Treatment of severe retromastoid pain secondary to C1–2 arthrosis by using cervical fusion

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Object. In this report the authors review their 5-year experience in the diagnosis and management of nine patients with severe retromastoid pain secondary to C1–2 arthrosis. Patients with symptomatic joints unresponsive to nonoperative therapy underwent cervical fusion procedures.

Methods. The mean age of the patients was 71 years, and the onset of prior symptoms ranged from 6 months to 18 years. All patients suffered similar discrete nonneuropathic pain without radicular symptoms ipsilateral to the diseased facet joint. Four patients experienced relief from pain with the use of nonoperative therapy. Five patients continued to experience intractable pain and underwent C1–2 fusion. The follow-up period ranged from 6 to 26 months. The cervical fusion procedure was successful in treating the retromastoid pain in all patients. In patients who underwent surgery, complete relief of pain was demonstrated in four and significantly reduced in the fifth.

Conclusions. The authors have drawn several conclusions. First, C1–2 arthrosis has a rather unique presentation and is a potential cause of upper posterior neck and head pain predominantly in elderly patients. Second, nonoperative management significantly improved the pain in nearly half of their patients and should be the first line of treatment. Last, C1–2 fusion was successful in treating the pain in patients in whom nonoperative management had failed to resolve symptoms.

KEY WORDS • C1–2 arthrosis • pain • fusion • pain relief

Upper posterior cervical and head pain are commonly seen in clinical practice. The term “occipital neuralgia” has been broadly applied to describe many different types of pain in this region. In reality, occipital neuralgia is a symptom rather than a specific disease entity. There are many potential causes of occipital neuralgia. The list of possibilities includes trauma, myofascial pain, Chiari malformation, vascular compression, migraine, infectious and neoplastic lesions, and cervical spondylosis. Despite this formidable list, occipital pain is most often associated with musculoskeletal disease. There can be significant homology and overlap in the clinical presentation of the various causes. Thus, the correct diagnosis can prove to be quite challenging to make. Not infrequently, multiple diagnoses are made, and patients undergo different procedures in the course of their lifetime to treat the chronic pain. This process can be frustrating for both patient and physician alike.

Nonoperative management is the first line of treatment for the majority of patients who complain of occipital neuralgia of musculoskeletal origin, which consists of modalities such as NSAIDs, spinal traction, cervical collar, physical therapy, and local anesthetic blocks. For those patients who do not respond favorably and continue to suffer incapacitating pain, a variety of surgical options are available. Nearly all of the established surgical procedures have a common goal: to effect a reduction in the transmission of pain mediated primarily through the dorsal ramus of the C-2 nerve root. Methods used in the pursuit of this goal include rhizotomy, nerve root decompression, ganglionectomy, neurectomy, and occipital nerve release. These procedures are generally performed to remedy entrapment or injury of the nerve itself. The quality of pain is often described as “shocklike” or “electric,” and radicular complaints such as hypesthesia and paresthesia are common. Presentation with these symptoms suggests a neuropathic source of pain.

In 1984, Ehni and Benner described a group of seven elderly patients with unilateral C1–2 arthrosis who presented with a “deep boring” pain in the upper cervical and suboccipital region but no radicular symptoms. The pain worsened when the head was moved in the direction of the affected side, and ROM was significantly diminished. Injection of the lateral border of the diseased atlantoaxial facet joint with a local anesthetic and a steroid agent provided instant but temporary relief. They concluded that the pain was caused by the arthrotic joint. Conservative therapy failed in two of the patients, who then underwent...
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a C-2 rhizotomy, and a third patient underwent a rhizotomy and a C1–2 fusion. All experienced permanent relief of pain. The risks of a neural ablative procedure such as rhizotomy are well documented in the medical literature.6,17,18 These include cerebrospinal fluid leak, vertigo, neuroma formation, deafferentation syndrome, and loss of scalp sensation. In this paper we describe the successful treatment of severe occipitocervical pain secondary to C1–2 arthrosis by performing fusion of the abnormal motion segment alone.

The C1–2 Facet Joint as a Source of Pain

The lumbar facet joints are a widely accepted source of low-back and referred lower-extremity pain in many patients.8,15 The lumbar facet syndrome is now a well-recognized diagnosis, and an assortment of treatment choices exist. By comparison, the cervical facets have not been as frequently identified as a potential source of head, neck, and upper-extremity pain. This is surprising because, other than soft-tissue and muscular disorders, osteoarthritis of the cervical spine is the most common rheumatological disease.11 In some patients the facet joint is ignored because attention may be predominantly directed to a degenerated intervertebral disc. Nonetheless, it is underappreciated that the cervical facet joints are well equipped to participate in pain pathways.

The cervical facet joints are amply supplied by nociceptive afferent nerve fibers.5,7,11 Facet joint pain is likely transmitted by uncapsulated receptors and free nerve endings that are found in the joint capsule and synovial lining.9,22 Several mechanisms of pain generation can be postulated. Facet degeneration occurs as a part of normal aging and can be accelerated by abnormal stress secondary to trauma, kyphosis, scoliosis, and disc degeneration. Pathological change begins with capsular laxity and synovitis.21 As the articular cartilage dehydrates, the content of proteoglycans and collagen diminishes. The synovium and subchondral bones deteriorate, leading to the formation of osteophytes, hypertrophy, and hypomobility of the joint.10,11,13 Pain can be transmitted at any of the stages of degeneration.

Arthrosis of the C1–2 facet joint is associated with a unique pain syndrome not demonstrated in the middle and lower cervical levels. The C1–2 facet joint differs substantially from subaxial facet joints because it is oriented relatively horizontally, bears a much greater load, and, in many ways, functions more like a subaxial intervertebral disc. This may also explain the mechanical nature of pain seen in these patients. Furthermore, pain related to facet joints below C-3 is generally associated with low cervical, shoulder girdle, and scapular pain.7 From our experience, as well as that of Ehni and Benner,4 pain generated from the C1–2 facet joint is located almost exclusively in the ipsilateral retromastoid region. This location has also been confirmed by a variety of diagnostic and therapeutic studies. Dreyfuss, et al.,3 administered fluoroscopically-guided injections of contrast medium into the lateral atlantooccipital joint and atlantoaxial joints of normal volunteers to determine patterns in facet joint pain. On distention of the atlantoaxial joint, all patients noted similar “dull, heavy” pain laterally at the C1–2 level and in the suboccipital region. The atlantooccipital injections produced mixed results: pain ranged from the vertex of the skull down to the spinous process of C-5.

Clinical Material and Methods

Patient Population

Nine patients with symptomatic C1–2 arthrosis were evaluated at UCLA Medical Center and the University of Tennessee between 1992 and 1997 (Table 1). The mean age was 71 years (range 49–80 years). Eight of the nine patients were women.

Presenting Symptoms

Location. Discrete unilateral retromastoid pain was observed in all patients. The pain was always located on the same side as that of the diseased facet joint. In eight of the patients, the pain was located on the left side and in one on the right side. Two patients noticed intermittent orbital pain as well.

Quality of Pain. The pain was described in nonradicular terms by all patients. Phrases such as “pressure,” “vise-like,” and “deep” were most frequently used. One patient with a C2–T1 syringomyelic cavity described a burning sensation. A description of shocklike or electrical pain was not reported in our patient population.

Associated Factors. In all patients neck motion elicited or worsened the pain, with ipsilateral rotation and/or lateral flexion being the most frequently associated movement (six patients). One patient noticed worsened pain with flexion only. Two patients heard clicking noises during lateral neck rotation. In six patients the pain was improved by sleeping with a rolled-up pillow under the neck.

Symptom Duration. The time between onset of symptoms and neurosurgical evaluation ranged from 6 months to 18 years, with a mean of 4.7 years. None of the patients could clearly identify an initiating event. All patients had been treated by other physicians before undergoing evaluation in our departments.

Medical History

Five patients had a history of osteoarthritis with predominant symptoms in the extremities. Four patients acknowledged previous trauma, including MVAs and falls. No patient had a history of migraine or other form of headache.

Physical Examination

A limitation in cervical ROM was the primary feature encountered during physical examination. One patient did not undergo ROM testing. Lateral flexion toward the affected side was diminished in all patients and was less than 20˚ in all patients except one. Ipsilateral rotation was less than 20˚ in six patients, slightly diminished in one,
and normal in the other. Extension was less than 10° in six patients and normal in two. Flexion was normal in all patients. Pain was frequently elicited with lateral flexion, lateral rotation, and extension.

Palpation over the symptomatic lateral atlantoaxial joint and/or adjacent area provoked pain in all patients. One patient’s head was tilted away from the affected side. One patient also had marked thoracic kyphosis. In all patients motor examination was within normal limits, and there was no sensory deficit in the head and neck region.

Radiological Studies

Unilateral C1–2 arthrosis was discovered in eight patients by performing conventional open-mouth radiography (Fig. 1). In the remaining patient adequate films were unobtainable, and the diagnosis was made by using coronal reconstruction of computerized tomography scans (Fig 2). Magnetic resonance imaging was performed in all patients. The most common finding was middle to lower cervical spine spondylosis, which was demonstrated in all patients. In two of the patients T1-weighted coronal-view MR images demonstrated a low signal intensity at the C1–2 facet joint, suggesting arthritis. Two patients also had a Chiari I malformation with tonsillar descent and cervical syringomyelia, and on MR imaging of the brain a prepontine meningioma was revealed in the patient in Case 5.

Previous Treatment

All patients had undergone prior medical treatment including oral medications such as NSAIDs, acetaminophen, opiate agents, prednisone, carbamazepine, and muscle relaxant agents. Other modalities involved use of cervical collar (four patients), cervical traction (three patients), epidural steroid injection (three patients), acupuncture (one patient), orthodontic braces (one patient), and spinal manipulation by a chiropractor (one patient). One of the patients (Case 7) with a Chiari I malformation had previously undergone decompressive surgery.

Results

Facet Injection

With the aid of fluoroscopy, C1–2 facet joint injections of local anesthetic and steroid agents were administered in six patients. Five patients experienced immediate and complete pain relief, which lasted between 36 hours and 3 days. The facet joint injection was ineffective in one patient (Case 4) who experienced not even transient relief. One patient (Case 7) received bilateral facet injections.

Operative Management

Five patients who continued to suffer intractable pain despite nonoperative management underwent C1–2 fusion procedures (Fig. 3). Gallie fusion was performed in two patients (Cases 2 and 3), Halifax clamps were used in one (Case 1), and transarticular screws were used in the other two (Cases 7 and 9). Autologous posterior iliac crest bone graft was used in all cases.

Patients were maintained in rigid cervical collars for the

<table>
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<th>Case No.</th>
<th>Age (yrs)</th>
<th>Sex</th>
<th>Symptom</th>
<th>Type of Imaging Study</th>
<th>Previous Treatment</th>
<th>Facet Injection Results</th>
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<td>transarticular screws: good</td>
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<td>transarticular screws: excellent</td>
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* AC = acetominophen; AP = acupuncture; BR = orthodontic braces; CC = cervical collar; CP = chiropractic care; CT = cervical traction; CZ = carbamazepine; DJD = degenerative joint disease; ES = epidural steroid; IN = intramuscular narcotics; MR = muscle relaxants; NS = nonsteroidal antiinflammatory drugs; OI = operative intervention; ON = oral narcotics; PR = prednisone; PT = physical therapy.
first 3 postoperative months. The patients were given the
option to wear soft collars once they discontinued wearing
the hard collars. Anteroposterior and lateral plain radio-
graphs revealed bony fusion to have taken place between
3 and 6 months after surgery. No patient developed pseud-
arthritis or infection.

The surgical procedures were successful (excellent or
good results) in all patients. An excellent result was de-
defined as complete resolution of pain without need for pain
medication or additional treatment. A good result was
defined as a greater than 75% reduction in pain, which
was controlled by a minimum amount of oral pain med-
ication. Excellent results were obtained in four patients.
The patients in Cases 2, 3, and 9 were completely pain
free when evaluated at their first postoperative visit. This
pain-free state continued until their follow-up visits at 12,
26, and 15 months, respectively. The patient in Case 1 had
experienced resolution of his pain by his 6-month postop-
erative visit, although he continued to take pain medica-
tion for other conditions. The patient in Case 7 was fol-
lowed for 7 months; her outcome was considered good
because she noticed a significant decrease in her preoper-
ative pain. She was able to discontinue intramuscular narc-
otics and lead a more active lifestyle.

Nonoperative Management

Four patients with C1–2 arthrosis were treated nonop-
eratively. The patient in Case 8 underwent a left-sided
C1–2 facet injection, which temporarily relieved her pain.
She was initially scheduled for fusion surgery, but she
canceled because her pain improved with the full-time use
of a cervical collar. She was seen at follow up 9 months
later, and her pain was controlled with the use of the col-
ar and oral analgesic medication. Only one patient (Case
6) did not experience pain relief from the facet joint injec-
tion. This patient opted against a repeated facet injection,
and she elected to try cervical collar therapy instead. Her
condition improved; however, she died of unrelated caus-
es 1 year later. The patient in Case 4 improved over 10
months of observation and did not receive an injection.
The patient in Case 5 was evaluated at follow up 3 years
postinjection and continued to experience pain that was
controlled with minimum oral analgesic medications.

Illustrative Case

Case 2

This 74-year-old female patient presented to her physi-
cian 2 years earlier with severe left-sided retromastoid
pain that worsened with movement. She also noted a
clicking noise at the base of her skull whenever she turned
her head. Her past medical history was significant for a
MVA and a fall, neither of which was associated with cer-
vical spine injury. An internist prescribed NSAIDs as well
as oral narcotic agents for pain control. She was also eval-
uated by a chiropractor who recommended an exercise
program that yielded minimum relief. She decided to
obtain neurosurgical consultation at UCLA Medical Cen-
ter after having become frustrated with the lack of im-
provement in her condition.

Neurological examination demonstrated normal find-
ings. However, she had marked limitation in extension,
lateral rotation, and lateral flexion of the neck. Severe pain
and crepitation were elicited on lateral rotation. The re-
gion of the left C1–2 facet joint was excruciatingly tender
to palpation. On MR imaging of the cervical spine, a low
signal area corresponding to the left atlantoaxial joint was
demonstrated, as was degenerative joint disease from C-5
to C-7. An open-mouth odontoid radiograph confirmed
the left C1–2 facet arthrosis.

A left-sided C1–2 facet joint injection of local anesthet-
ic and steroid agents provided immediate and complete
relief of pain for 3 days. She subsequently underwent
C1–2 Gallie fusion in which autologous iliac crest bone
graft was used. She underwent follow-up evaluation at 5
weeks, 5 months, and 1 year. Postoperative x-ray films re-
vealed that the wires were in good position and that the
fusion was solid (Fig. 3 center). Her preoperative pain was
completely relieved, and she was able to resume an active
lifestyle.

Discussion

Incidence of C1–2 Arthrosis

In a study of 252 outpatients with either peripheral os-
teoarthritis or degenerative disease of the spine, Halla and
subchondral sclerosis and osteophyte formation. The evaluation of patients with upper cervical and occipital pain continues to be difficult. This is no less true even after entities such as Chiari malformation and tumor are ruled out, and only musculoskeletal causes are suspected. It is widely accepted in the medical literature that pain derived from an arthrotic joint is not generally associated with neuropathic causes of occipital neuralgia. Therefore, it is important to rule out the possibility of nociceptive causes of pain, including degenerative joint disease of C1–2.

Notably, an MRI scan of the cervical spine is not particularly helpful in making the diagnosis of C1–2 arthrosis. In our series, pain was always noted on physical examination of patients with C1–2 arthrosis. Nearly 50% of axial neck rotation occurs at the C1–2 level from the pyramidal decussation.1 Lozano and Vanderlinden17 have found that the quality of pain was a key determinant in the outcome of microsurgical ganglionectomy in the treatment of chronic intractable occipital pain. They concluded that the patients who described their pain in neuropathic terms such as “electric” and “shocklike” following trauma were likely to have a favorable postoperative outcome, whereas those describing their pain as “pressurelike” were not. Pain derived from an arthrotic joint is not generally associated with neural injury; and a nonneuropathic pain quality is expected. All of our patients expressed the quality of their pain in nonneuropathic terms such as “pressure,” “dull,” and “heavy.”

A history of osteoarthritis was noted in five of our nine patients, and cervical spine involvement is common in many forms of arthritis. Osteoarthritis of C1–2 is associated with joint-space narrowing, subchondral sclerosis, and lateral subluxation.11 Four patients had a history of MVA- or fall-related trauma, which was minor in all except one patient (Case 7), who suffered a severe neck “sprain” following two separate MVAs. Her history of cervical trauma combined with a previous C-1 laminectomy perhaps explains her relatively premature C1–2 arthrosis at the age of 49 years.

Restriction in cervical ROM is the most pertinent positive finding on physical examination of patients with C1–2 arthrosis. Nearly 50% of axial neck rotation occurs...
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![Postoperative lateral cervical spine radiographs](image)

**Fig. 3.** Postoperative lateral cervical spine radiographs obtained in three patients in whom different techniques were used. All patients were successfully treated without complications.  Left: Status after C1–2 fusion in which Halifax clamps and wire were used; note the severe degenerative changes in the middle and lower cervical spine.  Center: Status after Gallie fusion.  Right: Status after C1–2 fusion in which transarticular screws were used.

Not surprisingly, lateral rotation was painful and markedly diminished in all but one of the patients, and extension was severely limited as well. This is most likely explained by coexisting middle and lower cervical spondylosis that was ubiquitous in our group. Limited and painful ROM is not restricted to C1–2 arthrosis but is seen in neuropathic causes as well. Several authors have described compression of the C-2 ganglion between the posterior arch of the atlas and superior articular process of the axis during extension and contralateral rotation of the head.\(^2\)\(^{25}\) Flexion of the neck can cause significant pain in cases in which the greater occipital nerve is entrapped within the cervical musculature. Another distinguishing feature of arthrosis demonstrated in two of our patients is palpable or audible crepitation.

Radicular symptoms such as abnormal cutaneous sensation in the head and neck region are not associated with C1–2 arthrosis and were absent in our patient population. In contrast, Dubuisson\(^6\) has noted preoperative sensory deficits in the greater occipital nerve distribution in 10 of the 11 patients in whom he performed partial rhizotomies for intractable occipital neuralgia. Poletti and Sweet\(^21\) have described occipital pain and dysesthesias in the C-2 distribution caused by a hypertrophied atlantoepistrophe ligament in patients with occipitocervical pain.

**Treatment for C1–2 Arthrosis**

The response to intraarticular facet injection of anesthetic and steroid agents is of the utmost importance in determining a treatment plan. Conceivably, the injection itself could be considered the definitive treatment if it provides long-term relief.\(^4\) In five of the six patients with C1–2 arthrosis, total pain relief lasting between 36 hours and 3 days was achieved. The patient in whom no response was elicited may have had a different source of pain. An alternate plausible explanation is a lack of appreciable anesthetic agent entering the joint cavity because of severe degenerative disease. If other structures such as the C-2 nerve, ganglion, or even muscle are inadvertently anesthetized, a false-positive result may occur, and the surgeon could be mistakenly led to believe that the facet joint is the source of pain.

In our series, posterior C1–2 fusion was successful in relieving the intractable pain caused by C1–2 arthrosis. Four patients were cured of their preoperative pain, and the other noted significant improvement. Atlantoaxial arthrodesis was attained without complication by use of Gallie fusion (two patients), Halifax clamps (one patient), and placement of transarticular screws (two patients). Halifax clamps were used in the first operative case and required supplementation with cable to address the rotational/translational forces at this level. Although fusion was achieved in this patient, the construct did not appear to be the most stable. Consequently, the Gallie technique was used in the next two cases and emerged as the preferred method. It was technically the simplest and provided excellent arthrodesis. Transarticular screws were placed in the patient in Case 7 because she had previously undergone a C-1 laminectomy. Overall, the time required for fusion to occur (up to 6 months) was longer than expected and can most likely be explained by the advanced age of our patient population.

In the remaining patients, whereas nonoperative therapy helped considerably, it did not cure their pain. The most effective nonsurgical treatment appeared to be cervical collar therapy. This was not an unexpected observation because pain transmission should be decreased by limiting motion across the pathological joint. This also is the exact philosophy on which the fusion procedure is based. These patients may have had stiffening of the joint to such an extent that their pain was improved.
Conclusions

Arthropsis of C1–2 is a common cause of severe upper posterior cervical and head pain in elderly, predominantly female, patients with a particular retro-mastoid predilection. The syndrome has unique features in terms of the patient medical history, presentation, physical examination, and radiographic studies that are associated with it. Nonoperative therapy, particularly the use of a cervical collar, was helpful in improving the pain in nearly half of our patients. However, the pain status in some patients will not improve, and some patients simply are not satisfied with the continuing need for medication and for an orthosis to aid participation in their daily activities. In these cases, operative management must be considered. Atlantoaxial fusion was successful in all of our patients in whom nonoperative management failed, with complete relief of preoperative pain obtained in four cases. This procedure allowed for the avoidance of the aforementioned risks of rhizotomy as seen in the study by Ehni and Benner, a particularly important consideration when undertaking surgery in an elderly population.

We do not advocate C1–2 fusion as the best method to treat all forms of intractable occipital pain. Patients with signs and symptoms of nerve root entrapment or injury would more likely benefit from other procedures such as nerve root decompression or ganglionectomy, as described by authors elsewhere. However, in a select population with intractable pain and documented C1–2 arthropsis, cervical fusion alone can be a highly successful procedure.

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