonaccidental). No prevertebral soft tissue swelling or sharp, irregular bony edges were noted to suggest an acute fracture

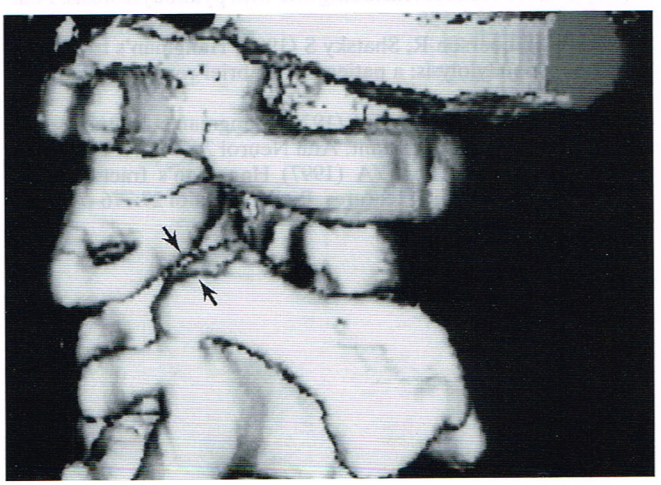

Fig. 2 3-D reformatted CT image of the upper cervical spine (3.0 mm slice thickness with 0.5 mm spacing) shows a bony defect between the articular surfaces of the pars interarticularis and the posterior arch of the C2 vertebra *(arrows)*. In addition, the apophyseal joint between C2 and C3 appears to be fused

Fig. 3 Lateral radiograph of the cervical spine in flexion shows a typical congenital absence of the pars interarticularis of C2: a defect involving the posterior elements of C2 with anterior displacement of C2 body relative to C3. In acute trauma cases, this appearance may be confused with that of an acute hangman's fracture. The osseous defect involving the C2 pedicles is obvious *(arrow)*. The apophyseal joint between C2 and C3 appears to be fused. Extension brought no significant change in this appearance (not shown). No significant prevertebral soft tissue swelling is present

ment of the vertebral foramen should include: (1) spinal tumors, especially dumb-bell tumors – neurofibroma, meningioma, ganglioneuroma, chondroma, dermoid, teratoma, fibroma, metastasis, and plasmacytoma; (2) bone tumors – osteoblastoma, osteofibroma, osteochondroma, aneurysmal bone cyst, giant osteoid osteoma, and osteogenic sarcoma; (3) pathology of the vertebral artery – tortuosity, aneurysm, and angioma; (4) spondylitic lesions; (5) fractures; and (6) meningocele [5].

When the appearance of a hangman's fracture (fracture of the posterior elements of C2 with anterior displacement of the C2 body relative to C3) is noted in a patient with recent trauma, an acute fracture may not be easily differentiated from primary spondylolysis (due to congenital absence of a pedicle) or a sequela of remote fracture (accidental or nonaccidental) on a radiographic study alone. However, one should always search for sharp edges and cortical disruption on a plain

radiograph, which clearly indicate an acute injury. CT in acute fracture usually demonstrates sharp, irregular fracture margins with adjacent soft tissue swelling, whereas it shows smooth sclerosis of the margins of bony defects in primary spondylolysis or in cases of sequelae of remote fracture. The margins of an acute Hangman's fracture may, however, be quite smooth with little sclerosis or proliferative change, and thus an acute traumatic basis should be carefully excluded in all cases of C2 spondylolysis. Differentiating an acquired spondylolysis from a congenital abnormality (primary spondylolysis) may also be difficult if the abnormality is present in an otherwise normal individual. This differentiation is usually much easier when primary spondylolysis is related to pyknodysostosis or Crouzon disease (craniofacial synostosis) [6, 7], both of which are hereditary diseases that have other striking clinical and skeletal findings, e.g., characteristic facies or mental retardation. There are only two case reports of a hangman's fracture secondary to violent shaking in babies [8, 9], both of which were diagnosed in infancy and had other findings of the shaken baby syndrome. Our case, by contrast, is in a 19-year-old teenager who presented with neck pain and came to medical attention following a motor vehicle accident. The plain radiographs as well as axial and 3D reconstruction CT images demonstrated the findings of the congenital or developmental absence of C2 pars interarticularis, although no other significant associated skeletal abnormalities suggesting this etiology were present. The lateral radiographs obtained in flexion and extension demonstrated a very mild anterior offset of C2 over C3 with no significant instability, which probably accounts for the patient's being relatively asymptomatic. Keeping the possibility of chronic hangman's fracture in mind, further questioning of the patient revealed neglect in his childhood by his mother who had left the family a few years previously, but he did not have any memory of severe trauma or hospitalization. No pertinent hospital records were available either.

In conclusion, absence of pars interarticularis is a rare, complex developmental abnormality, which is most commonly seen in the lower cervical spine (C6, C5) and may lead to misdiagnosis in the acute trauma setting. Primary spondylolysis of C2 is also described, which may be an isolated congenital or developmental abnormality in otherwise normal individuals or may be a component of severe hereditary skeletal diseases, e.g., pyknodysostosis. Additionally, hangman's fracture in an abused infant has been reported in two cases, in both of other signs of child abuse were noted. Although the presented case raised a diagnostic dilemma on the basis of the facts discussed, developmental absence of the C2 pars interarticularis was considered foremost in the differential diagnosis. A stable chronic hangman's fracture secondary to remote accidental or nonaccidental trauma, however, should also be considered in the differential and these patients should be questioned carefully with regard to the possibility of nonaccidental trauma in their past. If remote child abuse is diagnosed, this may be of great significance for the future wellbeing of younger children or infants in the same family.

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