

Unilateral Atlantoaxial Osteoarthritis a 10-Patient Retrospective Analysis

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Abstract

Study design: A retrospective study.

Objective: The purpose of this study is to report our experience concerning unilateral atlantoaxial osteoarthritis and to discuss clinical symptoms, diagnostic and therapeutic strategies.

Summary of Background Data: Atlantoaxial osteoarthritis (AAOA) is a rare condition of the cervical spine. Its prevalence is between 5% and 18%. Unilateral atlantoaxial osteoarthritis (UAAOA) is a specific part of AAOA. This painful pathology presents clinical and radiological pathognomonic signs. Little data is available regarding this condition.

Method: A continuous retrospective analysis was performed between 2007 and 2018 in a university hospital. All patients included in the study suffered from cervicalgia and showed signs of UAAOA. Clinical and radiological assessments were conducted during all follow-ups.

Results: Ten patients were included in the study. The average age was

77.1 years. Minimum follow-up was 12 months. The average neck pain according to the visual analogue scale (VAS) was 65. All patients had lateralised cervicalgia and occipital pain associated with limited axial head rotation. All patients had an open-mouth odontoid-view radiograph. A CT scan confirmed the diagnosis of UAAOA. First line of treatment was conservative. If treatment failed, C1-C2 steroid CT-guided injections were performed. In cases of severe UAAOA, or if the above therapeutic methods failed, a surgical treatment was performed. Posterior C1-C2 fusion led to pain relief in all patients.

Conclusion: This study led to a more clearly-defined UAAOA. Clinical symptoms of UAAOA are lateralised cervicalgia and occipital pain, associated with a limited axial rotation of the head. Conservative treatment is the first part of therapeutic management. CT-guided steroid injections had a low impact on painful symptoms and may cause major side effects. Posterior C1-C2 fusion had great results in severe UAAOA or when previous treatments failed.

Keywords: Atlantoaxial; C1-C2 Fusion; Arthrodesis; Cervicalgia; Osteoarthritis; Cervical Spine; Occipital Neuralgia; Range of Motion; Rhizotomy; Steroid Injection; Computerised Tomography; Neck Pain

Keypoints

Unilateral atlantoaxial osteoarthritis is a rare condition of the cervical spine; the clinical symptoms are lateralised cervicalgia and occipital neuralgia, associated with limited axial head rotation; Posterior C1-C2 fusions offer good results where other treatments have failed.

Introduction

Atlantoaxial osteoarthritis is a rare condition. The prevalence of C1-C2 osteoarthritis is 4.8% among the population and can be up to 18.2%. Two main causes are reported: traumatic or degenerative [1]. The majority of patients are female [2]. The main clinical symptoms are cervicalgia, occipital pain and limited cervical range of motion [1,3]. Treatment range is from conservative to surgery [1,2]. Few reports have been produced regarding unilateral atlantoaxial osteoarthritis (UAAOA) [4,5]. The purpose of this study is to report our experience and to discuss the diagnosis and treatment management of UAAOA.

Materials and Methods

A continuous retrospective analysis was conducted in the Spine Unit 1 at the University Hospital of Bordeaux, France. The clinical evaluation included: anamnesis, pain evaluation according the visual analogue scale (VAS; 0 = no pain; 100 = maximum pain), Disability according Neck Disability Index (NDI; 0 = no disability; 50 = maximum disability) and physical examination with evaluation of the cervical range of motion, using a 2-arm goniometer [6,7]. The radiological assessment included standard cervical radiographs and a cervical CT scan. Facet joint osteoarthritis was evaluated according to unilateral grade III or IV atlantoaxial facet joint osteoarthritis, according to Kellgren., *et al.* scale [8-10]. Therapeutic strategy was defined according the clinical and radiological findings. The inclusion criteria were neck pain and unilateral atlantoaxial osteoarthritis. Unilateral atlantoaxial osteoarthritis was defined as grade III or IV on one side of the facet joint and grade 0 or I on the other side [8,9]. Patients with bilateral atlantoaxial osteoarthritis (Grade II or higher on both facet joints), occipitocervical osteoarthritis, atlantodental osteoarthritis or rheumatoid arthritis were ruled out. A single senior surgeon performed the patient evaluation, surgical procedure and follow-up.

Results

From 2007 to 2018, ten patients were treated for UAAOA (5 females and 5 males). The average age of the patients was 77.1 years (67 - 86 years). Minimum follow-up was 12 months.

Clinical history

The mean evolution period of the symptoms was 24.7 months (6 - 96 months). All patients suffered from neck pain. The average neck pain according to the visual analogue scale was 65 (25 - 90). The average Neck Disability Index was 28 (21 - 40). No patient suffered from bilateral cervical pain. The pain was always lateralised, on either the right side (30%) or the left side (70%) of the neck. In seven patients, the pain

ascended unilaterally to the occiput and the parietal skull. One patient had unilateral scapular pain irradiations. Two patients had no pain irradiation. The axial rotation range of motion of the head was evaluated using a universal 2-arm goniometer [6,7]. The ratio of the angles between right and left rotation were measured. The average reduction of the axial cervical range of motion was 58% (50 - 70%).

Radiological diagnosis

Unilateral atlantoaxial osteoarthritis was visualised in all patient standard cervical radiographs and open-mouth cervical radiographs. A cervical CT scan was performed in all the patients. It led to a confirmation of the diagnosis in ten patients and ruled out two patients with atlantodental osteoarthritis (Figures 1a-1c). Six patients had a grade IV and four had a grade III according to the Kellgren., *et al.* scale. Joint osteoarthritis was localised in the left side in seven patients and on the right side in three patients (Table 1). All patients had lateralised cervicalgia on the same side of the atlantoaxial osteoarthritis.



Figure 1a: Coronal CT view of C1-C2 joints showing right unilateral atlantoaxial osteoarthritis.



Figure 1b: Lateral left CT view of C1-C2 joints showing a normal atlantoaxial joint.



Figure 1c: Lateral right CT view of C1-C2 joints showing atlantoaxial osteoarthritis.

	Cl-C2 facet joint osteoarthritis grade according Kellgren., et al.	
	Left	Right
Patient 1	Ι	IV
Patient 2	III	Ι
Patient 3	IV	Ι
Patient 4	IV	Ι
Patient 5	Ι	IV
Patient 6	IV	Ι
Patient 7	Ι	III
Patient 8	III	0
Patient 9	III	Ι
Patient 10	IV	Ι

 Table 1: CT scan evaluation of the C1-C2 facet joint osteoarthritis grade according to Kellgren., et al.

Therapeutic strategy (Figure 2)



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The first treatment option was conservative. Initial therapy for all ten patients consisted of physiotherapy, non-steroidal antiinflammatory drugs (NSAIDs), analgesics and soft collars. After a three-month treatment, no patient experienced relief from cervical pain. Four of the patients who presented severe UAAOA (VAS above 60, a NDI greater than or equal to 25 and grade IV unilateral facet joint osteoarthritis) were treated with posterior fusion (Gallie technique, n = 4 and Magerl technique, n = 2) [11-13]. Six patients, who had

less severe pain, were treated with steroid injections. Two of them experienced total pain relief; two underwent a C1-C2 posterior fusion. One patient, who had more severe occipital pain than cervicalgia, underwent a C2 rhizotomy with no success [14]. One had anaesthetic contraindications and could not have received a posterior fusion.

In all 6 patients who were treated with C1-C2 posterior fusion, full relief from occipital and neck pain was achieved. After 1 year of follow-up, average neck pain according to the VAS was 3 (0 - 10), average NDI was 5 and 100% fusion was achieved.

Discussion

Clinical findings

There are several upper cervical joint lesions. This may result from segmental hypermobility or hypomobility, such as osteoarthritis [1]. The are two main causes of upper cervical osteoarthritis: degenerative or traumatic [1]. In our study, none of the patients had a previous history of trauma. Cervical and subocciptal symptoms as a result of upper cervical conditions are described from the age of 6 until the ninth decade [15]. The estimated prevalence range for cervicalgia associated with occipital irradiations in the population is from 0.4% to 4.1% with a mean age of 42.9 years [15,16]. With aging, these symptoms result from osteoarthritis. In the sixth decade, atlantoaxial osteoarthritis is found in 5.4% of the population and in up to 18.2% in the ninth decade [17,18]. Atlantoaxial osteoarthritis afflicts atlantodental articulation in 17%, the facet joint in 12% and both in 71% [2]. Harata., *et al.* described atlantoaxial osteoarthritis in thirty-one patients but only four patients had isolated facet joint involvement [2].

Females are more affected by cervicogenic headaches (sex ratio 4:1) [1,15]. These data differed from our study where the sex ratio was 1:1. The upper cervical spine conditions as a source of severe neck pain and occipital irradiations are often underestimated [1]. One of the patients suffered for more than 8 years before the diagnosis was made. After reviewing the clinical history of the ten patients, four assessable symptoms were found: lateralised cervicalgia, pain irradiation and reduced upper cervical range of motion. All patients suffered from cervicalgia. This symptom is found in both bilateral and unilateral atlantoaxial osteoarthritis in 100% of the patients [1,4,19]. Pain is lateralised in 12% to 100% of the patients who suffer from atlantoaxial osteoarthritis [2,17,19]. All patients in this study had lateralisation of pain, as did those in the Schaeren., et al. and Fuentes and al. studies [4,5]. All the patients had unilateral and ipsilateral pain to the UAAOA. Schaeren., et al. found the same pain irradiation in all their patients [5]. Chronology of the pain is important. In our study, pain irradiation occurred after cervicalgia. Sjaastad., et al. showed that, in 97% of patients who suffered from cervicogenic headaches, pain irradiation appeared after occipitocervical symptoms [16]. Ehni., et al. reported other various symptoms such as dizziness, nausea, vomiting, phonophobia and photophobia. The final symptom that was present in 100% of the patients was reduced axial rotation of the head. A measurement of axial rotation angles was performed with a 2-arm goniometer. The accuracy of the visual evaluation is 2.9° [7]. The reproducibility of the measurements was guaranteed through the use of the goniometer by the same individual senior surgeon during all follow-ups [6,7]. This reduced range of motion was found in 60 to 100% of the patients who presented bilateral or unilateral atlantoaxial osteoarthritis [1,2,5]. Star, et al. found that, if bilateral atlantoaxial osteoarthritis was present, the loss of axial rotation was greater than 50% [1]. In our study, all patients had over 50% of reduced axial head rotation on the same side of the UAAOA.

UAAOA: diagnostic imaging

The diagnosis of osteoarthritis is performed via diagnostic imaging. Standard radiographs constitute the first-line imaging method in UAAOA. Static and open-mouth radiographs of the cervical spine are the gold standard [20,21]. Open-mouth radiographs of the odontoid allow for a viewing of the atlantoaxial facet joints. UAAOA was defined by a C1- C2 lateral facet joint with grade III or IV osteoarthritis

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and the other lateral facet joint with grade 0 or I, according to the Kellgren., *et al.* osteoarthritis scale [8-10]. These views were not sufficient due to a high false-positive rate [22]. A second imaging method was necessary to assess the diagnosis of UAAOA. Our experience showed that CT is the gold standard for diagnosing UAAOA. It allowed us to accurately view any abnormal morphology of C1-C2 joints (osteophytosis or obliteration of joint space) [23].

Therapeutic management (Figure 3)



Once the diagnosis of UAAOA was made, the first line of treatment was conservative [1,4,5,24]. When analgesics, NSAIDs, relaxing muscle agents, soft collar and physiotherapy failed in achieving pain relief, CT- guided C1-C2 steroid injections were the second line of treatment. Lateral atlantoaxial intra-articular steroid injections may provide short-term analgesia in AAOA [25,26]. Some major side effects have been reported in recent years during CT-guided procedures [27,28]. Chevrot., et al. showed a high rate of injection failure in the event of severe C1-C2 osteoarthritis due to obliteration of joint space [29]. According to these studies, CT-guided steroid injections were performed in six patients who had an NDI less than 25 after conservative treatment. Only two of them obtained cervical pain relief with an NDI less than 4. No major side effects occurred during the procedures. Surgical strategy was preferred when patients had an NDI greater than or equal to 25, or if CT-guided steroid injections failed. In one patient who had more severe occipital pain irradiation than cervicalgia, rhizotomy was performed. No pain relief was achieved using this procedure. When UAAOA is responsible for C2 neuralgia, results of rhizotomy are mediocre as the main aetiology is not treated [14,30,31]. Posterior C1-C2 fusion is indicated when the disability is severe or when other treatments failed. In our cohort, first C1-C2 fusions were performed according Gallie's technique [11]. Magerl's technique has been the treatment of choice over the past five years [12]. Transarticular screw fixation offers greater stability and a higher rate of fusion [32]. The complication rate in C1-C2 posterior fusion is 5% [33]. In our cohort, only one patient had a superficial wound infection after posterior C1-C2 fusion. The average reduction in axial rotation after C1-C2 posterior fusion is 32 degrees on both sides [34,35]. This reduced range of motion was not an issue for the patients. Their lack of mobility was balanced by a pain-free upper cervical spine and by mobility of the lower cervical spine. After posterior fusion, all patients felt immediate relief [3,5,11]. At the one-year followup visit, the patients were asked: "Would you have the same treatment again if you had the same condition?". The six patients who had undergone surgery answered, "Yes, definitely" [36]. Results of C1-C2 posterior fusion in medical literature were graded excellent in 80 to 100% of patients [1,3,11].

Conclusion

UAAOA is a rare condition of the upper cervical spine. This pathology is characterised by unilateral cervicalgia, lateralisation of pain irradiation and reduced axial head rotation. Open-mouth and cervical radiographs are the first-line imaging method. The diagnosis is assessed by CT scan. The first line of treatment is conservative. CT-guided steroid C1-C2 joint injections are the second line of treatment. When other treatments failed or when UAAOA was severe, posterior C1-C2 fusion achieved a high rate of patient satisfaction.

Bibliography

- Star MJ., et al. "Atlantoaxial lateral mass osteoarthritis. A frequently overlooked cause of severe occipitocervical pain". Spine 17.6 (1992): S71-S76.
- 2. Harata S., et al. "Osteoarthritis of the atlanto- axial joint". International Orthopaedics 5.4 (1982): 277-282.
- 3. Grob D., *et al.* "Transarticular screw fixation for osteoarthritis of the atlanto axial segment". *European Spine Journal* 15.3 (2006): 283-291.
- Fuentes S., et al. "[Atlanto-axial lateral mass osteoarthritis. Three case reports and review of the literature]". Neurochirurgie 47.1 (2001): 51-54.
- 5. Schaeren S and Jeanneret B. "Atlantoaxial osteoarthritis: case series and review of the literature". *European Spine Journal* 14.5 (2005): 501-506.
- 6. Williams MA., *et al.* "A systematic review of reliability and validity studies of methods for measuring active and passive cervical range of motion". *Journal of Manipulative and Physiological Therapeutics* 33.2 (2010): 138-155.
- 7. Hirsch BP., et al. "Improving visual estimates of cervical spine range of motion". *American Journal of Orthopedics* 43.11 (2014): E261-E265.
- 8. Côté P., *et al.* "Apophysial joint degeneration, disc degeneration, and sagittal curve of the cervical spine. Can they be measured reliably on radiographs?" *Spine* 22.8 (1997): 859-864.
- 9. Ofiram E., *et al.* "Cervical degenerative index: a new quantitative radiographic scoring system for cervical spondylosis with interobserver and intraobserver reliability testing". *Journal of Orthopaedics and Traumatology* 10.1 (2009): 21-26.
- Kellgren JH and Lawrence JS. "Osteo-arthrosis and disk degeneration in an urban population". *Annals of the Rheumatic Diseases* 17.4 (1958): 388-397.
- 11. Joseph B and Kumar B. "Gallie's fusion for atlantoaxial arthrosis with occipital neuralgia". Spine 19.4 (1994): 454-455.
- Magerl F and Seemann P-S. "Stable Posterior Fusion of the Atlas and Axis by Transarticular Screw Fixation". In: M.D PPK, Weidner P- DD med A (eds) Cerv. Spine I. Springer Vienna (1987): 322-327.
- 13. Vernon H and Mior S. "The Neck Disability Index: a study of reliability and validity". *Journal of Manipulative and Physiological Therapeutics* 14.7 (1991): 409-415.
- 14. Gille O., *et al.* "Surgical treatment of greater occipital neuralgia by neurolysis of the greater occipital nerve and sectioning of the inferior oblique muscle". *Spine* 29.7 (2004): 828-832.

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- 15. Haldeman S and Dagenais S. "Cervicogenic headaches: a critical review". Spine Journal 1.1 (2001): 31-46.
- 16. Sjaastad O and Bakketeig LS. "Prevalence of cervicogenic headache: Vågå study of headache epidemiology". *Acta Neurologica Scandinavica* 117.3 (2008): 173-180.
- 17. Zapletal J and de Valois JC. "Radiologic prevalence of advanced lateral C1-C2 osteoarthritis". Spine 22.21 (1997): 2511-2513.
- Zapletal J., *et al.* "Relationship between atlanto-odontoid osteoarthritis and idiopathic suboccipital neck pain". *Neuroradiology* 38.1 (1996): 62-65.
- Halla JT and Hardin JG. "Atlantoaxial (C1-C2) facet joint osteoarthritis: a distinctive clinical syndrome". Arthritis and Rheumatology 30.5 (1987): 577-582.
- 20. Halla JT and Hardin JG. "The spectrum of atlantoaxial facet joint involvement in rheumatoid arthritis". *Arthritis and Rheumatology* 33.3 (1990): 325-329.
- 21. Nguyen HV., et al. "Rheumatoid arthritis of the cervical spine". Spine Journal 4.3 (2004): 329-334.
- 22. Daniel WW. "False-positive findings in the atlantoaxial facet joints using the open-mouth view of the odontoid". *Arthritis and Rheumatology* 34.1 (1991): 125-126.
- 23. Zapletal J., et al. "Atlanto- odontoid osteoarthritis. Appearance and prevalence at computed tomography". Spine 20.1 (1995): 49-53.
- 24. Ghanayem AJ., *et al.* "Osteoarthrosis of the atlanto-axial joints. Long-term follow-up after treatment with arthrodesis". *Journal of Bone and Joint Surgery-American Volume* 78.9 (1996): 1300-1307.
- 25. Narouze SN., *et al.* "The longitudinal effectiveness of lateral atlantoaxial intra-articular steroid injection in the treatment of cervicogenic headache". *Pain Medicine* 8.2 (2007): 184-188.
- 26. Glémarec J., *et al.* "Intraarticular glucocorticosteroid injection into the lateral atlantoaxial joint under fluoroscopic control. A retrospective comparative study in patients with mechanical and inflammatory disorders". *Joint Bone Spine* 67.1 (2000): 54-61.
- Edlow BL., et al. "Posterior circulation stroke after C1-C2 intraarticular facet steroid injection: evidence for diffuse microvascular injury". Anesthesiology 112.6 (2010): 1532-1535.
- 28. Tang E. "Complications of C1-C2 facet injection". Anesthesiology 114.1 (2011): 222-224.
- 29. Chevrot A., et al. "C1-2 arthrography". Skeletal Radiology 24.6 (1995): 425-429.
- Bovim G., *et al.* "Neurolysis of the greater occipital nerve in cervicogenic headache. A follow up study". *Headache* 32.4 (1992): 175-179.
- Dubuisson D. "Treatment of occipital neuralgia by partial posterior rhizotomy at C1-3". Journal of Neurosurgery 82.4 (1995): 581-586.
- 32. Elliott RE., *et al.* "Outcome comparison of atlantoaxial fusion with transarticular screws and screw-rod constructs: meta-analysis and review of literature". *Journal of Spinal Disorders and Techniques* 27.1 (2014): 11-28.
- 33. Elliott RE., *et al.* "Outcomes of fusion for lateral atlantoaxial osteoarthritis: meta- analysis and review of literature". *World Neurosurgery* 80.6 (2013): e337-e346.

- 34. Dangerfield PH., *et al.* "Rotation of the atlantico-axial joint, investigated using CT and MRI". *Studies in Health Technology and Informatics* 88 (2002): 336-339.
- 35. Roche CJ., et al. "The atlanto-axial joint: physiological range of rotation on MRI and CT". Clinical Radiology 57.2 (2002): 103-108.
- Daltroy LH., *et al.* "The North American spine society lumbar spine outcome assessment Instrument: reliability and validity tests". *Spine* 21.6 (1996): 741-749.

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