

NEUROSURGICAL TREATMENT OF UNSTABLE CERVICAL SPINE DUE TO RHEUMATOID ARTHRITIS: CASE SERIES

Tratamiento neuroquirúrgico de la columna cervical inestable debido a artritis reumatoidea: Serie de casos

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ABSTRACT

Introduction: Rheumatoid arthritis (RA) is a systemic disease characterized by synovitis, which causes damage to the ligaments and joints. The cervical region is the region of the spine most affected, with neck pain the most frequent symptom. The three forms of presentation are atlantoaxial subluxation, cranial settlement, and subaxial subluxation. The clinical evolution of patients treated conservatively is poor, with surgery being a key element to prevent the progression of neurological deterioration.

Methods: A retrospective study of 10 patients with RA operated between 2015 and 2019 was carried out. Ranawat criteria were used for clinical classification and imaging studies were performed to confirm cervical instability. Anterior and posterior cervical arthrodesis techniques were used. Control after surgery was performed by tomography and the Ranawat score was determined in the outpatient control.

Results: In the period 2015 and 2019, 10 patients with RA who presented symptoms of cervical instability were operated on. All patients were female, aged between 52 and 73 years. The most frequent symptom was neck pain. Most of the patients presented inflammatory markers (ESR, CRP) in high values. The most frequent cervical involvement was atlantoaxial instability. The most common surgical technique used was posterior C1-C2 arthrodesis via the inter-articular approach. Ranawat's classification improved in 90% of patients after surgery. Complications were surgical site infections and rupture of occipitocervical fixation bars, which were adequately resolved.

Conclusions: Surgery for patients with vertebral instability due to RA should be aimed at treating intractable pain and stopping the progression of cervical instability, with the aim of promoting neurological recovery and reducing mortality.

Keywords: Spine, Arthritis, Rheumatoid, Joint Instability, Synovitis, Neck Pain, Arthrodesis. (source: MeSH NLM)

RESUMEN

Introducción: La artritis reumatoide (AR) es una enfermedad sistémica caracterizada por sinovitis, que genera lesión de los ligamentos y de las articulaciones. La región cervical es la región de la columna vertebral más afectada, siendo la cervicalgia el síntoma más frecuente. Las tres formas de presentación son la subluxación atlantoaxial, el asentamiento craneal y la subluxación subaxial. La evolución clínica de los pacientes tratados en forma conservadora es pobre, siendo la cirugía un elemento clave para evitar la progresión del deterioro neurológico.

Métodos: Se realizó un estudio retrospectivo de 10 pacientes con AR operados entre el 2015 y 2019. Se utilizó los criterios de Ranawat para la clasificación clínica y se realizó estudios de imágenes para confirmar la inestabilidad cervical. Las técnicas de artrodesis cervical anterior y posterior fueron utilizadas. El control luego de la cirugía se realizó mediante tomografía y se determinó la puntuación de Ranawat en el control ambulatorio.

Resultados: En el periodo 2015 y 2019 fueron intervenidos quirúrgicamente 10 pacientes con AR que presentaron síntomas de inestabilidad cervical. Todos los pacientes fueron del sexo femenino con edades comprendidas entre los 52 y 73 años. El síntoma más frecuente fue la cervicalgia. La mayoría de los pacientes presentaron marcadores inflamatorios (VSG, PCR) en valores elevados. La afectación cervical más frecuente fue la inestabilidad atlantoaxial. La técnica quirúrgica más frecuente fue la artrodesis posterior C1-C2 vía interarticular. La clasificación de Ranawat mejoró en el 90% de pacientes luego de la cirugía. Las complicaciones fueron infecciones del sitio operatorio y la ruptura de una barra de fijación occipitocervical, las cuales fueron resueltas en forma adecuada.

Conclusiones: La cirugía de los pacientes con inestabilidad vertebral por AR debe dirigirse a tratar el dolor incoercible y detener la progresión de la inestabilidad cervical, con el fin de promover la recuperación neurológica y disminuir la mortalidad.

Palabras clave: Columna Vertebral, Artritis Reumatoidea, Sinovitis, Dolor cervical, Artrodesis. (Fuente: DeCS Bireme)

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Rheumatoid arthritis (RA), a chronic, progressive, systemic inflammatory disease, is characterized by synovial hyperplasia and cartilage destruction, causing joint deformities and disability over time. RA affects approximately 1 to 2% of the world's adult population and is most common among women between the ages of 40 and 50.¹⁻⁴ Although the most prominent effects of RA are seen in small peripheral joints, the second most affected region is the cervical spine.^{3,4,6,7}

Recent estimates suggest that more than 80% of RA patients have radiographic involvement of the cervical spine, some as early as 2 years after the initial diagnosis of RA.^{2,3,6,8} Chronic inflammation of the cervical spine initially leads to proliferation of fibrovascular tissue and pannus formation, resulting in bone erosion and ligament laxity. This cascade can lead to instability of the cervical spine in the form of atlantoaxial instability (AAI), cranial settlement (CS) and subaxial subluxation (SAS) or a combination of the three.^{2,3,4,6,8} In addition, RA can cause inflammatory discitis and traumatic odontoid erosion or fracture.⁸

The most common clinical manifestations of cervical disease in RA is neck pain, specifically pain at the cranio-cervical junction.^{2,8,13} Involvement of the cervical spine is of particular importance because, if not treated in time, it can lead to greater neurological compromise and consequently worsen quality of life.^{2,6,9}

Although the medical treatment of RA has been improved

with the use of new medications, which have decreased the incidence of cervical involvement, these have not been able to slow its progression once it begins.^{3,6,8} When the involvement of the cervical spine becomes symptomatic, surgical stabilization should be considered as it has been shown to delay and sometimes prevent disease progression by improving functional status in certain patients.^{2,3,4,6,8,17}

In this study, we report 10 RA cases that were treated in our hospital by fusion surgery, with techniques that include cervical arthrodesis and occipitocervical fixation.

METHODS

Our team carried out a retrospective study of patients with RA between 2015 and 2019 who presented involvement of the cervical spine and who underwent surgery. These patients are referred to our hospital from different parts of Peru. The patients were subject to surgery after evaluation by the Medical Board of the Neurotrauma and Spine Service.

The criteria used to classify the type of cervical instability that the patients presented was based on the imaging study that included X-rays, tomography, and magnetic resonance imaging. The classification of the degree of neurological involvement was based on the Ranawat scale for rheumatic cervical myelopathy both pre- and post-surgery. All patients were operated on by a surgical team from the Neurotrauma and Spine Service. Immediate tomographic control studies were performed on all patients after surgery.

Table 1. Summary of cases of rheumatoid arthritis with cervical instability surgically treated in the Neurotrauma and Spine Service of the Guillermo Almenara National Hospital, Lima - Peru, 2015-2019.

Patients	Age	Sex	Clinical	Diagnosis	Surgery	Ranawat pre surgery	Ranawat post surgery
Case 1	66	F	Cervicalgia	AAI + SAS	Posterior arthrodesis C1-C7	II	II
Case 2	57	F	Cervicalgia + Brachial paresis (right)	AAI	Posterior arthrodesis C0-C3	II	I
Case 3	67	F	Occipitocervical pain Quadriparesis	AAI	Posterior arthrodesis C0-C4	IIIA	II
Case 4	60	F	Cervicalgia	SAS	Anterior and posterior arthrodesis, (2 surgeries)	II	I
Case 5	52	F	Cervicalgia + Quadriparesis	AAI + CS	Occipitocervical arthrodesis	IIIA	I
Case 6	56	F	Cervicalgia + Quadriparesis	AAI	Posterior arthrodesis C1-C2	IIIA	II
Case 7	73	F	Cervicalgia	SAS	Previous arthrodesis	II	II
Case 8	61	F	Cervicalgia+ Hemiparesis	AAI	Posterior arthrodesis C1-C2	IIIA	II
Case 9	51	F	Cervicalgia Quadriparesis	AAI	Posterior arthrodesis C1-C2	IIIA	II
Case 10	60	F	Cervicalgia	AAI	Posterior arthrodesis C1-C2	II	II

Source: Data base from Guillermo Almenara National Hospital, Neurotrauma and Spine Service. *(AAI) Atlantoaxial instability, (CS) cranial settlement, (SAS) subaxial subluxation

Cervical stabilization consisted of anterior or posterior arthrodesis depending on the previous surgical indication. The anterior arthrodesis performed in cases of subaxial subluxation were performed with anterior plates and cylindrical titanium meshes. Techniques that included C1-C2 arthrodesis and occipitocervical fixation using a rod and screw system were used for posterior arthrodesis.

In the postoperative period, patients were kept with a soft or semi-rigid collar depending on the degree of stability achieved. All data were collected from the medical records and epicrisis of the patients during hospitalization and outpatient control.

RESULTS

We carried out a retrospective study of patients with rheumatoid arthritis with cervical instability treated between 2015 and 2019. Ten patients with RA with symptoms of cervical instability were operated on, after a case discussion at the Neurotrauma and Spine medical board. All patients were female, aged between 52 and 73 years, mean 60.3 (*Table 1*).

The most frequent symptom in our patients was neck pain.



Fig 1. (Case 1): 66-year-old woman, with a history of rheumatoid arthritis (RF +) with osteoporosis, right foot arthroplasty and cholecystectomy for 30 years. She received treatment with prednisone, methotrexate, leflunomide, sulfasalazine. She had posterior neck pain and laboratory tests showed elevated CRP and ESR. (A, B, C) Functional cervical radiography demonstrated atlantoaxial instability and subaxial subluxation. (D, E, F) Cervical tomography and magnetic resonance imaging showed C1-C2 subluxation and a moderate narrow canal. (G, H, I) The surgery performed was C1-C7 posterior arthrodesis with good clinical evolution, maintaining grade II on the Ranawat scale.

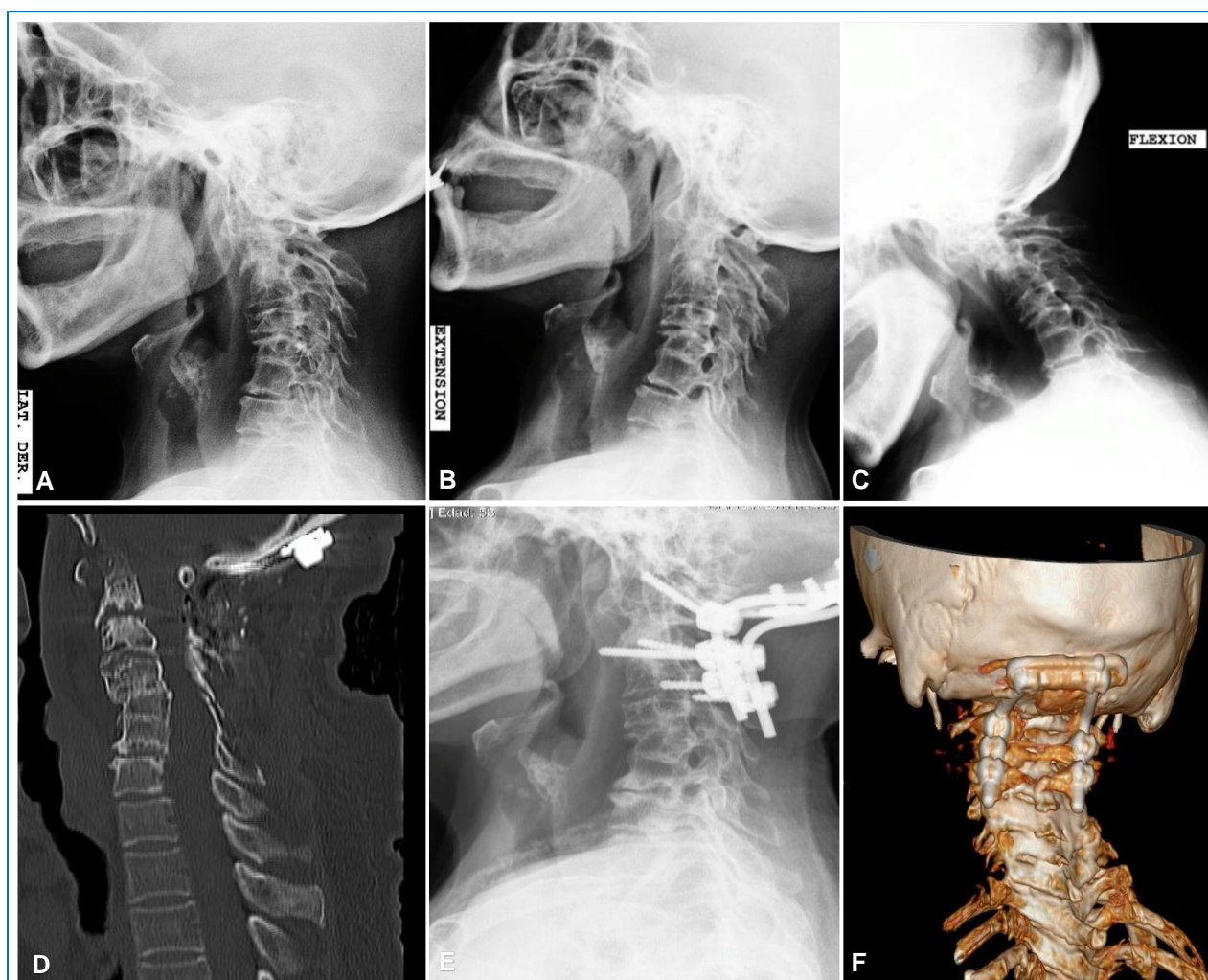


Fig 2. (Case 2): 57-year-old woman with a 30-year history of rheumatoid arthritis, treated pulmonary TB, knee surgery and thoracic surgery. She received therapy with prednisone and methotrexate. The patient presented symptoms of posterior neck pain, brachial paresthesias in the right upper limb. Laboratory tests showed elevated CRP and ESR. **(A, B, C)** Functional cervical radiography demonstrated the presence of atlantoaxial instability. **(D, E, F)** The surgery performed was C0-C3 posterior arthrodesis with good clinical evolution on the Ranawat scale, going from grade II to grade I.

They all had an average illness period of more than 10 years. In most of our patients, inflammatory markers (ESR, CRP) were found at high values. The most frequent cervical involvement was atlantoaxial instability.

The surgical techniques used were both anterior and posterior cervical arthrodesis, the C1-C2 posterior arthrodesis being the inter-articular approach the most frequently performed procedure. A halo-traction system was not used in any case. The Ranawat classification improved in 90% of the patients after surgery, the majority had a Ranawat II before surgery.

Regarding postoperative complications, there were recurrent surgical site infections and rupture of one of the occipitocervical fixation bars, which were properly resolved.

At follow-up, 3 months after the operation, improvement was obtained in all patients and there were no signs of spinal cord compression. There were no deaths because of arthrodesis surgery and cervical stabilization.

DISCUSSION

The involvement of the cervical spine in patients with rheumatoid arthritis has been published in multiple research papers, so early management is particularly important to avoid the progression of neurological compromise.

Most of the affected patients are female,^{1,2,3,4,16} as was shown in our series where 100% were women. The age of presentation ranges from 50 years with a history of having been diagnosed with RA on average 15 years before,^{1,2,3,4,16} these data coincide in a certain way with our work that the mean age was 60 years and most were 30 years old after being diagnosed with RA.

Compared to other series, most of our patients also received therapy with corticosteroids, disease-modifying antirheumatic drugs (DMARDs), and biological agents, 16 which were able to delay the onset of cervical disease but not its progression, as shown by others series,^{3,6,8,16} in contrast to its success in treating peripheral joint manifestations.

The clinical manifestations of cervical disease in RA are varied and difficult to interpret in the context of joint arthropathy, muscle wasting, decreased range of motion, compressive neuropathy, and poor functional status in many

patients.^{3,17} It is important to note that the incidence of asymptomatic cervical involvement in RA is high with 33% to 50% reports of patients having no symptoms, and therefore greater awareness of the frequency of cervical

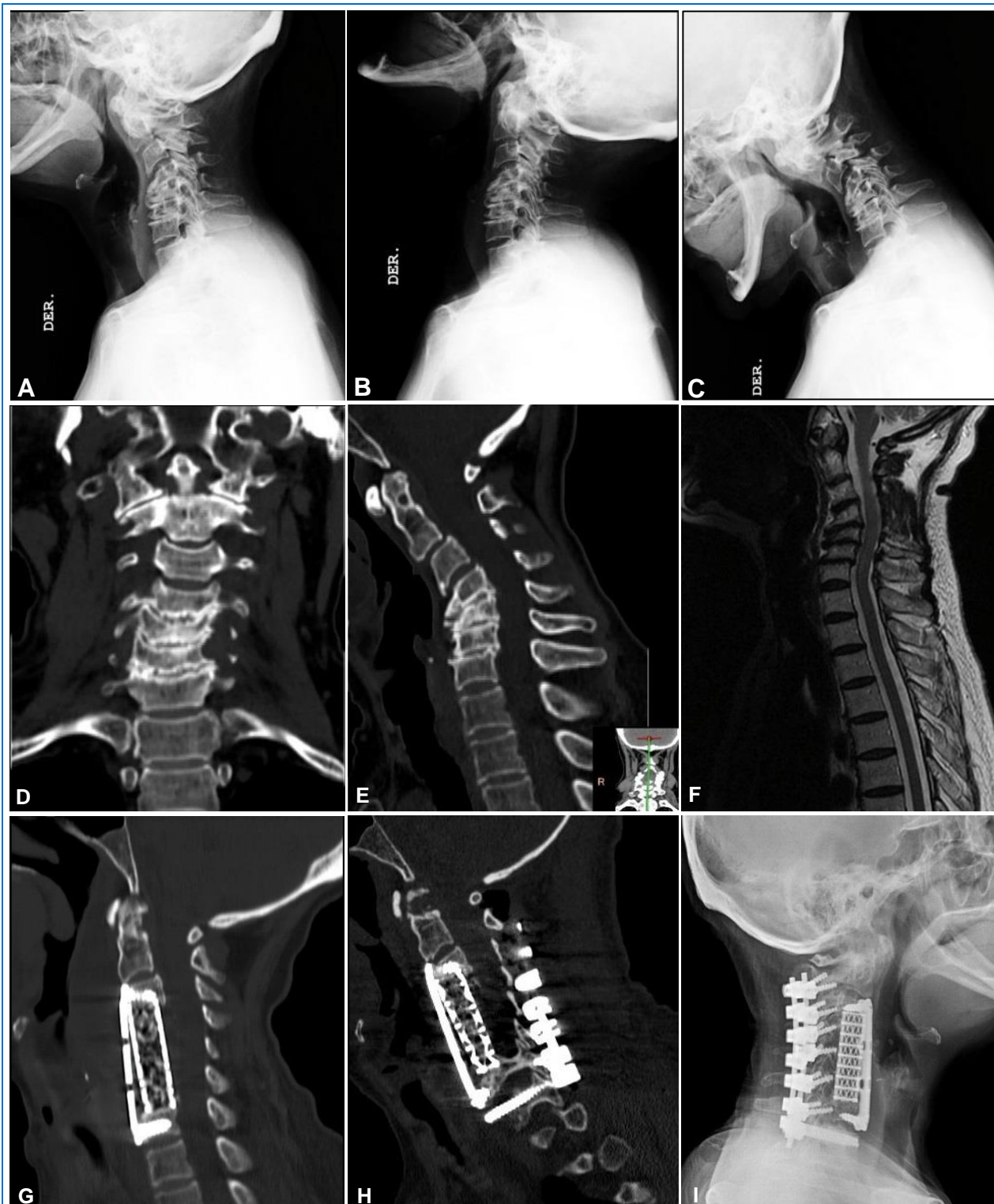


Fig 3. (Case 4): 60-year-old female patient with rheumatoid arthritis for 30 years, under treatment with prednisone, Etanercept and methotrexate. History of high blood pressure, chronic gastritis, wrist arthrodesis, elbow dislocation surgeries and appendicitis. Neck pain was the main symptom, being classified on the Ranawat scale as type II. Inflammatory serum markers: CRP 11 and ESR 38. X-ray images show involvement of the subaxial cervical spine with signs of instability on dynamic images in (A) neutral position, (B) hyperextension and (C) hyperflexion. (D, E) Tomography images and (F) Cervical MRI that more clearly show the bone and soft tissue involvement at the cervical spinal level. The patient underwent surgery on two occasions, (G) initially an anterior arthrodesis was performed by performing a corpectomy with placement of a cylindrical titanium mesh and anterior plate. (H, I) In a second stage, a posterior arthrodesis was performed with screws to lateral masses.

involvement is paramount in the early detection of the initial stages of the disease even in the absence of symptoms.¹³ In our series, patients who underwent surgery were referred from the medical service, 100% with symptoms. Neck pain, specifically pain in the cranio-cervical junction is one of the most common presentations, according to a report this symptom occurs in 69% of patients with cervical instability.^{2,8,13,16} Occipital headache also is a common complaint, present in 60% of patients with atlantoaxial instability and in 90-100% of patients with cranial seating, and can be attributed to compression of the major and minor occipital nerves as they pass between C1 and C2.^{2,8} In

our series, posterior cervical pain was the most predominant, and specific pain at the occipitocervical junction was observed in atlantoaxial instability and cranial settlement.

Cervical spine involvement is of importance because, if left untreated, it can lead to significant neurological morbidity, worsen quality of life, and sometimes cause sudden death from stroke, obstructive hydrocephalus, or cardiac arrest.^{2,6,9,17}

When cervical spine involvement becomes symptomatic,

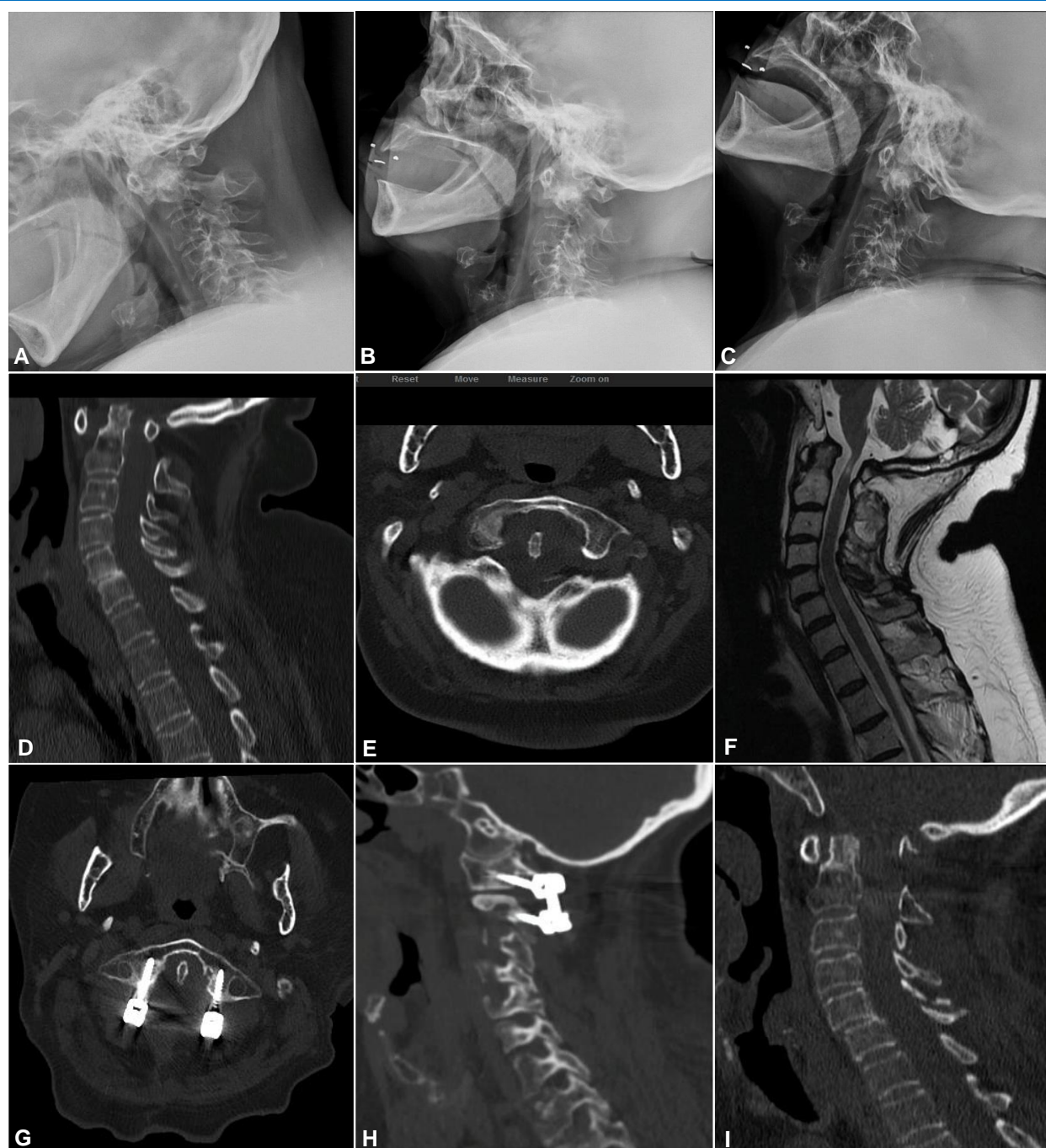


Fig 4. (Case 6): 56-year-old female patient with a 12-year history of rheumatoid arthritis, cesarean section, cholecystectomy, hysterectomy, bilateral hip replacement, on treatment with prednisone, methotrexate, azathioprine, leflunomide, and sulfasalazine. She presented posterior cervicalgia and quadriparesis. Laboratory tests showed elevated CRP and ESR. (A, B, C) The cervical functional radiograph showed the presence of atlantoaxial dislocation. (D, E, F) Cervical tomography and magnetic resonance imaging showed C1-C2 subluxation and a moderate narrow canal. (G, H, I) Surgery was performed by C1-C2 posterior cervical arthrodesis, achieving reduction of C1-C2. The clinical evolution was good, going from grade III on the Ranawat scale to grade II.

surgical stabilization should be considered as it has been shown to delay and sometimes prevent disease progression and improve functional status in certain patients.^{2,3,4,6,8,17} The presence or development of peripheral joint erosions, DMARD failure, prolonged use of corticosteroids, and increased disease activity (elevated ESR or CRP) are significant risk factors for the development or presence of spinal involvement cervical.^{3,6,10,11,12,13,17} It should be noted that most of our patients had elevated inflammatory markers at the time of diagnosis of cervical instability.

The objectives of treatment in these patients are relieve pain and avoid the development of irreversible neurological deficit and sudden death. Surgical indications include the presence of instability, neurological deficit, and intractable neck pain. Certain radiological parameters may indicate surgery in the presence or absence of neurological symptoms, such as atlantoaxial instability with "PADI" equal to or <14mm; migration of the odontoid over the Mc Gregor line> or equal to 5mm and a sagittal diameter of the medullary canal equal to or <14mm, in the presence of subaxial subluxation.¹⁰ To these criteria could be added the combination of the alteration of Ranawat measurements, Redlund-Johnell and Clark Station. Finally, a cervico-medullary angle <135° should also be considered in the Magnetic Resonance or evidence of medullary compression in images.^{14,17,18,19} Added to the clinical features of the patients in our series, imaging tests (functional radiography, tomography, and magnetic resonance imaging) showed the presence of cervical instability guided by the criteria mentioned above.

Surgical stabilization using arthrodesis techniques has evolved in recent years from the appearance of sublamina fixation techniques first described by Gallie in 1939 through the C1-C2 transarticular fixation with screws described by Magerl in 1986 and the C1-interarticular fixation. C2 with screws and plate by Goel in 1994, or with poliaxial screws and bars described by Harms 2001.^{15,17,18} We perform posterior cervical arthrodesis techniques using mostly poliaxial screws and bars, in some cases of our series we place cylindrical titanium meshes and plates for anterior arthrodesis.

Reducible atlantoaxial subluxation can be managed with a C1-C2 fusion. If there is irreducibility of this alteration, or a partial reduction of the canal, a decompression (C1 laminectomy) is necessary. In cases of atlantoaxial impaction, the preoperative use of traction, using a halo, is recommended to try to reduce the impaction and avoid the need for decompression. If traction is successful, instrumentation and posterior arthrodesis from C0 to C2 can be performed. Preoperative images indicate whether the fusion should be prolonged caudally, including all subluxated segments. If the atlantoaxial impaction is irreducible or a posterior fusion without reduction remains symptomatic, odontoidectomy is indicated, and can be performed transorally or with a high retropharyngeal approach.¹⁸ We use posterior C1-c2 interarticular arthrodesis techniques in cases of atlantoaxial instability and posterior C0-C1-C2 arthrodesis when we also observe cranial settlement.

Subaxial instability can be treated by posterior or anterior fusion and decompression techniques as our patients were managed. The results of surgical procedures are more variable in relation to neurological recovery. One of the most important predictors of post-operative recovery is the

previous neurological condition, which can be assessed using the Ranawat Classification of rheumatoid myelopathy. The greater the preoperative deterioration, the lower the expected recovery.^{14,17,18} In our study and thanks to surgery, we managed to improve the neurological clinic (90%) in most of our patients.

Due to the serious and life-threatening complications of cervical spine disease in RA, its early diagnosis and treatment should be a priority in patients with RA.

CONCLUSIONS

Current treatment of cervical spine involvement by RA is aimed at stabilizing and stopping the progression of neurological involvement. These patients with vertebral instability may present asymptomatic or with posterior neck pain, as in most patients. After clinical and imaging evaluation, the type of instability (AAI, CS or SAS) can be determined and the appropriate surgical treatment can be carried out accordingly.

Similarly, the surgical risk must be weighed against the risk of conservative treatment. The optimal result depends on the adequate selection of patients evaluating the clinic and images. Surgery can promote neurological recovery and decrease mortality.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: All the authors. *Drafting the article:* Zumaeta J. *Critically revising the article:* Cari E, Basurco A. *Reviewed submitted version of manuscript:* Zumaeta J. *Approved the final version of the manuscript on behalf of all authors:* Zumaeta J.

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