Dynamic CT scan of the craniovertebral junction: a role in the management of os odontoideum

Rola dynamicznej tomografii komputerowej złącza szczytowo-potylicznego przy ustalaniu postępowania w przypadkach stwierdzenia os odontoideum

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Abstract

Os odontoideum is an uncommon abnormality of the craniovertebral junction (CVJ) that exists as a separate ossicle apart from a hypoplastic dens. Its genesis and natural history have been debated, and its proper treatment remains uncertain.
A 48-year-old woman complained of persistent upper neck pain and paraesthesia of her left side. Magnetic resonance imaging of the CVJ demonstrated an os odontoideum. Dynamic computed tomography scan of the CVJ showed a reduction of the space available for the spinal cord to 50% from extended to flexed position. The patient underwent posterior spinal fusion of C1-C2 using a sublaminar titanium hook and rods fixed in moderate extension.
We discuss the usefulness of the dynamic computed tomography (CT) scan in the evaluation of atlantoaxial motion and the management of this pathology.

Key words: os odontoideum, dynamic CT scan, craniovertebral junction, posterior approach, space available for the spinal cord.

Streszczenie

Os odontoideum jest rzadko spotykaną nieprawidłowością złącza szczytowo-potylicznego, w której słabo wykształcony ząb obrotnika tworzy dodatkową kość. Sposób jej powstawania i historia naturalna są przedmiotem dyskusji; istnieje również niepewność co do właściwego postępowania.
Czterdziestoośmioletnia kobieta zgłosiła się z powodu utrzymującego się bólu górnej części szyi oraz lewostronnych parestezji. W badaniu złącza szczytowo-potylicznego za pomocą rezonansu magnetycznego uwidoczniono *os odontoideum*. Dynamiczna tomografia komputerowa tej okolicy wykazała zmniejszanie się przestrzeni dostępnej dla rdzenia kręgowego o 50% podczas zgięcia w porównaniu z prostowaniem szyi. U chorej wykonano tylne zespolenie kręgów C1-C2 za pomocą tytanowego haka i prętów, uzyskując stabilizację szyjnego odcinka kręgosłupa w umiarkowanym wyproście.
Autorzy omawiają przydatność dynamicznej tomografii komputerowej (TK) w ocenie ruchomości stawu szczytowo-obrotowego i w leczeniu wspomnianej patologii.

Słowa kluczowe: *os odontoideum*, dynamiczna TK, połączenie czaszkowo-kręgowe, dostęp tylny, przestrzeń dostępna dla rdzenia kręgowego.

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Introduction

As a rare anomaly of the second cervical vertebra, os odontoideum has been infrequently documented since Giacomini first described it in 1886 [1]. Although it is uncommon, os odontoideum might lead to atlantoaxial instability and spinal cord compression. On the radiographs, os odontoideum is defined as an ossicle with smooth circumferential cortical margins representing the odontoid process that has no osseous continuity with the body of C2 [2,3]. The aetiology of os odontoideum remains debatable in the literature with evidence for both acquired and congenital causes. The aetiology, however, does not play an important role in its diagnosis or subsequent management [3]. Dynamic imaging of the craniovertebral junction (CVJ) has been used by the authors to confirm the instability of C1-C2. We discuss the usefulness of dynamic computed tomography (CT) scan of the CVJ in the evaluation of atlantoaxial motion and to plan the management of this pathology.

Fig. 1. Spin echo T2-weighted MRI images in sagittal plane in neutral position showing the os odontoideum with predental soft tissue atlantoaxial subluxation, anterior thecal space obliteration by the odontoid process, posterior cord compression by posterior arch of the atlas and increased cord signals at C1/C2 levels

Case report

A 48-year-old woman complained of upper neck pain that lasted about 6 years. The symptoms persisted after several weeks of physical therapy, and she progressively developed paraesthesiae of her left upper and lower limbs and weakness of her lower extremities when walking. She had no recent or distant history of trauma.

The initial physical examination revealed a limited range of motion of her cervical spine to rotation and forward flexion, but no point of tenderness over the posterior spinous processes. Her muscle strength was normal in both the upper and lower extremities; bilateral symmetric hyperreflexia of the upper and lower extremities with Babinski sign was seen. Proprioception and vibratory sensation were impaired on the left side. The rest of the examination and all laboratory findings were normal.

Anteroposterior and lateral plain radiograph examination of the cervical spine demonstrated an irregular ossicle in the expected position of the odontoid process. Lateral radiographs with the patient's neck in flexion and extension confirmed the diagnosis of os odontoideum with instability between the first and second cervical vertebrae. Magnetic resonance imaging (MRI) was performed with the neck in a neutral position and revealed marked retrolisthesis of the os odontoideum with consequent spinal cord compression (Fig. 1). Dynamic CT scan of the CVJ was performed to confirm the atlantoaxial dislocation. The space available for the spinal cord (SAC), being the minimum distance from the posterior aspect of the odontoid or axis to the nearest posterior structure (foramen magnum or posterior ring of the atlas), was measured in flexed and extended positions. It was 16 mm in the extended position and was reduced to 8 mm when the neck was flexed (Fig. 2).

The risks and benefits of surgery were explained to the patient, who consented to cervical stabilisation via a posterior approach. The patient underwent a posterior spinal fusion of the first and second cervical vertebrae using a sublaminar titanium hook and rods fixed in moderate extended position. At the follow-up after 12 months, paraesthesiae had disappeared but mild cervical pain persisted. The control lateral plain radiograph of the cervical spine showed no atlantoaxial instability (Fig. 3), and the patient regained her full activities.

Discussion

The C1-C2 articulation is the most mobile part of the vertebral column. The integrity of the odontoid process

Fig. 2. CT reconstruction images in sagittal plane in flexion and extension showing the os odontoideum and marked reduction of the space available for the cord

and the ligaments determines the stability of the atlantoaxial articulation [4]. When the odontoid process is disrupted due to traumatic or non-traumatic causes, the atlas is displaced along with the odontoid process, leading to atlantoaxial instability. Congenital diseases of the craniovertebral junction, such as os odontoideum, increase the stress on the C1-C2 articulation and lead to hypermobility and eventual frank instability [2].

The pathogenesis of os odontoideum has been extensively discussed in the literature. Some authors have argued that it is congenital and represents the centre of either the proatlas or atlas. However, most authors believe that it is a result of remote trauma leading to a chronic non-united fracture of the odontoid process [5]. Regardless of the origin, the clinical management remains the same [6]. Os odontoideum can present with a wide range of clinical symptoms and signs; it can also be an incidental finding on imaging. The chief symptoms include occipito-cervical pain alone, myelopathy, intracranial symptoms or signs of vertebrobasilar ischaemia [7]. Patients with os odontoideum and myelopathy have been further categorized into three situations: 1) transient myelopathy (commonly after trauma), 2) static myelopathy, and 3) progressive myelopathy [8].

The diagnosis of os odontoideum is not usually considered until imaging is obtained, because such symptoms will reveal other pathologies of the CVJ. The presence of an os odontoideum is usually first suggested after obtaining plain cervical spine X-rays, which most often are sufficient for a diagnosis [3]. In os odontoideum, there is a joint-like articulation between the odontoid and the body of the axis, which appears radiologically as a wide radiolucent gap. This gap may be confused with the normal neurocentral synchondrosis before the age of 5. Therefore, the diagnosis of os odontoideum in children is confirmed by demonstrating motion between the odontoid and the body of the axis. In adults, the diagnosis of os odontoideum is suggested by observing a radiolucent defect between the dens and the body of the axis [4].

CT has been used to better define the bony anatomy of the os odontoideum and the odontoid process [3]. It has the advantage of adequately delineating the bony abnormalities, the extent of cervicomedullary compression and the presence of Chiari malformation [9]. As in our case, dynamic sequences of CT in flexion/extension have been used to depict the degree of abnormal motion between C1 and C2. The odontoid ossicle, however, is fixed firmly to the anterior ring of the atlas and moves along with it in flexion, extension, and lateral slide. The anterior portion of the atlas is usually hypertrophied, and the posterior portion of the ring may be hypoplastic or absent. Most often, there is anterior instability, with the os odontoideum subluxing forward in relation to the body of C2. On the other hand, the instability might also be posterior, with the basion moving posteriorly into the spinal canal during neck flexion. In this situation, attention should be directed to

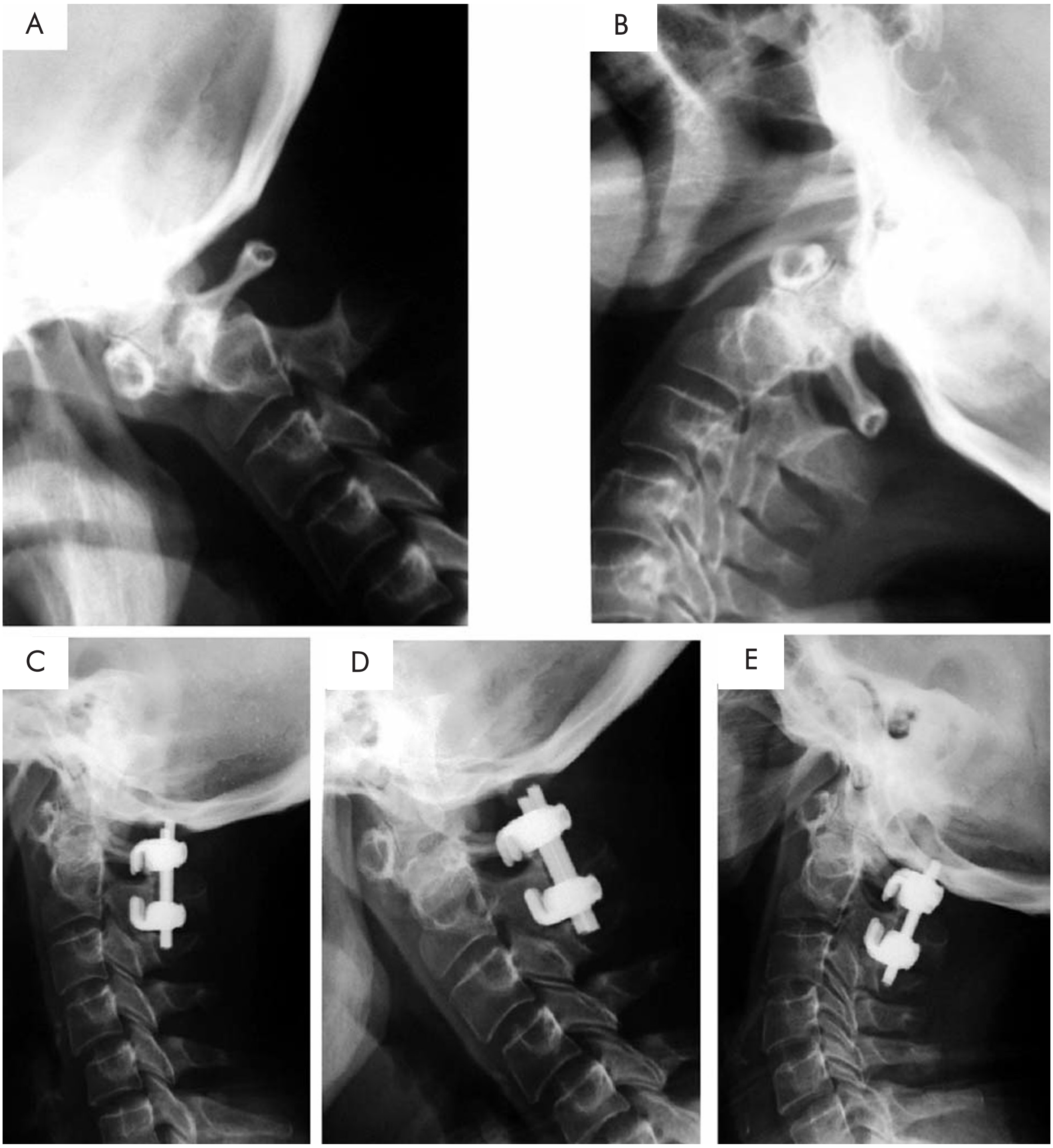

Fig. 3. Lateral x-rays of the cervical spine in flexion and extension. Ventral subluxation of C1 over C2 in flexion (A), which decreases on extension (B). Post-operative control, with neutral (C), flexed (D) and extended (E) position, demonstrating the absence of instability over flexion and extension

the amount of SAC. This was accomplished by measuring the distance from the posterior aspect of the odontoid or axis to the nearest posterior structure (foramen magnum or posterior ring of the atlas) [4]. McRae was first to draw attention to the relationship between neurological symptoms and the SAC. He noted that among his patients with atlanto-occipital fusion, those presenting less than 19 mm of SAC were always symptomatic [10]. This measurement has been defined more specifically as more clinical data became available. It is generally accepted that a reduction of the SAC to 13 mm or less may be associated with neurological problems [3].

Cervical spinal cord compression resulted from the dynamic aspect of our patient's spinal cord compression occurring in the axially flexed position. Therefore, flexion of the neck causes atlantoaxial dislocation and canal compromise, which is also due to the backward slippage of the axis relative to the ring of the atlas, whereas the extension of the neck causes reduction of the atlantoaxial dislocation and increases the SAC. These have to be considered in the management of this pathology when osteosynthesis has to be performed in the extended position. Abe *et al.* in 1976 defined the instability index as an indicator of atlantoaxial instability; this is the change of rate of SAC between flexion and extension. The instability index is calculated by subtracting the maximum distance for the spinal cord between the body of C2 and the anterior portion of the posterior atlantal arch, as well as the minimum distance on flexion-extension radiographs. This sum is divided by the maximum distance and multiplied by 100, and the final value is an indicator of anteroposterior instability [11]. In our case, the instability index was 50%.

Although CT provides indirect evidence of cord compression by showing a decreased spinal canal diameter, it has certain limitations for the visualization of the spinal cord and associated spinal ligaments due to beam hardening artefacts in this site. A preoperative MRI helps to assess soft tissue anomalies, syringomyelia, the extent of cervicomedullary compression and cord changes. Kinematic MRI is an imaging technique that provides real-time images with excellent soft tissue definition in multiple planes, as recently reported [12]. In our opinion, this technique had lower sensitivity to bone anomaly and SAC measurement when compared with dynamic CT. Follow-up MRI is not possible due to the presence of metallic wires or rods used as implants for posterior stabilization, unless titanium implants are used [12,13].

Surgical treatment is not required for every patient in whom os odontoideum is identified. Patients with os odontoideum found incidentally, who have no neurological deficit and no instability at C1-C2 in flexion/extension studies, can be managed without surgical intervention, and longitudinal clinical and radiographic surveillance is recommended [3,5]. Most investigators of this disorder favour surgical stabilization and fusion of C1-C2 instability associated with os odontoideum. Stabilization should be offered: 1) to patients with neurological deficits whether or not one can demonstrate instability; 2) to those with instability and SAC of 13 to 14 mm; and 3) to those in whom grossly abnormal motion is demonstrated [14]. Patients with atlantoaxial dislocation and normal posterior arch of atlas with no associated congenital bony anomalies usually have reducible atlantoaxial dislocation. The reduction may not be evident on plain radiographs of the craniovertebral junction unless sufficient neck extension has been provided. Those patients are at significant risk of neurological deterioration both by flexion and extension of the neck during intubation and patient positioning [9].

Various techniques have been described for surgical stabilization of the os odontoideum, including C1-C2 posterior fusion with Gallie-type wiring, Brooks C1-C2 wiring, occipitocervical fusion, and C1-C2 transarticular screw fixation [15]. As in our case, dynamic CT of the CVJ has the advantage of demonstrating the best position (flexion or extension) in which the SAC is greater, and consequently the neck must be fixed in this position. However, it is difficult and dangerous to perform an atlantoaxial fusion with patients who present irreducible deformities, because sublaminar wiring may cause neurological complications. Thus, an occipitocervical fusion with or without resection of the posterior atlantal arch will be instituted with less risk of serious complications [2]. Ventral decompression of irreducible deformity, in conjunction with C1-C2 or occipitocervical fusion and internal fixation, will be indicated. Odontoid screw fixation has no role in the treatment of this disorder [16].

We conclude that dynamic CT was able to detect reduction of SAC and to confirm indirectly the cord compression that was not seen in a neutral position. This technique may provide new information for evaluation and treatment of many pathological conditions of the cervical spine, such as evaluation of fractures, ligamentous injuries, degenerative processes and congenital abnormalities. Coupled to MRI, it could provide new criteria for classification or revised indications for surgery.

Disclosure

The authors report no conflict of interest.

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