Retro-odontoid Degenerative Pseudotumour Causing Spinal Cord Compression and Myelopathy: Current Evidence on the Role of Posterior C1–C2 Fixation in Treatment

Francesco Certo, Massimiliano Maione, Massimiliano Visocchi, and Giuseppe M. V. Barbagallo

Abstract *Background:* A retro-odontoid pseudotumour compressing the spinal cord and causing myelopathy is often associated with an inflammatory condition such as rheumatoid arthritis. A degenerative non-inflammatory retro-odontoid pseudotumour responsible for clinically relevant spinal cord compression is a rare condition described in small clinical series and is likely associated with craniovertebral junction hypermobility or instability–like conditions. For several years, direct removal of the lesion through an anterior or lateral approach has been advocated as the best surgical option. However, in the last decade the posterior approach to the craniovertebral junction, to perform C1–C2 fixation and C1 laminectomy without removal of the retro-odontoid tissue, has demonstrated its efficacy in reducing retro-odontoid pannus as well as in obtaining improvement of myelopathy.

Methods: In this paper we analyse the clinical and radiological outcomes of seven patients (five males and two females) treated with posterior C1–C2 fixation and C1 laminectomy for a degenerative non-inflammatory retro-odontoid pseudotumour responsible for spinal cord compression. C1 laminectomy provided immediate spinal cord decompression. We also review the relevant literature focusing on associated cervical degenerative conditions that may contribute to triggering or acceleration of atlantoaxial hypermobility or 'instability', causing formation of the retro-odontoid tissue.

Results: The mean follow-up period (of six followed-up patients) was 55.8 months (range 10–96 months). In all cases the Nurick score at the latest follow-up visit demonstrated clinical improvement; magnetic resonance imaging during follow-up demonstrated progressive reduction of the retro-odontoid pseudotumour in all but one patient, who died of surgery-unrelated disease in the early postoperative period. No vascular or neural damage secondary to C1–C2 fixation was observed.

Conclusion: C1–C2 fixation associated with C1 laminectomy is an effective surgical option to treat myelopathy secondary to a degenerative retro-odontoid pseudotumour. In these cases, direct removal of intracanalar tissue compressing the spinal cord is not required, as C1–C2 fixation is sufficient to cause its disappearance.

Keywords Atlantoaxial fixation · Craniovertebral junction · Cervical myelopathy · Odontoid process · Retro-odontoid pseudotumour

Introduction

A retro-odontoid pseudotumour [1], also known as pannus or a phantom tumour [2], is non-neoplastic, fibroreactive tissue involving the odontoid process and the surrounding structures of the craniovertebral junction [3, 4], causing different degrees of spinal cord compression.

Although the aetiology and pathophysiology of retro-odontoid pseudotumour formation and growth are unclear, various predisposing factors associated with its development have been identified, including inflammatory diseases (rheumatoid arthritis [5] and psoriatic arthritis [6]) and, less frequently, non-inflammatory pathologies (including post-traumatic pseudoarthrosis of the odontoid process, unstable odontoid fractures [7], os odontoideum [3, 8–10], postlaminoplasty kyphotic cervical instability [11], long-term haemodialysis [12], craniocervical junction malformations [10] and chronic atlantoaxial subluxation/instability [1, 13]). In cases related to inflammatory conditions, chronic damage of the C1–C2 ligamentous complex has been postulated as a likely factor inducing the development of retro-odontoid pannus [14]. Conversely, the pathogenesis is not clear in non-inflammatory cases, although atlantoaxial instability is

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often identified [13, 15]. Barbagallo et al. hypothesized that extensive subaxial cervical spine spondylotic changes, with reduced segmental hypomobility, might be coupled with compensatory hypermobility in the C1–C2 area and that such a phenomenon would eventually induce formation of retro-odontoid pannus [16].

Surgical treatment of a retro-odontoid pseudotumour can be performed by direct removal of the mass through a transsphenoidal/transoral/transpharyngeal approach [5, 17], a high cervical lateral approach [18] or a posterior extradural approach via laminectomy. Recently, posterior approaches to the craniovertebral junction, such as occipitocervical [10, 11, 19–22] or atlantoaxial [7, 9, 19, 23–28] fixation or C1 laminoplasty [2], have been reported by several authors, as they can induce a progressive, spontaneous reduction in the volume of the pseudotumour, avoiding the need for its direct resection.

We report our institutional experience with a cohort of patients presenting with compressive myelopathy related to a degenerative, non-inflammatory retro-odontoid pseudotumour, surgically managed with C1–C2 fixation combined with laminectomy of the C1 posterior arch. The timing of the pseudotumour reduction over medium-term follow-up and its relation to each patient's clinical and radiological features are analysed.

Materials and Methods

Patients

Between July 2009 and August 2016, seven patients (five male and two female), with a mean age of 63.7 years (range 55–76 years), were treated. Clinical and radiological data were retrieved from the patients' charts and/or the institutional electronic database. Clinical and radiological follow-up evaluations were performed at 3, 6 and 12 months after surgical treatment, and then every year.

Clinical and Radiological Assessments

All patients underwent neurological evaluation before surgery, postoperatively and at each follow-up visit. The Nurick score was used to assess the severity of myelopathy. The preoperative imaging protocol was the following: cervical x-rays with flexion–extension views, magnetic resonance imaging (MRI) and computed tomography (CT) angiography of the craniovertebral junction with multiplanar reconstruction. Retro-odontoid pannus features were classified according to MRI findings, as proposed by Yonezawa in 2013 [29]: type 1 pannus (hyperintense on T2 and hypointense on T1) was observed in two cases; type 2 pannus (hypointense on T2 and iso- or hyperintense on T1) was identified in one case; and type 3 pannus (a combination of hypo- and hyperintense on T2 and T1) was depicted in four cases. Interestingly, a patient with type 2 pannus had eggshell calcification surrounding the retro-odontoid pannus, clearly visible on the CT scan. Preoperative radiological evaluation also documented the presence of atlas assimilation in two patients, platybasia associated with C1 assimilation and consequent Chiari malformation type I in one patient, and diffuse idiopathic skeletal hyperostosis (DISH) in two other patients (Fig. 1). Both MRI and CT scanning clearly documented signs of subaxial spondylosis in all patients. Two patients had already undergone surgery for cervical spondylotic myelopathy at the C5–C6/C6–C7 and C5–C6 levels, respectively (Table 1). The preoperative radiological evaluation documented signs of advanced subaxial cervical spondylosis in all patients.

Cervical x-rays, CT scanning and MRI were also performed immediately after surgery. Moreover, the patients underwent MRI over the follow-up period to document the progressive reabsorption of retro-odontoid tissue.

Surgical Treatment

Depending on the patients' individual associated conditions (i.e. the presence of atlantoaxial assimilation), different surgical approaches were performed. Three patients underwent C1–C2 fixation according to the Goel–Harms technique. Occipitocervical fixation was performed in three patients: two with atlantoaxial assimilation and one with radiological signs of pre-existing C0–C1 fusion (in one patient, screws were inserted into the occipital condyles; in the remaining two cases, an occipital plate was implanted). In two other patients, C1–C2 fixation with C2 translaminar screws was performed (Fig. 1). Resection of the C1 posterior arch was performed in four of the seven patients, and C1–C3 laminectomy and posterior decompression of the foramen magnum were performed in two other cases (Table 1). All surgical procedures were performed by the senior author (GMVB).

Results

The mean follow-up period (in the six followed-up patients) was 55.8 months (range 10–96 months).

All surgical procedures were performed without perioperative complications. Postoperative x-rays and CT scans demonstrated correct device positioning in all cases. One patient died 10 days after the surgery because of acute kidney disease and multiorgan failure. The mean Nurick score of the other six patients decreased from 4 (range 3–5) to 2.3 (range 1–3) by the time of the latest follow-up. The neurological status

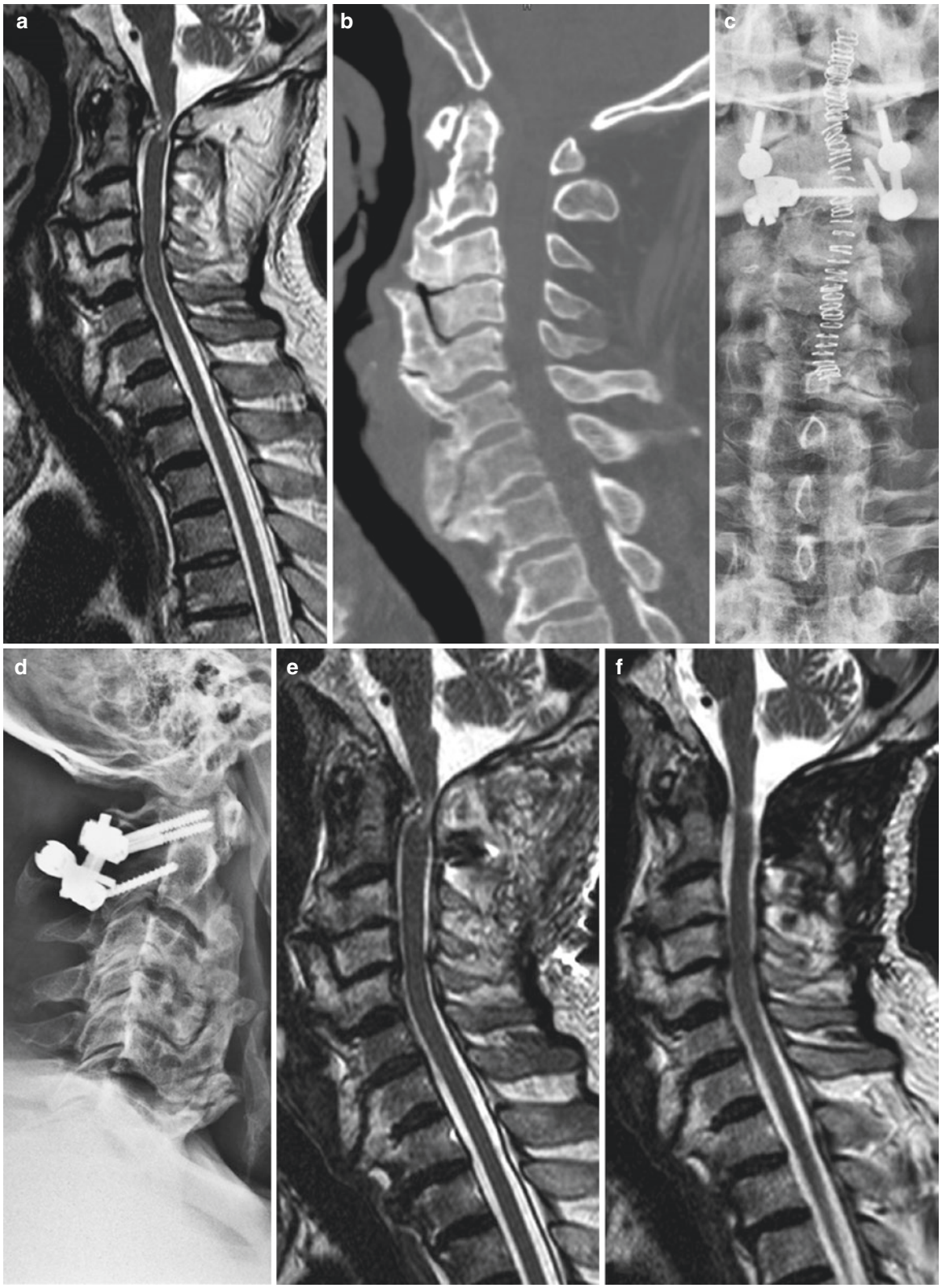

Fig. 1 Images of patient #7. (**a**) Preoperative sagittal T2-weighted magnetic resonance imaging (MRI) showing the presence of retro-odontoid pannus compressing and dislocating the spinal cord. (**b**) Sagittal reconstructed computed tomography (CT) scan showing multiple anterior osteophytes limiting the physiological motion of the cervical spine. (**c**) Postoperative anteroposterior x-rays and (**d**) postoperative lateral X-rays demonstrating C1–C2 fixation; because the right C2 pedicle was too narrow, a translaminar screw was used (a hybrid construct). Postoperative 1-month and 6-month spine MRI documented (**e**) the progressive reduction and (**f**) the disappearance of the retro-odontoid pseudotumour

Table 1 Review of institutional series of degenerative retro-odontoid pannus treated by posterior fixation

Patient (age/sex)	AAI or AAS (Y/N)	Subaxial spondylosis (level/s)	Associated conditions	Surgical procedure	Time to pannus reduction/ disappearance	Follow-up
55/M	N	C2–C3	–	C1–C2 fixation with C1 laminectomy (C1 lateral mass and C2 pedicle screws)	8 months	96
63/M	N	C5–C6 (fusion)	DISH	C1–C2 fixation with C1 laminectomy (C1 lateral mass and C2 pedicle screws)	12 months	83
76/F	N	C4–C5/C5–C6	Atlas assimilation	C0–C3 (C2 translaminar screws and C3 lateral mass)	NA	NA
58/F	N	C4–C5	Atlas assimilation	Occipito-C5 fixation (C2 translaminar screws and C3 lateral mass, C4 and C5 pedicle screws on one side and lateral mass screw on the other side)	6 months	78
64M	N	C4–C5/C5–C6	Atlas assimilation, platybasia and Chiari malformation	Occipito-C3 fixation in 1 pt with C1 assimilation (C2 translaminar screws and C3 lateral mass)	14 months reduction (pannus still present at 24 months FU)	46
59/M	N	C5–C6	Previous ACDF	C1–C2 fixation with C1 laminectomy (C1 lateral mass and C2 pedicle screws)	12 months	22
71/F	N	C3–C4/C4–C5/ C5–C6	DISH	C1–C2 fixation with C1 laminectomy (C1 lateral mass and C2 one translaminar screw and one pedicle screw)	6 months	10

AAI atlanto-axial instability, *AAS* atlanto-axial subluxation, *DISH* diffuse idiopathic skeletal hyperostosis, *ACDF* anterior cervical discectomy and fusion

remained unchanged immediately after surgery in all but one patient (the one showing calcified pannus). This patient reported transient worsening of pre-existing right upper limb weakness, which progressively recovered within 6 months after the surgery. A clinical improvement over the follow-up period was documented in all patients.

Radiological follow-up was performed in six of the seven patients. A significant and progressive decrease in retro-odontoid tissue was documented in all cases during the follow-up period. In the patient with calcified retro-odontoid tissue, incomplete reabsorption of the pseudotumour was documented 2 years after the surgery, albeit that the patient had experienced a slight clinical improvement (the Nurick score varied from 4 to 3). A direct anterior transoral approach to remove the pseudotumour was advised, but the patient did not consent to further surgery.

Discussion

Compressive myelopathy due to a retro-odontoid pseudotumour has been described as being related to chronic inflammatory conditions involving the skeletal system and joints, such as rheumatoid or psoriatic arthritis [5, 6, 24, 30]. Rare cases of retro-odontoid pannus associated not with an inflammatory condition but, rather, with a purely degenerative aetiology have been described [3, 7–12, 25]. The formation and growth of degenerative retro-odontoid pannus has been correlated with underlying instability of the atlantoaxial joint, which is responsible for continuous mechanical stress on the ligamentous and cartilaginous complex of the C1–C2 joint. This condition may lead to altered tissue turnover, causing formation of retro-odontoid pannus. However, even more rare cases of retro-odontoid pannus without overt C1–C2 instability have been also described [26, 31].

Symptomatic patients usually present with symptoms and signs of pyramidal tract involvement as a result of direct compression of the anterior columns of the spinal cord. Direct decompression can be achieved by resection of the abnormal retro-odontoid tissue through an anterior (transoral, transsphenoidal or transpharyngeal) [5, 17] or anterolateral route [18]. Posterior approaches to the craniovertebral junction have been more recently proposed as an alternative to anterior or anterolateral approaches, with the rationale of managing the retro-odontoid pseudotumour and treating the C1–C2 instability, which may have caused its formation. Different surgical techniques have been proposed to stabilize the craniovertebral junction, according to the different anatomical features of patients [32–34]. In general, when the anatomy of the C0–C1–C2 joints is normal and there are no signs of instability at the C2–C3 level, C1–C2 posterior fixation using a Magerl or Goel–Harms technique is sufficient to ensure solid fixation of the craniovertebral junction [32, 33, 35–40]. Conversely, in cases of atlas assimilation, platybasia, etc., occipitocervical fixation appears to be the most effective and safe surgical option [41]. Controversies exist regarding the best surgical strategy for patients without overt C1–C2 instability [26, 31]. In such cases a different pathogenesis, not related to the presence of C1–C2 instability, has

been postulated. Indeed, some authors have suggested that simple posterior neural decompression may be effective [2, 31]. Conversely, a case of a patient affected by a retro-odontoid pseudotumour not associated with C1–C2 subluxation, which was successfully managed by C1–C2 fixation, was reported in 2009 by Tanaka et al. [26], who observed a clinically and radiological significant reduction of the retro-odontoid pseudotumour 6 months after the surgery.

Atlantoaxial instability was not present in any patient included in the present series. In all cases we observed 'delayed' indirect neural decompression after C1–C2 fixation, related to the progressive reduction and disappearance of the retro-odontoid pannus. Moreover, we found a constant presence of subaxial spondylosis in all cases. This findings has not been well investigated in the literature. Chikuda et al. published findings from a series of ten patients who had retro-odontoid pannus without associated inflammatory diseases [4]. Eight patients in that series had no signs of C1–C2 instability, and four of those eight also had subaxial spondylosis. We have already hypothesized in a previous paper that the genesis of degenerative retro-odontoid pannus in cases not associated with an inflammatory condition or C1–C2 instability may be explained by abnormal mechanical stress on the craniovertebral junction, caused by reduced motion at subaxial levels [16]. In our series, the hypothesis of subaxial reduced motion as a cause of retro-odontoid pannus development is supported by the fact that two of our patients had previously undergone anterior cervical discectomy and fusion at subaxial levels and two other patients suffered from DISH.

In our previous publication on this topic, we also performed a systematic literature review of studies reporting retro-odontoid pannus treated by a posterior approach [16]. We found only a few reports on retro-odontoid pannus that was not associated with inflammatory disease or with atlantoaxial subluxation or instability.

The timing of the reduction/disappearance of the retro-odontoid tissue was variable in our series, as in the other published series [7, 10, 11, 13]. However, a retrospective classification of preoperative MRI scans according to the classification proposed by Yonezawa et al. [29] for rheumatoid retro-odontoid pannus showed that the timing of its disappearance in two patients with type 1 pannus and in three of four patients (one patient died 10 days after surgery) with type 2 pannus ranged from 6 to 12 months. One patient with type 2 pannus had a reduction but not disappearance of the pannus at follow-up. In this patient the presence of eggshell calcification of the retro-odontoid tissue probably limited the reabsorption process. The timing of pannus reduction/disappearance reported in our series is similar to those described in other recent series. We did not observe immediate or early postoperative disappearance of the pseudotumour, as reported by Shah et al. [42] and Ito et al. [43].

Posterior decompression including the C1 posterior arch was performed in four of the seven cases in the present series. We believe that this further surgical adjunct may ensure early neural decompression in patients with a more severe clinical status [44].

A further interesting finding in the present series is related to the medium-term follow-up results. Indeed, we observed that the progressive reduction of the retro-odontoid pseudotumour demonstrated by sequential follow-up MRI was accompanied by consequent improvement in the clinical status, documented by the reduction in the Nurick score. In a subsequent phase, both the clinical status and MRI findings remained stable. In two patients, worsening of subaxial spondylosis required further surgery 13 months and 38 months after the previous surgery, respectively. The clinical history of such cases supports the hypothesis that a retro-odontoid pseudotumour represents a clinical and anatomopathological manifestation of a larger and dynamic degenerative phenomenon, involving—at an earlier stage—the subaxial spine and then extending also to the craniovertebral junction.

Conclusion

A degenerative retro-odontoid pseudotumour not associated with inflammatory disease is a rare condition requiring proper management. Posterior craniovertebral junction fixation is a reliable and effective option leading to 'delayed' neural decompression related to progressive pannus reabsorption. This condition remains stable at medium-term follow-up, albeit that associated subaxial spondylosis, which is often observed in cases without atlantoaxial instability, may require further surgical procedures. Multicentre studies and larger clinical series are encouraged in order to clarify the optimal management of these complex conditions.

Competing Interests The authors declare that they have no competing interests.

Compliance with Ethical Standards No financial support was received for this work.

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