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Os Odontoideum. Case report and literature review

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ABSTRACT

Os odontoideum is an uncommon skeletal abnormality defined by the separation of the odontoid process from the body of the axis vertebrae. This condition can be congenital or acquired, and symptoms may include neck pain, myelopathy, or abrupt neurological deficits. Os odontoideum is commonly diagnosed using imaging modalities such as X-rays, CT scans, and MRI. Treatment options differ depending on the presence of symptoms and the stability of the atlantoaxial joint. Conservative therapy may be appropriate for asymptomatic instances, although surgical intervention, such as C1-C2 vertebral fusion, is frequently required for symptomatic cases or those at risk of spinal cord injury. Long-term follow-up is critical for monitoring potential problems and ensuring optimal results for patients with os odontoideum.

Keywords
Os odontoideum, atlanto-axial instability

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INTRODUCTION

Os odontoideum (OO) first described by Giacomini in 1886 [1] is a circumferentially corticated ossicle separated from a hypoplastic odontoid process of C2 (dens). It is cranially situated relative to the expected position of the odontoid tip. The OO's attachment to the anterior arch of C1 via an undamaged transverse ligament might cause a C1 subluxation relative to C2 during neck movement, whether in flexion or extension. The clinical presentation might vary, from asymptomatic imaging findings to neck soreness and neurological deficits. Prompt imaging is crucial for reducing morbidity and mortality [2].

CASE REPORT

A 60-year-old female presented to the neurosurgery department with neck pain. There was trauma history 8 months prior. Clinical examination revealed a tetraparesis dominating on the left side.

A lateral radiograph of the cervical spine shows a well-corticated, spherical ossicle placed dorsally and slightly cranial to C1's anterior arch. The prevertebral soft tissue width is normal (see Fig. 1). CT images show a corticated margin of this ossicle within 04mm from the

anterior arch of C1, a split atlas malformation, and a posterior tilt of C2 into the ventral spinal canal (Fig. 2).

Magnetic resonance imaging (MRI) demonstrates recession of the posterior wall of the vertebral body of C2 with thickening of the posterior common ligament without signs of rupture, responsible of signal abnormality and thinning of the spinal cord at this level most consistent with myelomalacia (Fig. 3).

The patient underwent surgery; she had an occipitocervical fixation (Fig.4.)

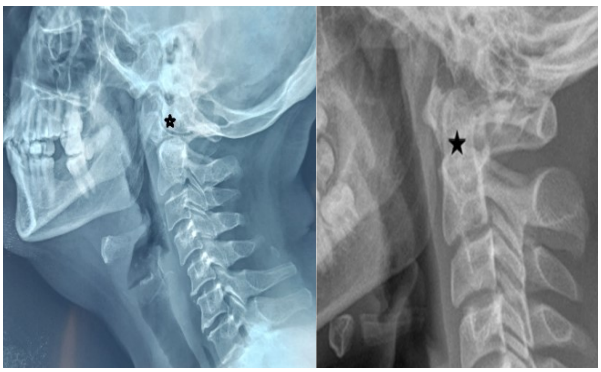

Figure 1. Lateral radiograph of OO. Left, a corticated ossicle is located dorsal and superior to the anterior arch of C1 consistent withan ectopically located os (star). Right, Normal comparison radiograph demonstrating the normal morphology of the odontoid process (star) and A-a distance.

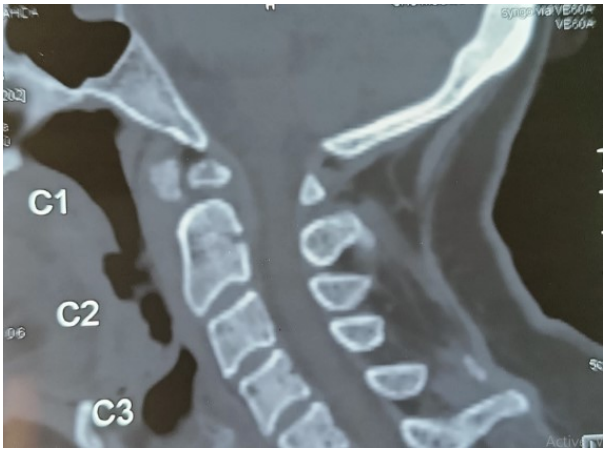

Figure 2. CT images of OO confirm a circumferentially corticated ossicle.

Discussion

Os-odontoideum is defined as a condition where at least half of the assumed odontoid process detached from the rest of the process and body of C2.The detached odontoid process has a smooth inferior border and is not anatomically or functionally connected to the rest of the odontoid process or the body of the axis. Fractures of the odontoid process and os-odontoideum can be distinguished by their irregular margins and history of trauma. Other anomalies connected to the odontoid process include os-terminale, ossification, and osteophyte development at the apical ligament [4]. Although typically an unintentional discovery, it has been linked to significant spinal cord and vertebral artery injury [3].

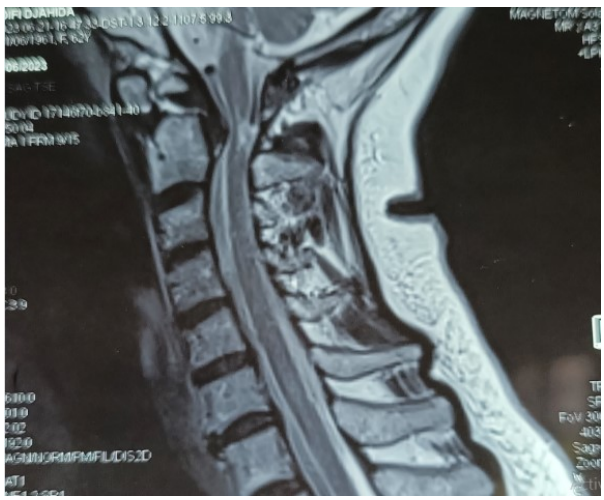

Figure 3. Sagittal T2W magnetic resonance images showed the os odontoideum and compression of the spinal cord.

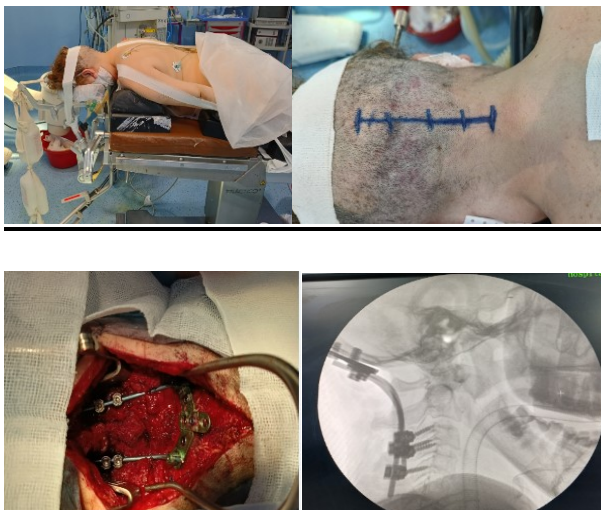

Figure 4. intraoperative images of the occipitocervical fixation with a radiological control image.

Giocomini first reported os odontoideum in cadaveric research in 1886. However, its true

occurrence is unknown, and it is often discovered incidentally [4, 5]. The condition is thought to have both hereditary and traumatic causes. Congenital hypotheses suggest a failure in fusion between the dens and body of the axis. Evidence for this includes the existence of os odontoideum in identical twins, higher gene expression in patients with os odontoideum, and the disorder's relationship with Down syndrome, Morquio syndrome, and Wolcott-Rallison syndrome [5].

The traumatic hypothesis suggests that an initial lesion to the dens causes avascular necrosis and osseous remodeling, ultimately leading to os odontoideum. The development of os odontoideum is likely caused by two distinct factors. Regardless, atlantoaxial instability is clinically significant since it might impact neuronal and circulatory structures in this region [5].

Clinical symptoms may vary from no symptoms, atlanto-axial instability, to spinal cord compression [6]. Cervical myelopathy worsens with age, leading to diminished atlanto-axial stability and symptoms such as neck pain, torticollis, numbness, weakness, and activity limitation. Syncopal events due to os odontoideum have been described [5]. In severe cases, paralysis or death may result. [1].

Zhao et al. published the biggest case series of Os odontoideum, which causes atlantoaxial instability. In a survey of 279 individuals, 84.9% reported pyramidal symptoms, with the majority experiencing paralysis, numbness, or neck pain. Surprisingly, only 40.1% of patients had any history of trauma [10].

In our case the patient had cervicalgia with tetraparesis.

The diagnosis and management of OOs remains identical regardless of their origin. Cervical spine radiographs are commonly used to identify an OO, as shown with our case.

Cervical spine flexion-extension exams can assess atlantoaxial instability [10].However, the degree of atlantoaxial instability is less effective than the absolute diameter of the spinal canal in predicting negative outcomes. Spierings and colleagues propose a critical canal diameter of 13 mm as a high risk for cord damage [6]. CT and MRI scans are frequently obtained to guide subsequent therapy. Both may be used to confirm a suspected anomaly (CT/MRI) or assess cord injury (MRI).A hypertrophied anterior arch of C1 generally indicates underlying chronic instability [1].

Surgical treatment of os odontoideum is indicated for symptomatic cases [9]. Our patient underwent surgery due to neck pain and spinal cord compression.

Several surgical procedures have been developed over time to treat atlantoaxial instability. The Goel approach is particularly suitable for C1-2 instability. Vertebral artery damage is one of the consequences of C2 transpedicular screwing, with a prevalence of 2-8%. Vertebral artery injuries can occur when the C2 pedicle is thin and/or the vertebral artery foramen is within the pedicle. In such circumstances, alternatives to C2 transpedicular screwing, such as C2 laminar screwing, have been proposed. In 2017, Senturk et al. suggested a new alternative approach of C2-C3 transfacet screwing [6].

In a series of 190 patients Atlantoaxial segmental stabilization resulted in clinical symptomatic and neurological improvement in 100% patients [4].
Our patient underwent occipito-cervical fixation as a result of a developed venous drainage that rendered proceeding with a C1-C2 fixation infeasible.

Even though the debate is still ongoing between conservative therapies with imaging follow-up versus prophylactic spinal fusion, asymptomatic os odontoideum with radiological instability can be handled conservatively. However, careful clinical and radiologic follow-up is required as these cases may become symptomatic [8].

Conclusion

Os odontoideum is a rare condition that can cause nonspecific symptoms such as neck soreness or irreversible paralysis. It is important to be aware of its presence. Early diagnosis of a symptomatic os odontoideum decreases patient morbidity and mortality.

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