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Atlantoaxial osteoarthritis: case series analysis and literature review

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ABSTRACT

Objective. To present rare clinical cases of atlantoaxial osteoarthritis (AAOA), review modern surgical methods and describe our experience in surgical treatment of this pathology.

Material and methods. The study included five patients who underwent surgery for AAOA. All patients presented with persistent drug-resistant pain. Unilateral degenerative damage to the paired atlantoaxial joint was present in four patients, bilateral damage — in 1 case. Posterior atlantoaxial fusion (Gallie technique) was performed in one case. Screw C1—C2 fixation (Harms technique) and arthrodesis of damaged atlantoaxial joint were carried out in four cases.

Results. All patients decreased VAS score of pain by 6—7 points (mean 6.6) after surgery. A follow-up examination confirmed successful fusion in each case. All patients experienced significant limitation of head rotation due to atlantoaxial articulation blockage associated with concomitant spondyloarthrosis.

Conclusion. Drug-resistant pain and/or cervical myelopathy are indications for surgical treatment of AAOA. Posterior fusion is the only effective method for pain relief. Relief of clinical symptoms minimizes lost head rotation. Harms fusion technique enables complete consolidation thanks to stable fusion. Atlantoaxial arthrodesis enhances C1—C2 vertebral block.

Keywords: *atlantoaxial osteoarthritis, atlantoaxial fusion, atlantoaxial arthrodesis.*

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Introduction

Atlantoaxial osteoarthritis (AAOA) is a rare degenerative spinal disease and the leading cause of occipital neuralgia in elderly people. Degeneration typically affects one of the lateral atlantoaxial joints. X-ray signs of AAOA are detected in 5.4% of population in the sixth decade of life. By the ninth decade, they are detected in 18.2% with higher incidence among women [1—4].

There are few Russian and international studies devoted to this pathology. All available studies are case reports or small case series [1—3].

The main symptoms of AAOA are occipital pain and impaired head rotation. In addition to arthrosis of the lateral atlantoaxial joint, pathogenesis of pain syndrome is underpinned by mechanical irritation of the C2 root (Arnold's nerve) and reflex spasm of the oblique muscles of the head with subsequent muscular-tonic and angiodystonic syndromes [1—5]. In delayed stages of disease, joint destruction and head «sinking» lead to odontoid process inclination into the foramen magnum, «corrugation» of posterior longitudinal ligament, hypertrophy and ossification of odontoid ligamentous complex, compression of rostral spinal cord and signs of myelopathy [5, 6].

AAOA is often not recognized at all or verified at delayed stages of disease due to some subjective and objective factors associated with diagnosis of this pathology. Conservative treatment of AAOA, including nonsteroidal anti-inflammatory drugs, physical therapy, massage, shock wave therapy, novocaine myofascial blockades, intra-ar-

ticular steroid injections, and radiofrequency denervation successfully manages symptoms in most patients [1, 3, 4]. Surgery is the only treatment for intractable pain and/or cervical myelopathy [1—4, 6—8].

Material and methods

A retrospective study enrolled 5 men aged 64—75 years who underwent surgery for AAOA between 2013 and 2019. All patients presented with persistent pain resistant to conservative therapy. Anamnesis was more than 5 years. Unilateral degenerative lesion of the paired atlantoaxial joint was present in 4 patients, and bilateral lesion was present in 1 man. We assessed postoperative pain (VAS score), bone consolidation and head rotation. Analysis of bone fusion was based on criteria proposed by Eck K.R. et al. [9]. Head rotation was assessed by measuring the angle between extreme positions of head during lateral rotation. One patient underwent posterior atlantoaxial fusion (Gallie technique), 4 patients — C1—C2 screw fixation (Harms procedure) and arthrodesis of the affected atlantoaxial joint.

A 68-years-old patient has been suffering from degenerative lesions of the atlantoaxial joint for 7 years. There were severe pain (VAS score 8) within skull base radiating to occipital and parietal regions, impaired mobility in cervical spine, crepitus and shooting pain in the back of the head when turning the head to the right. Spondylography revealed degenerative lesion of cervical spine involving atlantoaxial joint **(Figure**, **a)**. CT found arthrosis of the right lateral atlantoaxial joint with severe narrowing of the joint space, subchondral sclerosis and cystic transformation of bone, osteophytes and involvement of the Cruveilhier joint **(Figure**, **b, c)**.

Surgery

The patient underwent surgery in prone position with the head fixed in a Mayfield bracket. We exposed posterior supporting structures of the CI—CII vertebrae and mobilized them to lateral borders through a midline incision. Subperiosteal skeletonization of the lateral mass of the atlas and the C2 vertebral arch root up to the atlantoaxial joint was performed. Unilateral Arnold's nerve was mobilized after stepwise coagulation of venous plexus **(Figure d)**. Then, we inserted screws into the lateral mass of the atlas and C2 body using freehand technique. After C2 root displacement and distraction with a bur, we resected the articulating surfaces of the damaged atlantoaxial joint. Its cavity was filled with autologous bone chips harvested from the ilium **(Figure e)**. Next, careful decortication of posterior arches of the C1—C2 vertebrae was followed by placement of bone chips between them **(Figure f, g)**.

Cervicoccipital pain decreased after surgery (VAS score 1). Regression of symptoms alleviated significant limitation of head rotation caused by atlantoaxial joint fusion and spondyloarthrosis of subaxial spine.

A follow-up examination 24 months after surgery revealed successful posterior fusion and arthrodesis of atlantoaxial joint (Grade I according to K.R. Eck et al.) **(Figure h, i)**.

Results

The minimum follow-up period was 12 months (range 12—78). Symptoms and postoperative outcomes are presented in **Table.** In all patients, VAS scores of pain decreased to 6-7 points (mean 6.6) postoperatively. In all cases, a 12-month follow-up examination confirmed successful fusion (Grade I according to K.R. Eck et al.) with complete integration and remodeling of autologous bone. Preoperatively, all patients had significantly reduced head rotation range (mean 42°), as measured by goniometry, due to underlying disease and concomitant subaxial spondyloarthrosis. The loss of additional head rotation (mean 26°) after atlantoaxial fusion was subjectively offset by improvement of symptoms.

Discussion

Atlantoaxial joint, consisting of paired lateral and medial (Cruveilhier's) atlantoaxial joints, is the most mobile spinal segment. Like all other joints, atlantoaxial joint is susceptible to age-related degeneration. Despite high epidemiological prevalence rates, symptomatic forms of AAOA are much less common. This can likely be explained by latent or mild course of disease in most cases. Furthermore, degenerative damage to atlantoaxial joint is often underestimated as a cause of cervicoccipital pain and impaired head mobility. The last ones are often mistaken for manifestations of subaxial spondyloarthrosis that leads to delayed diagnosis [1, 3, 7].

Unilateral neck pain radiating to occipital and parietal regions, and sometimes to the eye, aggravated by turning the head towards the affected side, and impaired rotation are typical manifestations of disease. Dreyfuss P. et al. reproduced clinical patterns of AAOA in healthy individuals by injecting a contrast agent into the lateral atlantoaxial joint. Thus, the authors experimentally demonstrated a relationship between atlantoaxial joint degeneration and cervicoccipital pain [1—3]. A pathognomonic symptom is also sensation of painful crepitus following neck movements. This forces patients to support their head with hands [1, 2]. Some authors observed conduction disorders in patients with AAOA. Goel A. [6] identified progressive myelopathy in 82% of patients with atlantoaxial joint degeneration.

According to the literature, conservative therapy is effective in most cases. Cervical myelopathy and/or refractory pain syndrome are the only indications for surgical treatment [1—4, 6—8].



Pre- and postoperative examination.
a — sagittal spondylography: severe degeneration of the upper cervical and subaxial spine; b, c — preoperative sagittal (b) and frontal (c) CT scans: arthrosis of the right lateral atlantoaxial joint with joint space narrowing, subchondral sclerosis and cystic bone transformation, osteophytes and involvement of the Cruveilhier joint; d — intraoperative image: the arrow indicates the Arnold's nerve mobilized from venous plexus; e–g — postoperative frontal (e), parasagittal (f) and axial (g) CT scans: atlantoaxial fixation (Harms technique), arrows indicate the resected atlantoaxial joint, autogenous bone chips between the arches of the CI–CII vertebrae and along metal elements; h, i — frontal (h) and parasagittal (i) CT scans in 24 months after surgery: arrows indicate arthrodesis of the lateral atlantoaxial joint and posterior fusion of the CI–CII vertebrae with complete integration and remodeling of autologous bone.

In 1978, Whitesides T.E. and McDonald A.P. first implemented the idea of atlantoaxial joint blockade to eliminate painful irritation. They performed posterior fusion of the C1—C2 vertebrae for AAOA and laid the foundation for surgical treatment of this pathology [1]. Throughout the 20th century, numerous atlantoaxial fusion techniques were developed from the simplest ones (cerclage wire and autologous bone grafts) to widespread techniques described by F. Magerl, A. Goel, and J. Harms. Thanks to stable C1—C2 fixation, these methods allow for earlier bone consolidation [10].

In 1939, Gallie W.E. described the first of many atlantoaxial fusion techniques developed throughout the 20th century. Rotational instability and failed bone fusion were a significant drawback of this technique. According to the literature, their incidence varied from 25% to 80%.

Postoperative outcomes.

No.	Age, years	VAS scores		Bone fusion (K.R. Eck et al.)	Rotation range, °	
		Before surgery	After surgery		Before surgery	After surgery
1	64	7	0	I	60	20
2	68	8	1	I	50	20
3	75	8	2	I	20	10
4	71	7	0	I	40	10
5	66	7	1	I	40	20

To increase rigidity of fixation and improve bone consolidation rates, Brooks A.L. and subsequently other authors modernized this method by modifying the bone graft wire attachment [10].

Transarticular technique, introduced by Magerl F. in 1979, provides the most stable fixation of atlantoaxial joint. However, screw placement trajectory limits this approach in some injuries and diseases, as well as in patients with age-related anatomy changes. This is why we did not use this method in our sample [10].

Fixation of the lateral mass of the atlas and articular process of the C2 vertebra with a plate and screws, developed by A. Goel in 1994, became not widespread due to technical nuances of metal implant placement, including mandatory transection of C2 nerve root to expose the lateral mass of the atlas [7, 10].

In 2001, Harms J. resolved the problem of posterior atlantoaxial fixation. Polyaxial screws and another placement technique without C2 root transection significantly simplified the method. To date, this is the most popular surgical treatment for craniovertebral injuries and diseases [7, 10].

Currently, atlantoaxial fusion is a generally accepted method for AAOA. A meta-analysis found that 92.6% of patients with this pathology experienced regression of symptoms after C1—C2 fusion, regardless of technique [3, 7]. Harms technique is the leading method of atlantoaxial fusion for AAOA thanks to obvious advantages. Currently, only technical nuances of this method, namely advisability of C2 nerve root transection and atlantoaxial arthrodesis, remain the subject of discussion [7].

Importantly, the Arnold's nerve transection for relief of occipital neuralgia caused by AAOA was introduced by Ehni et al. as early as 1984. However, it was not widely used as a standalone procedure. The method developed by Goel A. in 1994 implied routine C2 nerve root transection due to no serious complications [6, 7, 10]. In modern literature, there are also few case reports on decompressive surgeries for degenerative lesions of atlantoaxial joint [7, 8].

Today, some authors recommend combination of atlantoaxial fusion with C2 root transection. They believe that total removal of compressing bony structures anterior to spinal nerve is difficult to achieve without root transection [6, 7]. They consider lost occipital scalp sensitivity to be an acceptable complication. Ganglionectomy is recommended to prevent postoperative neuropathic pain [7, 11]. In our opinion, the need to sacrifice the nerve root for resection of osteophytes seems a questionable argument in favor of this tactic. At the same time, nerve root transection allows for easy hemostasis in case of massive bleeding from venous plexus, as well as complete skeletonization of the lateral mass of the atlas and atlantoaxial joint for arthrodesis. These are more reasonable strengthens in favor of this procedure.

Atlantoaxial joint arthrodesis with special implants or autografts was developed by Goel A. and initially used for basilar invagination. Now, this approach is becoming a part of surgical practice for AAOA [6, 7].

In our opinion, simple placement and primary stability of metal structure make Harms technique the only alternative for atlantoaxial fusion, including patients with AAOA. This technique does not require C2 root transection that is also an undeniable advantage. Arnold's nerve mobilization as a key step of the procedure is achieved through careful subperiosteal skeletonization of the lateral mass of the atlas and C2 vertebral arch root. This allows for free-hand screw implantation, as well as complete decortication and arthrodesis using atlantoaxial joint distraction. Our experience demonstrates these procedures significantly improve postoperative outcomes.

Conclusion

In older patients with appropriate clinical findings, evaluation should focus on diagnosis of degenerative lesions of atlantoaxial joint. Drug-resistant pain and/or cervical myelopathy are indications for surgical treatment. Posterior fusion is the only effective method for manifestations of AAOA. Postoperative outcomes of the Gallie procedure were no different from those of the Harms atlantoaxial fusion. However, high risk of ineffective fusion due to rotational instability of autograft precludes its widespread use. Relief of symptoms alleviates lost head rotation. Indeed, the last one was limited even before surgery. The Harms technique allows for complete consolidation thanks to stable fixation. Atlantoaxial arthrodesis enhances C1—C2 vertebral block.

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COMMENT

The authors present postoperative outcomes in 5 patients with drug-resistant atlantoaxial osteoarthritis. The authors describe in detail clinical presentation, diagnostic methods, surgical technique (Gallie and Harms fusions) and outcomes. The report includes review, historical aspects of surgical treatment and discussion of advisability of C2 root transection. The manuscript fully complies with the journal's scope. Indeed, it addresses a rare but clinically significant disease and contains new data and practical recommendations. The novelty of the study is limited by small (n=5) sample size and retrospective design. However, the authors position their report as a case series and do not claim to make generalizations. Practical significance lies in demonstrating the feasibility of Harms fusion without C2 root transection with favorable outcomes. Technique of Arnold's nerve mobilization and arthrodesis is described in detail. Interestingly, lost head rotation after surgery is subjectively offset by pain relief. The article is of clear interest to neurosurgeons, vertebrologists and neck pain specialists. Despite small sample size, the article expertly organizes literature data, proposes technical solutions (preservation of C2 nerve root, arthrodesis) and describes functional losses and their compensation.

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