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Case Report

Hypertrophic Dens (Retro Odontoid Pseudotumor) Spinal Compression Treated with C1-C2 Fixation: Case Report and Review

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Abstract

Hypertrophic dens is an inflammatory process at the C1-C2 level leading to mass formation behind the dens. The cervical region, which contains vital nerve fibers, plays a crucial role in maintaining body control; hence, dysfunction at this level is harmful. However, surgical treatment is risky and may greatly impact the quality of life, even with a successful procedure. Consequently, recent studies focus on identifying the most effective decompression technique while minimizing postoperative consequences. We present a case of a 61-year-old male complaining of cervical pain associated with progressive limb weakness. An MRI showed hypertrophic growth of the soft tissue behind the dens, reducing the spinal canal at C1/C2 level. The patient underwent C1/C2 posterior decompression and fixation without anterior mass removal. His postoperative time was uneventful, with important pain relief, sphincter function restored, strength improvement, and regained the ability to walk again. The authors performed a PubMed search adhering to PRISMA guidelines. One hundred four cases that underwent C1-C2 posterior fusion were analyzed, all presenting mass regression, with 43 progressing with mass disappearance after fusion. There were no cases of mass maintaining or increasing in size, allowing us to infer that excess mobility is involved in the physiology of the pannus. The C1 - C2 fixation through fusion eliminates the hypermobility of the C1-C2 segment, and mass regression occurs. Posterior decompression and fixation using the Goel-Harms technique, without mass removal, is the preferred approach for treating retro-odontoid pseudotumor. This technique effectively resolves Atlantoaxial Instability, thereby addressing the inflammatory process and reducing mass formation.

Keywords: Atlanto-axial fixation; Hypertrophic dens; Odontoid process; Pannus; Retro-odontoid pseudotumor; Spine

Introduction

To date, the operative treatment is separated into fusion and decompression. The latter is also subdivided into anterior, composed of ventral decompression, which is performed endoscopically via transoral approach, and posterior procedures; dorsal decompression is widespread and can lead to regression of the retro-odontoid mass without resection. Fusion procedures are more recently advocated based on the pathophysiology of cervical instability, considering that the resolution of such alteration leads to mass reduction without resection. It involves Occipital-cervical Fusion, usually performed in cases where the retro-odontoid mass is compressing the medulla, avoiding the growth of the mass, and C1-C2 fusion, which is a less restrictive procedure. However,

there has yet to be a consensus in the literature regarding which procedure is the standard for hypertrophic dens. Thus, our study aims to provide a better view of whether fusion or decompression is a better treatment option regarding site accessibility and surgeon skills. This study describes a case while presenting a literature review of the physiopathology and treatment of hypertrophic dens, also discussing the concepts involving upper cervical spine stability.

Case Report

A 61-year-old male patient presented with cervical pain associated with progressive limb weakness. He started complaining of cervical pain and difficulty walking, evolving with dropping objects, especially with the right hand, and finally restricted to a wheelchair. Upon physical examination, the patient presented a muscle strength of grade 2 on the right side of the body, including right foot drop and grade 3 force on the left side. There was also global loss of sensation from the neck down, pain and temperature worst on the left side, but vibration worst on the right side. Pathological hyperreflexia, with increased reflex area, was also present, in addition to bilateral positive Hoffmann's sign. Indeed, he also had sphincter dysfunction, reporting difficulty in initiating urination. An MRI scan showed a hypertrophic growth of the soft tissue behind the dens, reducing the spinal canal at C1/C2 level and compressing the cervical spine (Figure 1). The patient was operated on with a C1/C2 posterior decompression and fixation using the Harms technique without any attempt to remove the anterior mass. The patient recovered well, with important pain relief, sphincter function restored and strength improvement, and walking again. The comparison between pre-op and post-op sizes is shown in Figure 2.

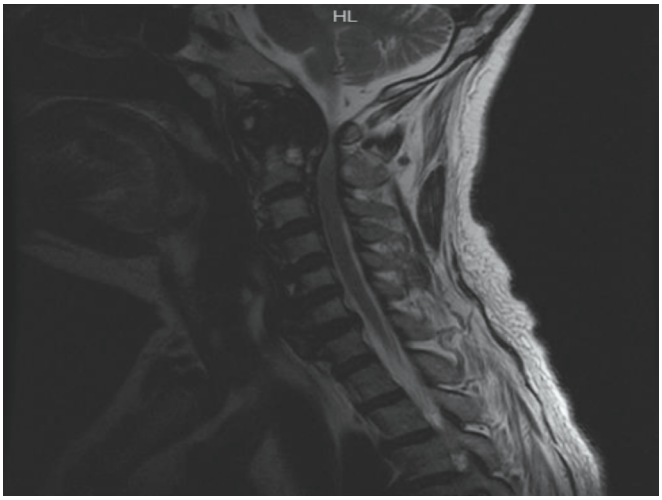

Figure 1: This image shows the ROP in pre-operation.

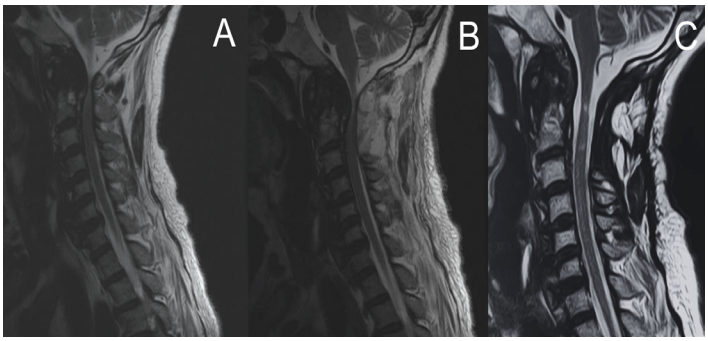

Figure 2: Comparison between ROP size in pre operation (A), immediate post operative (B) and a week of postoperative (C).

Methods

A PubMed search adhering to PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines was performed. The search for publications using the following descriptors: retroodontoid Pseudotumor, retro-odontoid pseudotumor, periodontoid mass, retro odontoid soft mass or odontoid pannus AND treatment, according to database algorithm. The search encompassed all available articles in English and Portuguese, published between 2012 and 2022. The inclusion criteria were: treatment description, post-operative description and non-tumor patient. The following characteristics were used to exclude studies: literature reviews, animal studies, correspondence or letters, articles not available in full text and articles with incomplete information. Titles and abstracts were analysed to screen articles with positive exclusion criteria. From eligible articles the data were obtained, which included: 1) Patients, 2) Age, 3) Sex, 4) Etiology, 5) Surgery, 6) Follow Up, 7) Regression in C1-C2 fusion, 7) Size Pre-op, 8) Size Post Op.

Results

The literature search yielded 99 articles. The title and abstract of 99 articles were screened and based on exclusion criteria, 77 articles were eliminated. After this initial screening, 16 articles were assessed for eligibility, of which 9 were excluded because of the type of treatment not adhering to the inclusion criteria. Works that were duplicated and whose content was not within the context studied were excluded. Therefore, 7 studies (Table 1) were eligible for analysis.

Paper	Patients	Age	Sex	Etiology	Surgery	Follow Up	Regression in C1-C2 fusion	Size Pre-op	Size Post Op
[6]	63	NA	37M;26F	35 OO; 16 DA; 5 RA; 7 NI	Goel	81 m	All cases	vertical 4.5-10.5 mm (7.9 mm); transverse 2-5.2 mm (3.4 mm).	NA
[7]	29 (8 C1-C2 PF)	72.4 (+-9.2)	14M-3F	RA - 4; HEMO-DIALYSIS - 2; OALL - 5; AAS - 15.	C1-C2 PF	54 m (12-96)	All cases	NA	NA
[1]	11 (1 fusion)	68 (48-79)	4M-7F	5 RA; 6 non RA	C1-C2 PF	51.6 m (6-100)	All cases	8.9 ± 3.0 mm	5.3 ± 3.5 mm
[8]	7 (3 fusion)	63.7 (55-76)	5M - 2F	Type 1 (2); Type 2 (1); Type 3 (4).	Goel Harms	55.8 m (10-96)	All cases	NA	NA
[9]	2 (1 fusion)	34 (16-42)	2M	NA	Goel		All cases	NA	NA
[10]	5 (2 fusion)	64.8 (55-76)	3M -2F	NA	Harms Tech-nique	32 m (22-45)	All cases	NA	NA
[11]	26	35 (15-54)	9M - 17F	OO (5), neoplasms of the CVJ (2), basilar invagination os occipitalization of the atlas (16)	C1-C2 PF	20.7 m (6-40)	All cases		

OO (Os Odontoideum); RA (Rheumatoid Arthritis); DA (Degenerative Arthritis); OALL (Ossification of Anterior Longitudinal Ligament); AAS (Atlantoaxial Subluxation); NI (Non Identified); NA (Not Available); M (Male); F (Female); m (months).

Discussion

Types and Classifications of Pannus

Usually, the Retro-Odontoid Pseudotumor (ROP) is classified either as rheumatoid or non-rheumatoid. On the occasion that it is linked with rheumatoid arthritis, leading to inflammation, it is denominated as pannus. In this way, histopathologically, the pannus can be hypervascular, hypovascular, and fibrous [2]. Moreover, retro-odontoid pseudotumor is associated with Atlantoaxial Instability (AAI), Dish Syndrome, ossification of the posterior longitudinal ligament, and trauma. Also, calcium and amyloid depositions may lead to pseudotumor. The AAI has been considered a fundamental part of the pathophysiology of the ROP once the hypermobility of the joint results in the tearing of the ligaments and deposition of inflammatory tissue or abnormal substances in the posterior aspect of the dens, leading to a mass formation [2]. This idea is supported by the fact that mass reduction was observed in cases treated with stabilization alone [6], which means that the stabilization interrupts the deposition process, culminating in mass reduction due to the resolution of the inflammatory mechanism. It's a noticeable historical trend in using smaller surgeries with stabilization instead of larger surgeries for mass resection [12]. This trend results from the development of new methods and parameters using MRI imaging to assess the integrity of ligament structures. Therefore, evaluating AAI is central to the treatment choice and may help differentiate cases that need decompression (mass resection or laminectomy) or stabilization alone.

How Types Influence Treatment Choice

Dividing the disease into two major groups: with and without radiologic instability, allows the surgeon to decide whether to perform stabilization. Cases with clear radiological instability are candidates for stabilization-only procedures. On the other hand, we may consider only anterior or posterior decompression in cases without radiological instability. Still, stabilization alone may not lead to a sufficient mass reduction in cases without clear radiological instability, incurring a need for decompression [13]. One exception for decompression in patients' without instability' is C1-C2 hypermobility, which works as a dynamic instability, despite technical classification [14]. Goel and collaborators [6] reported considerable mass reduction after stabilization. Therefore, the stabilization alone argument is reinforced since even when there is no clear instability, there is significant mass reduction after surgical fixation. Stabilization alone may be a sufficient procedure for most patients, disregarding the need for laminectomy.

Regression of the Pannus After Surgery

The authors analyzed 104 cases (Table 1) operated with C1-C2 posterior fusion, all presenting mass regression, with 43 progressing with the mass's disappearance after fusion. None of the patients did the mass maintain or increase in size. This fact allows us to deduce that excess mobility is involved in the physiology of the pannus growth process, and once this mechanism is extinct, there is a mass regression [6]. Not only did the mass decrease in size, but all patients clinically improved after surgery, being able to deambulate independently, and myelopathic symptoms significantly improved [4,6,11].

C1-C2 Fixation As The Best Treatment Choice

Although there are different techniques, we chose the posterior fixation of C1-C2 using the Goel-Harms technique. However, this procedure requires smaller exposure of the posterior elements of the C1 and C2 vertebrae of the entry angle [15]. Indeed, the direct visualization of the cervical spine is reached more easily with this technique, thus allowing less angulation in the surgical procedure. Moreover, the degree of fixation among Goel-Harms and other techniques is the same; hence since it concedes better visualization, less angulation, and the same degree of fixation [15], the Goel-Harms technique ought to be the best surgical treatment of choice when facing retro odontoid pseudotumor. The good outcome after fixation alone is indicative that C1-C2 by the Goel-Harms technique is a safe and effective treatment for retro odontoid pseudotumor, regardless of its etiology. It's an easier technique than atlantoaxial transarticular fixation (Magerls technique), requiring smaller exposure and with excellent clinical outcomes. Furthermore, this procedure is preferable to perform than other techniques using odontoidectomy, especially for being less invasive, accounting for fewer complications.

Our study has several strengths. First, we presented an anecdotal case of retro odontoid pseudotumor treated with C1-C2 fusion, which is the direction of consensus in the literature, proving the concept of mass regression after fusion. We also reviewed the latest relevant studies involving multiple techniques and compared the results, achieving a comprehensive approach to the current knowledge of this pathology and its treatment. Nevertheless, some weaknesses should be addressed. One inherent weakness of this study is that the reviewed studies lack data regarding the preoperative and postoperative size of hypertrophic dens mass, limiting our analyses of the proportion or mean of mass regression when performing each technique.

Furthermore, another area for improvement is that comparisons are not direct, involving different populations, surgeons, and techniques with no randomization or blinding procedure. However, at this current time, there are no head-to-head randomized trials published, justifying a simpler comparison of techniques as the best evidence to orientate conduct and future research. Thus, further randomized trials are needed to compare methods with more standardized preoperative assessment, evaluation of the postoperative quality of life, impact of surgery on functionality, and mean mass reduction. Furthermore, developing new scales and exams to assess hypermobility may help the understanding of the pathophysiology of mass development.

Conclusions

Atlantoaxial instability is a central aspect of the pathophysiology of the retro-odontoid pseudotumor, as it leads to inflammation and mass deposition in the posterior element of the dens. Therefore, stabilization discontinues the deposition process, culminating in mass reduction due to resolving inflammatory mechanisms. In that matter, stabilization-only procedures are optimal for cases with clear radiological instability. When dealing with retro odontoid pseudotumor with Atlantoaxial Instability, the Goel-Harms technique is the best surgical procedure, as it allows greater visualization, less angulation, and the same degree of fixation[15] while also being a more accessible technique than atlantoaxial transarticular fixation (Magerl's technique), for it demands lesser exposure. Moreover, the decompression and fixation without mass removal are favored in contrast to other approaches which employ odontoidectomy, notably for its less invasive characteristic. In synthesis, posterior decompression, and fixation using the Goel-Harms technique, without attempting to remove the anterior mass, is a better procedure in such cases.

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