

Os odontoideum: A case report

Madeline Crnec, BA, DC*

J. David Cassidy, DC, BSC, FCCS**

Dale Mierau, BSPE, DC**

K. Yong-Hing, MD, FRCS(C)***

Os odontoideum is an anomaly of the odontoid process and might be congenital or the result of trauma. Symptoms begin at any age, and the clinical presentation is variable. During radiological examination, it is often a chance finding in an unrelated problem, and just such a case is presented here. The patient presented with rheumatoid arthritis and thoracic backache, was found to have C1-2 instability due to an os odontoideum, and surgical stabilization was planned. Os odontoideum is an absolute contraindication to upper cervical manipulation.

KEY WORDS: odontoid, manipulation, chiropractic.

Introduction

Os odontoideum is an ossicle formed as a result of the disappearance of the central portion of the odontoid process. While it varies in size and shape, it always has a smooth cortical border. Location is either the usual position at the tip of the odontoid process (orthotopic) or nearer the base of the occiput (dystopic) where it occasionally fuses with the clivus.¹

The etiology is highly controversial. There are only two major studies on this topic. Prior to these, there were only small case studies. Fielding et al (1980), after reviewing 35 cases, were convinced that the majority of ossa odontoidea are related to trauma, often in early childhood. Spierings and Braakman (1982), however, were more inclined to see it as a congenital anomaly with the symptoms of trauma as secondary. This conclusion was reached after reviewing 37 cases and considering Fielding's findings as well.

It is clear from both studies that the diagnosis is often incidental and might be made in the later decades of life. Presentation might include neurological signs and will vary in degree and location of pain. Symptoms can include neck, shoulder and/or head pain, pain between shoulder blades, neck stiffness, headaches and pain radiating into the arms. Transient or permanent cord signs might present as weakness, parasthesia or even paralysis of one or all limbs. Not uncommonly, os odontoideum occurs with other congenital anomalies, although Fielding et al contend the relationship should be a more consistent finding if os odontoideum is indeed congenital in origin. Even Spierings and Braakman conceded that their patient pop-

L'odontoïde osseux est une anomalie du processus odontoïde qui peut être congénitale ou être le résultat d'un traumatisme. Les symptômes apparaissent à n'importe quel âge et la présentation clinique est variable. Lors de l'examen radiologique, on a fréquemment l'occasion de découvrir un problème non lié à l'examen. On a découvert chez ce patient qui souffrait d'arthrite rhumatoïdale et d'une douleur à l'arrière de la cage thoracique une instabilité C1-2 due à un odontoïde osseux, on a donc prévu une stabilisation chirurgicale. Une manipulation des cervicales supérieures est entièrement contre-indiquée dans le cas d'odontoïde osseux.

MOTS CLÉS: odontoïde, manipulation, chiropraxie.

ulation was older and might have forgotten childhood injuries. They made note of the nine cases of os odontoideum who had a record of previous normal x-rays which were reported by Fielding et al.

Spierings and Braakman compared three groups of radiological findings i) index of instability using flexion and extension views ii) inner diameter of atlantal ring (D_{at}) and iii) the smallest diameter of the spinal canal (D_{min}), during flexion. Only D_{min} demonstrated a significant difference between groups with and without cord signs, and those with permanent cord signs being the group with the smallest diameter (D_{min}). Even degree of instability in the sagittal plane had no statistical significance. They suggest that D_{min} , measured from the posterior aspect of the body of C2 and the posterior arch of atlas in flexion, becomes clinically significant if less than 13 mm.

Fielding et al reported that surgical treatment, consisting of fusion of either C0-C1, C1-2 or C0-1-2 alleviated pain and instability and resolution of transient neural deficits. Only five of his cases were managed conservatively. In Spierings' and Braakman's study, an operation did not always alleviate pain and in one case pain was made worse. They also reported two post-operative deaths. They concluded that those with only local pain or brain stem symptoms, such as vertigo or nystagmus, be managed conservatively, unless their D_{min} is less than 13 mm. Those patients with progressive cord signs or transient cord signs and a D_{min} of less than 13 mm should be considered appropriate candidates for surgical management.

The following case is an example of a patient with an os odontoideum that was an incidental finding after a trivial injury.

Case report

C.C. is a 31-year-old woman who presented to the office with upper thoracic pain which began two months before presentation, after pushing a car. At that time, she felt a snap in the neck and experienced immediate pain which radiated down both arms to the elbows. Her pain was further aggravated by a subsequent fall on ice during which she experienced a flexion-extension type injury to her cervical spine. Pain was relieved by rest and heat. She had previously been diagnosed as having

* Resident III, Chiropractic Sciences, C.M.C.C., 1900 Bayview Avenue, Toronto, Ontario

** Private Practice, Fourth Avenue Chiropractic Clinic, 208 - 119 4th Avenue South, Saskatoon, Saskatchewan, S7K 5X2

*** Department of Orthopaedics, University Hospital, Saskatoon, Saskatchewan, S7N 0X0

Address reprints to: Dr. JD Cassidy, Fourth Avenue Chiropractic Clinic, 208 - 119 4th Avenue South, Saskatoon, Saskatchewan S7K 5X2

© M Crnec, JD Cassidy, D Mierau, K Yong-Hing 1986

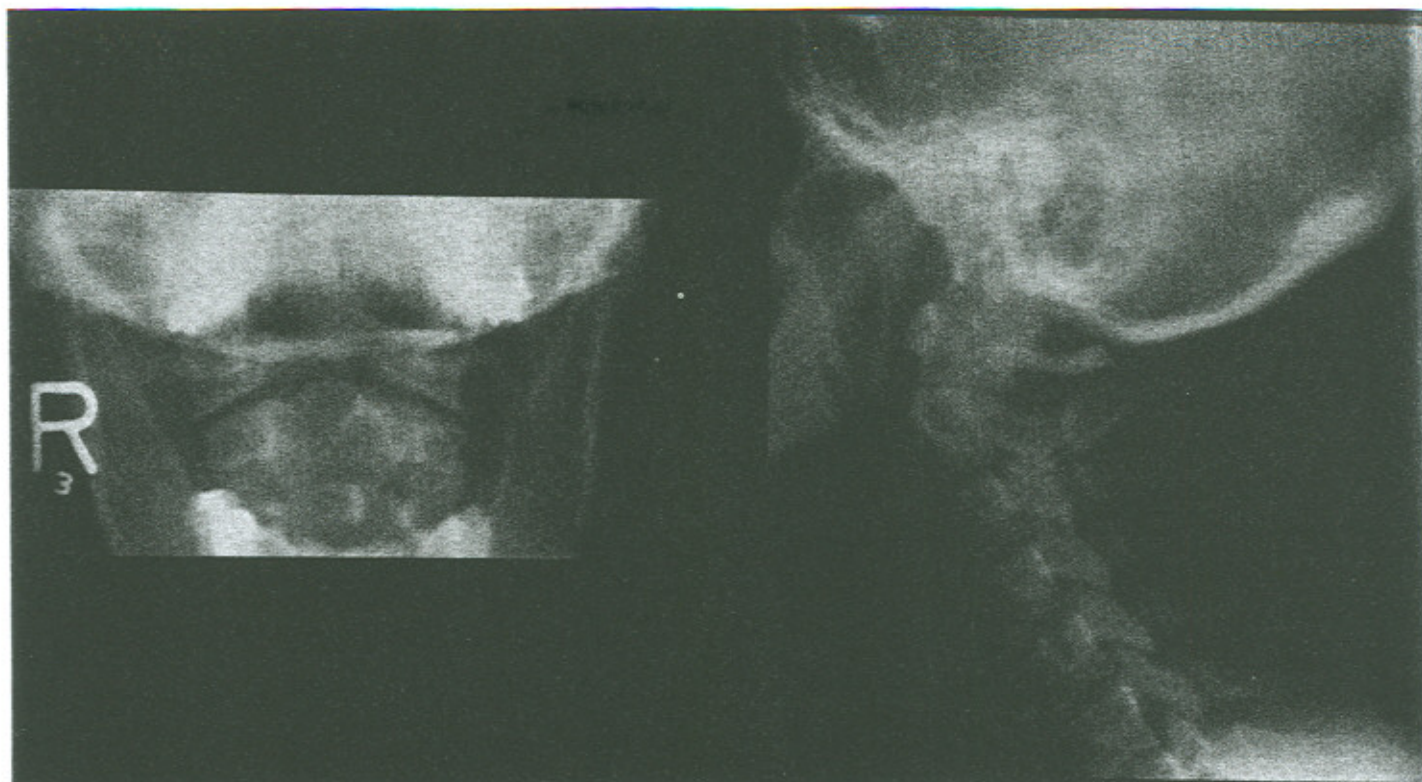


Figure 1: Open mouth view in AP projection and lateral views of cervical spine demonstrate a rudimentary odontoid process with a small, poorly visualized bone posterior to the anterior arch of C1.

rheumatoid arthritis and complained of bilateral wrist pain. Her mother was quite crippled with the disease. She also had hepatitis nine years before presentation.

On examination, she appeared pale and somewhat jaundiced. There was full and painfree movement of the cervical spine with no paraspinal tenderness. She was tender to palpation over the spinous processes of T2 and T3. Deep tendon reflexes, sensation and motor power were all normal in the upper extremities.

There was tenderness over the anatomical snuff boxes in both wrists, with no palpable increase in synovium. She had previously fractured both hands. There was full range of motion in both wrists. Both hands and feet demonstrated atrophic changes of the nails with redness and perspiration.

A working diagnosis of posterior joint dysfunction of the upper thoracic spine was made. However, the presence of rheumatoid arthritis suggested two other possible differential diagnoses, upper cervical instability which could produce bilateral arm pain and/or thoracic compression fracture as a result of rheumatoid osteopenia. Therefore, to make a definitive diagnosis, a radiological examination of the cervical and upper thoracic spine was done.

While the thoracic spine was normal, the cervical study revealed an os odontoideum and a marked instability of C1 on C2 of 8 to 9 mm, in flexion-extension. D_{min} , measured in flexion, was 16 millimetres.

She was referred to an orthopaedic surgeon for evaluation, and it was decided that a spinal fusion of C1-2 would be required to stabilize this segment. The surgeon in this case felt the instability was significant enough to warrant surgery, re-

gardless of the acceptable D_{min} .

It was also decided that the upper thoracic joint dysfunction could be treated, using manipulation, without involving the cervical spine. Nine anterior thoracic manipulations relieved the thoracic pain.

Discussion

There are several confounding factors which come into play in this case. It is the family history of rheumatoid arthritis plus the rheumatoid symptomatology present in the patient that made a convincing argument for a radiographic examination of what might otherwise have been considered simple mechanical back pain as a result of a recent trauma. Rheumatoid arthritis leads to instability of the atlanto-axial joint in approximately 25 percent of cases as a result of inflammation eroding the odontoid process and/or weakening or rupturing the transverse ligament.^{4,5} Osteopenia is also part of the pathology of this disease and another reason why a radiological examination should be performed on R.A patients before manipulation of any area is administered. Another consideration in the differential diagnosis in this case was the possibility that degenerative changes of the cervical spine might refer pain to the thoracic region. This patient also appeared somewhat jaundiced on presentation and a sickly appearance always warrants an in-depth investigation.

The os odontoideum was an asymptomatic incidental finding in this case. It is the degree of instability and the potential for permanent damage, should another hyperflexion/hyperextension injury occur, that persuaded the patient and the surgeon that a spinal fusion would be prudent.

The diagnosis of C1-2 instability is both a clinical and ra-

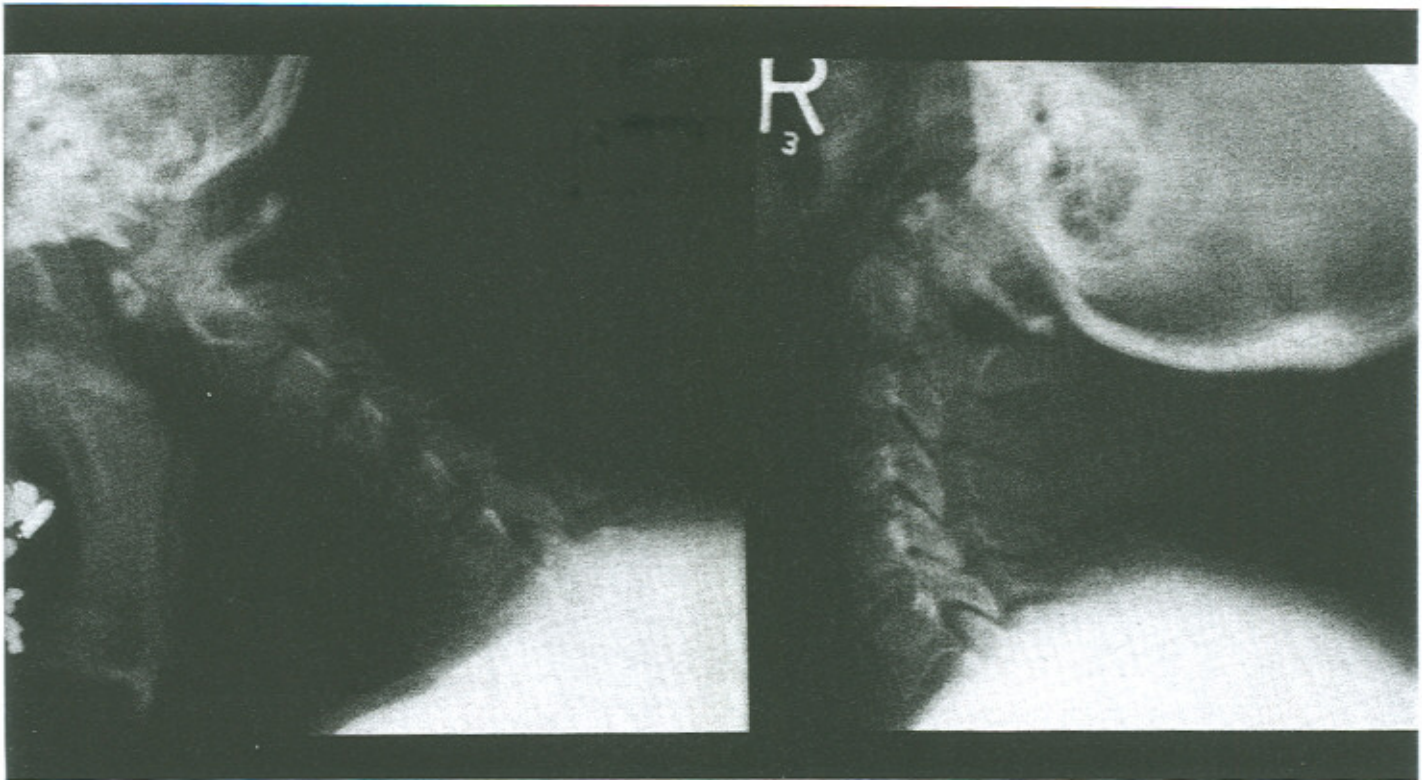


Figure 2: Flexion and extension views of the cervical spine demonstrate hypermobility at C1-2 of 8 to 9 mm.

diographic one. Spierings and Braakman categorize their patient population into those without cord signs and those with cord signs and describe the symptomatology on presentation. It is the group without extensive neurological involvement such as paraplegia which is most likely to present to a chiropractor's office. Symptoms in their study included uni- or bilateral neck, shoulder and/or arm pain, frontal or occipital headaches, tinnitus, syncope on extension of the neck, light-headedness, a twitching of the left leg on right rotation of the head or transient or progressive weakness or paresthesia of one or more limbs. In almost a quarter of these cases, the onset of symptoms was related to an injury, either hyperflexion or a blow to the head, but definitive diagnosis was delayed. Half of the cases without cord signs had no previous history of injury. Even more interesting, in the Spierings and Braakman study, was the finding that none of the cases involving permanent cord signs had a history of trauma. Fielding et al list the decreased range of motion of the cervical spine as the most common abnormal physical finding.

C1-2 instability is determined radiographically using the comparison of flexion and extension views to measure the i) atlanto-dental interval (ADI), that being the joint space between the atlas and the odontoid process and ii) spinous-lamina line. The normal upper limit of the ADI in flexion is 2.5 mm in adults and up to 4 mm in children.⁴

The spinous-lamina line will be disrupted in instability and can be used when the ADI cannot be measured, as in agenesis of the odontoid or when there is instability without disruption of the transverse ligament. In rheumatoid arthritis both the spinous-lamina line and the ADI will be changed.

It is Spierings' and Braakman's contention that the minimum diameter of the spinal canal, between the posterior body of C2 and the posterior arch of atlas, (D_{min}) in flexion is more clinically significant than the degree of instability. They found a D_{min} of less than 13 mm to be the point at which surgical fusion should be implemented.

Conclusion

When one considers the extreme variability of symptomatology in atlanto-axial instability and the grave consequences of manipulating such a segment, one is ethically bound to rule instability out of the differential diagnosis, especially in cases with i) a history of rheumatoid arthritis, ii) a recent or previous injury to the cervical spine involving hyperflexion or a blow to the head or iii) where etiology of symptoms cannot be reasonably determined.

This case demonstrates the value of routine radiological examination of the upper cervical spine prior to manipulative therapy.

References

- 1 Fielding JW, Hensinger RN, Hawkins RJ. Os Odontoideum J Bone Joint Surg April 1980; 62-A, 3:3, 76-283.
- 2 Spierings EL, Braakman R. The management of os odontoideum, analysis of 37 cases. J Bone Joint Surg 1982; 64-B, 4:422-428.
- 3 Panjabi M, White A. Clinical biomechanics of the spine. Toronto: J B Lippincott, 1978: 204-207.
- 4 Greenfield G B. Radiology of bone diseases 3rd ed. Philadelphia & Toronto: J B Lippincott, 1980: 686-728.
- 5 Sherk HH. Lesions of the Atlas and Axis. Clinical Orthop 1975; 109, 33-41.