### CASE REPORT/*OPIS PRZYPADKU*

# Rheumatoid atlantoaxial instability treated by anterior transarticular C1-C2 fixation. Case report

Reumatoidalna niestabilność C1–C2 leczona przednim zespoleniem stawów szczytowo-obrotowych bocznych. Opis przypadku

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# Abstract

The authors present a case that demonstrates the usefulness of anterior transarticular screw fixation in the treatment of instability due to rheumatoid arthritis. The surgical technique of this infrequently used procedure is presented.

A 35-year-old female patient with medical history significant for rheumatoid arthritis complained of persistent headache and upper neck pain. Examination revealed a decreased range of cervical rotational motion. Magnetic resonance imaging of the cervical spine revealed anterior displacement of C1, destruction of the left lateral atlantoaxial articulation and bony erosion of the C2 vertebral body below the base of the odontoid. Dynamic radiographs showed increased C1-C2 mobility. The authors used a right anterolateral approach to the cervical spine to perform fixation of lateral atlantoaxial articulations by means of titanium cannulated compressive screws. On 4-month follow-up examination, successful C1-C2 stabilization was documented. Despite restriction of neck rotation, the patient reported satisfactory improvement and returned to work.

**Key words:** atlantoaxial instability, cervical spine, spinal fusion, transarticular screws, rheumatoid arthritis.

## Streszczenie

W pracy przedstawiono opis przedniego zespolenia stawów szczytowo-obrotowych bocznych śrubami. Omówiono technikę i zalety tej nieczęsto stosowanej metody operacyjnej. Zespolenie wykonano u 35-letniej kobiety, u której doszło do niestabilności C1-C2 w przebiegu reumatoidalnego zapalenia stawów. Niestabilność powodowała u pacjentki silne bóle karku i głowy. W badaniu klinicznym stwierdzono ograniczenie zakresu skrętu szyi. Badanie metodą rezonansu magnetycznego ujawniło przemieszczenie przednie C1-C2, destrukcję lewego stawu szczytowo-obrotowego bocznego oraz strefę osteolizy w trzonie C2 na granicy trzonu i podstawy zęba kręgu obrotowego. Rentgenowskie zdjęcia czynnościowe w projekcji bocznej wykazały nadmierną ruchomość pomiędzy kręgiem szczytowym i obrotowym. Operację wykonano z dostępu przednio-bocznego po prawej stronie szyi. Do zespolenia stawów szczytowo-obrotowych bocznych użyto tytanowych kompresyjnych śrub tunelowych. Uzyskano częściowe zmniejszenie przedniego przemieszczenia kręgu C1 względem C2. W badaniu klinicznym po 4 miesiącach wykazano ograniczenie skrętu szyi podobne do przedoperacyjnego, radiologicznie potwierdzono stabilność zespolenia C1-C2. Pacjentka zgłasza zadowalającą poprawę, okresowo odczuwała niewielkie bóle karku, powróciła do pracy.

**Słowa kluczowe:** niestabilność szczytowo-obrotowa, kręgosłup szyjny, stabilizacja kręgosłupa, śruby przezstawowe, reuma-toidalne zapalenie stawów.

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# Introduction

Rheumatoid arthritis (RA) is an autoimmune disease that commonly affects the occipitocervical junction [1-3]. The rheumatoid process causes formation of granulation tissue also called rheumatoid pannus. Additionally, destruction of cartilages, ligaments and bony erosions develops, leading to atlantoaxial subluxation or to cranial settling [1-3]. Clinical symptoms of atlantoaxial instability include occipital headaches, occipital neuralgia, vertebrobasilar insufficiency, dysfunction of cranial nerves (fifth, ninth, tenth or eleventh) and cervical myelopathy [1-3]. The severity of myelopathy was systematized by the classification proposed by Ranawat [2,3]. Surgical treatment is focused on pain relief, spinal alignment, decompression and elimination of the instability [1,2]. To achieve C1-C2 stabilization, the surgeon may choose between various methods performed by a posterior approach - sublaminar or interspinous wires, hook-claw constructs, transarticular screw fixation, C1-C2 screw-rod constructs - or even may perform longer occipitocervical fusion, if necessary [1,2,4].

In 1971, Barbour described anterior transarticular screw fixation (ATS) of lateral atlantoaxial articulations (Fig. 1) [5-7]. Nowadays, this technique is used to stabilize odontoid fractures, chronically non-united odontoid fractures, Jefferson fractures, metastasis to lateral mass of C2, and as a salvage method in cases of posterior C1-C2 implant failure or odontoid screw failure [5,6,8,9].

We successfully used ATS to treat partially reducible atlantoaxial subluxation caused by RA. To the best of the authors' knowledge, this is the first report of this fusion method in Polish literature.

#### Case report

A 35-year-old woman was admitted to our department in February 2012 for treatment of atlantoaxial instability diagnosed in an external institution. She had been suffering from severe headaches and suboccipital neck pain of 18-month duration. Symptoms occurred at night, provoking sleep disorder, and intensified during the patient's daily social and professional activities (office work). Her medical history included RA diagnosed 25 years earlier (at the age of 10). On neurolo gical examination she demonstrated pain provoked by head motion as well as decreased range of lateral neck rotation  $-45^{\circ}$  to each side. The symptoms were assessed as Ranawat class I. Subjective perception of pain using a visual analogue scale (VAS) was scored as 8. Magnetic resonance imaging (MRI) revealed anterior displacement of C1, destruction of left lateral atlantoaxial articulation and bony erosion of the C2 vertebral body just below the base of the odontoid process; there was enough space available for the spinal cord, and the trajectories of vertebral arteries at C1 and C2 levels were normal (Fig. 2). Dynamic lateral radiographs of the cervical spine showed C1-C2 instability with the anterior atlantodental interval (AADI) varying from 4 mm in extension to 8 mm in flexion (Fig. 3).

The patient consented to surgery by an anterior approach, taking into account less invasive opening. The procedure was carried out in the patient's supine position. We used biplanar open-mouth and lateral fluoroscopy. Delicate extension of the neck and head allowed us to obtain reduction of subluxation. A longitudinal incision was made along the medial border of the right sternocleidomastoid muscle at the C3-C4 level. Typical blunt preparation of soft tissues between the carotid sheath and trachea with the oesophagus allowed us to reach the C2-C3 intervertebral disc. The anterior layer of the C2-C3 annulus was removed to prepare the entry points for K-wires under the pinafore of the C-2 body. The left K-wire was inserted near the midline, inclined 30° laterally and advanced through the left atlantoaxial articulation to reach the left C1 lateral mass. The right K-wire was inserted about 6 mm from the midline in order to omit bony erosion, inclined

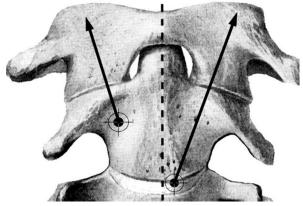


Fig. 1. The principle of anterior transarticular screw fixation with technical variations of screw entry points. Left: The screw entry point is situated 5 mm from the lateral border of the base of the odontoid process at the small groove beneath the superior articular facet of C2. Right: Entry point is situated 5-10 mm from the midline at the inferior border of C2. Screws are directed cranially through the lateral atlantoaxial articulations and engaged in lateral masses of C1

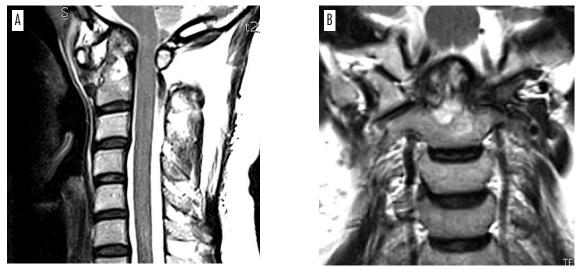


Fig. 2. Preoperative T2-weighted magnetic resonance imaging in sagittal (A) and coronal (B) plane, showing anterior subluxation of C1 over C2, destruction of the left lateral atlantoaxial articulation and bony erosion of the base of the odontoid process

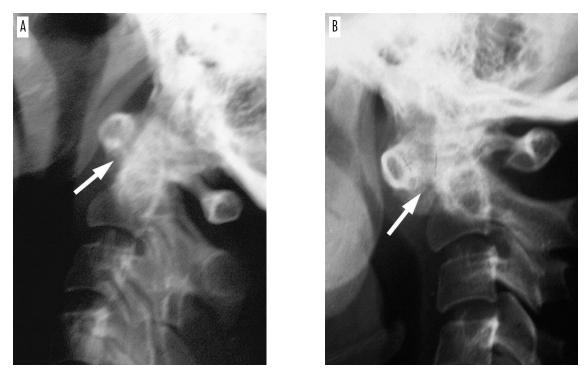


Fig. 3. Preoperative lateral radiographs showing increased mobility between C1 and C2. Anterior atlantodental interval (arrow) measured in extension (A) was 4 mm, whereas in flexion (B) it was 8 mm

 $15^{\circ}$  laterally and advanced into the C1 right lateral mass. On the lateral projection both K-wires were directed towards the superior articular facets of C1. Each wire served as a guide for a  $34 \times 3.5$  mm cannulated self-tapping compression screw (Synthes). After insertion of both screws, K-wires were removed, and the wound was closed with drainage. Intraoperative blood loss was

minimal. The neck was immobilized in a rigid collar for 6 weeks. Computed tomography (CT) after the procedure showed partial reduction of subluxation with AADI = 4 mm. Both screws traversed atlantoaxial articulations and were engaged in lateral masses of C1 (Fig. 4). Four weeks after the surgery the patient resumed professional activity. On 4-month follow-up examination restriction of neck rotation persisted and atlantoaxial stability was documented by dynamic lateral radiographs showing AADI = 4 mm in flexion as well as in extension (Fig. 5). Despite occasional moderate neck pain (VAS = 4), the patient reported satisfactory improvement.

## Discussion

In current surgical practice there are several posterior techniques of C1-C2 fixation, considered standard procedures for atlantoaxial fusion. The most important are sublaminar or interspinous wires (Gallie, 1939; Brooks and Jenkins, 1978), hook-claw constructs, transarticular screw fixation (Magerl and Seeman, 1986), and C1-C2 screw-rod constructs (Harms and Melcher, 2001; Wright, 2004) [1,4]. Each of the above-mentioned methods, combined with bone graft, may provide 100% fusion rates [4,5,8,11]. However, the wiring procedures may be complicated by nonunion, due to graft displacement and wire loosening [12]. On the other hand, posterior transarticular screw fixation is technically demanding: in up to 22% of individuals anomalous trajectory of the vertebral artery or other anatomical factors make screw placement impossible. C1-C2 subluxation must be reduced prior to screw placement to ensure proper screw trajectory, in case in patients with thoracic kyphosis access to the screw entry point in C-2 is difficult [5,10-12]. This method was complicated by injury of the vertebral artery (risk -2.2% per screw), cranial nerve XII, dural sac and spinal cord; screw breakage was also reported [5,8,10-12]. Screw-rod technique is considered to be safer, but the possibility of vascular or neural injury should be taken into account [1,4,12]. The posterior approach, however, possesses certain disadvantages: deep wound, damage to posterior neck muscles and bleeding, wide surgical exposure to visualize screw entry points in C1 and C2 [5,6,8,12]. These disadvantages justify the use of ATS in selected cases [5-7]. This technique is feasible by an anterolateral or retropharyngeal approach to the C1-C2 complex; additionally an endoscopic access was used [5-7,9,11,12].

The literature describes the following advantages of ATS: (1) surgical access through cervical soft tissues without muscular injury, (2) reduction of anterior dislocation of C1 by patient's head and neck extension in prone position, (3) more predictable screw trajectory against vertebral artery decreases risk of its injury, (4) surgeon may perform one-stage odontoidectomy, (5) stabilization may be completed by decortication of atlantoaxial artic-

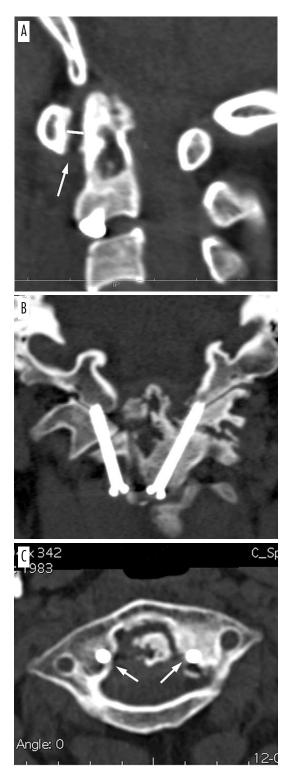


Fig. 4. Postoperative computed tomography (CT). (A) CT reconstruction image in sagittal plane in neutral position showing anterior atlantodental interval = 4 mm (arrow), (B) CT reconstruction image through trajectory of both screws, (C) CT axial scan at C1 level showing screws in lateral masses of the C1 vertebra (arrows)

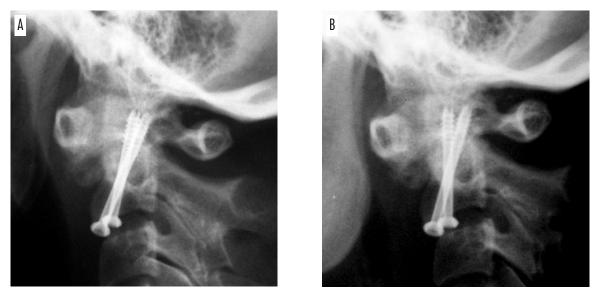


Fig. 5. Lateral radiographs taken 4 months after surgery in extension (A) and flexion (B), demonstrating the absence of mobility between C1 and C2

ulations and bone graft [6,8,9,11,12]. Biomechanical parameters of ATS are similar to posterior screw techniques [10-12]. Clinical efficacy of ATS (with no complications) has been reported by numerous authors [5,6,8,9]. Wang reported transitory dysphagia in 2 of 7 patients operated on by endoscopic access [8].

We have described the use of ATS in the treatment of instability due to RA. The patient's symptoms were typical for atlantoaxial instability. Given the absence of spinal cord involvement, the goal of surgical treatment comprised spinal alignment and elimination of pain, without the necessity of posterior decompression. According to suggestions formulated by Li [6], who indicated ATS in rheumatoid subluxation, we found that this procedure could be safely performed, offering reduction of C1-C2 displacement and immediate stability. Preoperative MRI showed normal passage of vertebral arteries at C1 and C2 levels, eliminating the necessity of further diagnosis by CT angiography. In terms of MRI, transarticular screw placement was not contraindicated. The patient's supine positioning allowed us to extend her head and neck to obtain reduction of subluxation under fluoroscopic control. AADI measured on the postoperative CT reconstruction image in a neutral position was 4 mm long, whereas normal AADI for adults is 0-3 mm [1]. On 4-month follow-up examination, lateral radiographs in flexion and extension confirmed the absence of mobility between C1 and C2.

In conclusion, our case proved the usefulness of ATS in the treatment of rheumatoid atlantoaxial instability. The patient's supine position allowed reduction of anterior subluxation between C1 and C2 with a good clinical result.

# Disclosure

The authors report no conflict of interest.

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#### COMMENT/*KOMENTARZ*

#### A comment to the paper Rheumatoid atlantoaxial instability treated by anterior transarticular C1-C2 fixaton. Case report by Zapałowicz K. et al.

The authors report on the case of transarticular screw fixation performed via anterior approach. These novel and interesting method already reported in the literature has the advantage of being less invasive when compared to the standard transarticular instrumentation known as Magerl technique which is performed through the posterior approach. The Magerl technique requires dissection of paraspinal muscles extended far laterally to expose C1/C2 joint and entry points for screws. This approach is devastating for paraspinal musculature resulting in its denervation and extensive scar. Magerl instrumentation of C1/C2 joint usually requires separate skin incisions distally to main surgical wound to achieve the correct angle and trajectory of screw insertions. In contrast to the posterior approach, the anterior approach spares paraspinal muscles, is straightforward, quicker, more familiar to spine surgeons, and more elegant. However, it has the disadvantage of providing limited possibilities of fusion with bone graft. Some authors who use the anterior C1/C2 transarticular instru-

mentation, do fusion by packing scarified fissure of the atlanto-axial joint with bone chips. Unlike the anterior approach, the posterior approach provides excellent possibilities of fusion with bone graft either interposed between C1 and C2 lamina and secured with sublaminar cables or packed within the fissure of atlanto-axial joint. The authors of this article do not mention if they performed any fusion (bone grafting) and therefore one must conclude that the case was instrumented without bone graft. Although one must appreciate the authors' surgical excellent skills and great experience required in this type of instrumentation, a concern arises about the long-term stability and risk of construct failure in the absence of healed fusion. The authors may only count on inadvertent fusion due to scarification of the joint fissure during instrumentation with screws. I hope the authors with their growing experience in this technique will present results of anterior transarticular fixation combined with some sort of fusion.

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