Case report

Cervical myelopathy involving os odontoideum and retro-odontoid cyst treated with Harms C1–C2 arthrodesis

S. Le Pape a, R. Gauthé a, C. Latrobe a, J. Leroux b, X. Roussignol a, M. Ould-Slimane a,∗

a Département de chirurgie orthopédique et traumatologique, institut régional du rachis, CHU Charles-Nicolle, 1, rue de Germont, 76031 Rouen cedex, France
b Clinique chirurgicale infantile, CHU Charles-Nicolle, 1, rue de Germont, 76031 Rouen cedex, France

A R T I C L E   I N F O

Article history:
Received 7 March 2016
Accepted 16 June 2016

Keywords:
Os odontoideum
Spinal cord compression
Retro-odontoid cyst
C1–C2 arthrodesis
Laminectomy

A B S T R A C T

An episode of acute decompression of cervical myelopathy occurred after an injury without fracture of an os odontoideum associated with a compressive retro-odontoid cyst. The 51-year-old female patient presented Fränkel C, Nurick grade 4 neurological status and pyramid syndrome. The initial MRI demonstrated an intramedullary T2 hyperintense signal in the context of spinal cord narrowing. The retro-odontoid cyst demonstrates atlantoaxial instability related to the os odontoideum. Harms C1–C2 arthrodesis without laminectomy was performed and the cyst disappeared completely. Spinal cord decompression was thus obtained on the MRI taken 3 months later. Neurological recovery was complete and continued at 1 year. © 2016 Elsevier Masson SAS. All rights reserved.

1. Introduction

Os odontoideum is a rare malformation of the upper cervical spine. Association with a retro-odontoid cyst has been described as a mark of local instability. Nonspecific symptomatic manifestations are erratic and vary from cervical pain to neurological disorders. Although guided by the severity of the clinical findings, treatment has not been fully codified. Resection of the cyst is technically difficult [1]. C1 or C2 laminectomies are generally associated with destabilization of the occipitocervical junction [2]. Treatment of the instability theoretically allows resorption of the cyst and thus decompression of the spinal cord can be obtained [3–5]. To our knowledge, there have been no published cases of treatment of acute decompression of cervical myelopathy with C1–C2 arthrodesis without laminectomy.

The present description of a case whose treatment was based on the pathophysiological hypothesis was the occasion for a literature review on the subject.

2. Observation

A 51-year-old female patient, with no medical or surgical history, was involved in a medium-level kinetic-energy traffic accident. No hemodynamic or respiratory problems were observed at initial care. The patient described cervical pain associated with paresthesia in the right upper limb. The neurological examination found a pyramidal syndrome and a Fränkel C, Nurick grade 4 sensorimotor status.

The initial whole-body scan demonstrated the existence of a bony continuity that had not been displaced within the odontoid processus (Fig. 1). The existence of corticalized edges led to correcting the initial diagnosis of fracture to os odontoideum. To better evaluate the existence of recent neurological disorders, magnetic resonance imaging completed the work-up, allowing the discovery of a retro-odontoid cyst with T2 hyperintensity and T1 hypointensity surrounded by a crown of echo gradient T2 hypointensity. This cyst exerted a mass effect on the spinal cord. The intramedullary T2 hyperintensity signified a spinal cord disorder (Fig. 2a and b).

Treatment aimed to stabilize the instability by limiting the cyst’s invasiveness. The cyst’s composition was probably related to the instability of the os odontoideum [6]. We found it useful to treat the etiology with a C1–C2 arthrodesis, applying the Harms technique [7], aiming for the C1 joint surfaces and the C2 pedicles. A corticocancellous bone graft from the iliac crest was attached to the rasped posterior arcs of these two vertebrae (Fig. 3).

Clinical progression was rapidly favorable with regression of the paresthesias and motor recovery to 4 out of 5 beginning the 3rd postoperative day. The patient was discharged on D7 with no scarring complications. She no longer presents paresthesia in the right upper limb, her neurological status was Fränkel E and Nurick grade 0. At 3 months, neurological recovery remained complete. Some cervical pain was reported (VAS = 3/10) without it

http://dx.doi.org/10.1016/j.jotrs.2016.06.008
1877-0568/© 2016 Elsevier Masson SAS. All rights reserved.
being incapacitating. The follow-up MRI demonstrated complete disappearance of the retro-odontoid cyst and signs of persistent spinal cord involvement (Fig. 4a and b). The patient returned to her profession as a check-out assistant 6 months after the intervention. At the last follow-up (1 year), no neurological deterioration had appeared. The x-rays showed no signs of degeneration of the occipitocervical junction.

3. Discussion

3.1. Nosology

Os odontoideum is a rare lesion of the craniocervical junction. This is an independent bony structure that always extends over a hypoplastic odontoid process [8]. Two main etiological theories have been described:

- an embryological origin suggesting a fusion abnormality between the odontoid process and the body of the axis;
- a traumatic origin suggesting progression of a fracture toward aseptic osteonecrosis [8].

Different names are associated with retro-odontoid structures involving spinal cord compression: mass, cyst, pseudotumor, granuloma, and pannus. Atlantoaxial instability related to os odontoideum could instigate the appearance of a retro-odontoid cyst [6]. Excessive solicitation of the transverse ligament may be involved in the pathophysiology according to several authors [6].
3.2. Treatment

The low number of cases identified makes it impossible to establish a consensus on treatment. With the neurological disorders and the imaging studies reported herein, the main objective of treatment is spinal cord decompression. Etiological treatment alone, therefore, appeared to be a wise choice based on the pathophysiological hypothesis [4]. Stabilizing os odontoideum with C1–C2 arthrodesis aimed to eliminate the cyst, to decompress the spinal cord. After verification if its feasibility [9], the Harms technique was selected. Abstaining from laminectomy as recommended by some teams [10,11], can be explained by the anterior position of the cyst and by the absence of posterior spinal cord compression. Moreover, the C1 blade is a good posterior bone graft surface. The clinical and imaging results argue in favor of this method’s efficacy with complete disappearance of the cyst and neurological problems.

Zhang et al. [12] proposed an analysis of ten similar cases. The time to treatment depended on the initial clinical severity. In cases with neurological symptoms, surgery was performed on the 4th day and consisted in C1–C2 arthrodesis associated with C1 laminectomy. If there was only a hyperintense intramedullary signal, surgery was undertaken on the 8th day. Our observation differs in that laminoplasty was not performed despite the existence of neurological impairment.

3.3. Alternatives

Several authors have proposed alternative strategies. Klimo et al., less conservative, associate C1–C2 arthrodesis with laminectomy, arguing that there is a risk of evolving toward myelopathy [3]. However, posterior approaches and, by extension, C1 and C2 laminectomies, result in muscle lesions that are reputed to be destabilizing. They require prolonged radiological monitoring. Some teams do not treat the instability but only the neurological compression by performing either a C1 laminectomy [10,11] or cyst resection via a transoral approach [1]. Others suggest simple radiological and clinical monitoring [13]. These methods do not treat the pathophysiological cause of the problem. They therefore expose the patient to persistence of the problem of recurrence of spinal cord compression.

4. Conclusion

Retro-odontoid cysts associated with unstable os odontoideum can lead to symptomatic spinal cord compression. C1–C2 arthrodesis allows resorption of the cyst and ensures spinal cord decompression.

Disclosure of interest

The authors declare that they have no competing interest.

References


