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Arnold's Neuralgia Complicating Rheumatoid Arthritis: About a Case Report and Literature Review

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Abstract

Introduction: Described in 1834, Arnold's neuralgia is a headache of cervical origin secondary to irritation or compression of the large posterior occipital nerve and/or the small occipital nerve. Headaches are unilateral, and evolve in paroxysms. The etiologies of NA are many and varied. They may be vascular, neurological, arthropathic or systemic, including rheumatoid arthritis. We report a Senegalese case of NA in a patient treated for RA, and then establish the current state of this association in the African region. **Observation:** This 47-year-old patient had been treated for 17 years for sequelae of rheumatoid arthritis (RA), and was seropositive for both rheumatoid factor and anti-citrullinated peptide antibodies. The levels of these autoantibodies were 103 IU/ml and 4000 IU/ml respectively. Polyarthritis was predominantly acromelic, characterized by ankylosing deformities. Since diagnosis, she had been treated with methotrexate (15 mg, then 20 mg per week), folic acid at a dose equivalent to methotrexate, and hydroxychloroquine (400 mg per day). Prednisone 10 mg corticosteroid therapy was administered on admission and gradually discontinued after two years' follow-up. Recent symptomatology was marked by a polyarticular, poly synovial flare-up over the past 6 months. In addition, the patient reported paroxysmal cervical pain. It was lateralized to the right, with tension and electric discharges. The rheumatological examination, in addition to the aforementioned joint disorders, revealed pain provoked by palpation and mobilization of the cervico-occipital hinge. Cervical mobilization was limited, particularly to flexion and extension movements. Biological tests revealed an inflammatory syndrome, with a sedimentation rate of 76 mm, a C-reactive protein (CRP) of 87 mg/l, and a microcytic hypochromic anemia with a he-

moglobin level of 8.6 g/dl. Cervical computed tomography revealed fusion of the atloido-axoid joint, with geodes and erosions on the edges of the anterior arch of C1, with no atloido-axoid dislocation detected. The electro-neuro-myogram (ENMG) showed a neurogenic pattern. This cervical symptomatology was compatible with Arnold's neuralgia. However, because of the ankylosis, the neurosurgeons did not recommend surgical decompression. Medical treatment was therefore reinforced by the addition of pregabalin (150 mg daily) and physiotherapy sessions. The short-term outcome was favorable, with partial pain regression. **Conclusion:** The originality of our case lies in the rarity of cervical spinal involvement in Africa. On the other hand, the fact that Arnold's neuralgia was discovered remains exceptional, whatever the population studied.

Keywords

Arnold's Neuralgia, Rheumatoid Arthritis

1. Introduction

Described in 1834, Arnold neuralgia (AN) is a headache of cervical origin secondary to irritation or compression of the posterior greater occipital nerve (GNO) and/or the lesser occipital nerve (**Figure 1**) [1]. Headaches are unilateral, with paroxysmal progression [1]. They are located just above the cervico-occipital hinge and radiate to the homolateral supraorbital region [1]. The diagnosis is made on the basis of suggestive clinical symptoms and radiological or electrical

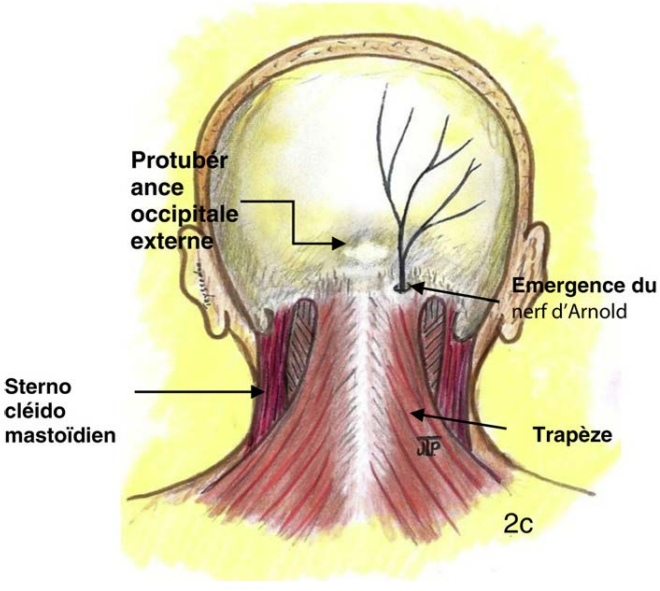


Figure 1. Anatomy of Arnold's nerve [1]. Protubérance occipitale externe -> External occipital protuberance; Emergence du nerf d'Arnold -> Emergence of the Arnold nerve; Sterno cléïdo mastoïdien -> Sternocleidomastoid; Trapèze -> Trapezius.

confirmation of nerve damage. The etiologies of NA are many and varied. They can be vascular, neurological, arthropathic or systemic, and include rheumatoid arthritis (RA) [1]. Indeed, certain factors predisposing to cervical involvement are specific to rheumatoid arthritis. These include the development of inflammatory pannus at the synovial C1-C2 joint, as well as long-term corticosteroid therapy. Identified risk factors include erosive polyarticular involvement, disease duration, and positivity for rheumatoid factor [2].

NA following RA is mainly reported in Western literature. In North Africa, as in sub-Saharan Africa, it is rarely described.

We report a Senegalese case of NA in a patient treated for RA, and then establish the current state of this association in the African region.

2. Observation

This 47-year-old patient had been treated for 17 years for sequelae of rheumatoid arthritis (RA), and was seropositive for both rheumatoid factor and anti-citrullinated peptide antibodies. The levels of these autoantibodies were 103 IU/ml and 4000 IU/ml respectively. Polyarthritis was predominantly acromelic, characterized by ankylosing deformities. Examination of the hands revealed button-hole fingers, Z-shaped thumbs, bilateral ulnar cupping and amyotrophy of the interosseous muscles (**Figure 2**). Since her diagnosis, she had been treated with methotrexate (15 mg, then 20 mg weekly), folic acid at a dose equivalent to methotrexate, and hydroxychloroquine (400 mg daily). Prednisone 10 mg corticosteroid therapy was administered on admission and gradually discontinued after two years' follow-up.

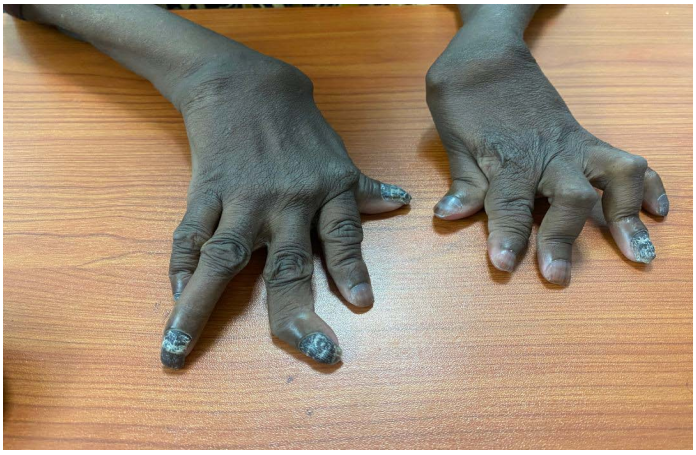


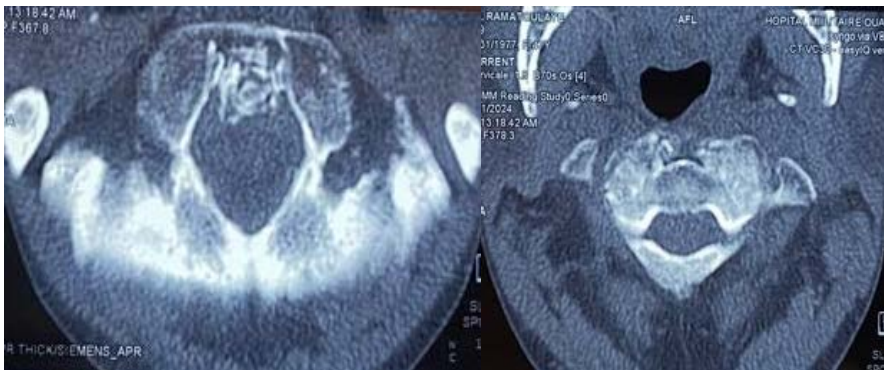
Figure 2. Characteristic RA deformities in the hands of our patient.

Recent symptomatology was marked by a polyarticular, poly synovial flare-up over the past 6 months. In addition, the patient reported paroxysmal cervical pain. It was lateralized to the right, and felt like tension and electric shocks. Its intensity was estimated at 8/10 on the visual analog scale.

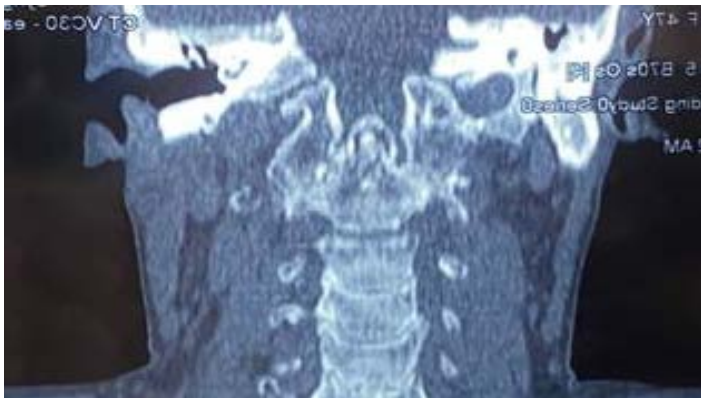
The rheumatological examination revealed, in addition to the aforementioned

joint disorders, pain provoked by palpation and mobilization of the cervico-occipital hinge. Cervical mobilization was limited, notably to flexion and extension movements.

Biological tests revealed an inflammatory syndrome, a sedimentation rate of 76 mm, a C-reactive protein (CRP) of 87 mg/l, and microcytic hypochromic anemia with a hemoglobin level of 8.6 g/dl. Cervical CT revealed fusion of the atloido-axoid joint, with geodes and erosions on the edges of the anterior arch of C1, with no atloido-axoid dislocation detected (**Figures 3(a)-(c)**). The electro-neuro-myogram (ENMG) showed a neurogenic pattern. This cervical symptomatology was compatible with Arnold's neuralgia.



(a) (b)



(c)

Figure 3. Cervical CT in our patient showing fusion of the associated atloido-axoid joint with erosions and geodes at the edges of the anterior arch of C1. No atlantoaxial dislocation detected ((a) and (b): axial section; (c): frontal section).

However, because of the ankylosis, the neurosurgeons did not recommend surgical decompression. Medical treatment was therefore reinforced by the addition of pregabalin (150 mg daily) and physiotherapy sessions.

The short-term course was favorable, with partial pain regression.

3. Discussion

We report the case of a patient suffering from Arnold's neuralgia complicating

sequelae of RA. This patient, who had been followed for 17 years, had characteristic RA deformities of the wrists and hands, as well as atloido-axoid ankylosis associated with NA. This C1-C2 involvement of Arnold's nerve is an integral part of the cervical complications of RA [3]. Our patient received pregabalin treatment combined with physiotherapy sessions, resulting in favorable short-term progress marked by a reduction in neuropathic pain and improvement in cervical stiffness.

Cervical involvement in general, and NA in particular, is rarely reported in African literature. Indeed, no case of cervical involvement has been reported in the majority of the largest series on RA in sub-Saharan Africa, notably in Nigeria with the study by Yerima A *et al.* [4]; in Burkina-Faso with Ouédraogo DD *et al.* and Ouédraogo NCJ *et al.* [5] [6] and in Senegal we have the work of Ndongo N *et al.*; Niasse M *et al.* [7] [8]. In North Africa, the same situation was observed in work carried out in Morocco by Benamour S *et al.* [9] and in Egypt by Sakr BR *et al.* [10].

The rarity of rheumatoid cervical spine in Africa could be linked to underestimation, as in Congo Brazzaville, Ntsiba *et al.* report rheumatoid cervical spine in 32% of patients [11]. However, the nature of the lesions has not been specified to confirm or affirm the presence of damage to the greater occipital nerve. On the other hand, in Western literature, rheumatoid cervical involvement in general is reported in more than half of patients after an average evolutionary duration of 10 years [12] [13]. This is consistent with the duration of the disease in our patient, who had been followed for 17 years. The magnitude and frequency of this range in Caucasians vary according to study populations and therapies administered [14] [15]. Prevalence is estimated at between 17% and 88% [14] [15]. In the studies of Blom M, Yurube T and Ahn JK, the prevalence of cervical spinal complications in RA was estimated at 16% [16], 70.4% [17] and 28.6% [18] respectively. Husham reported a lower prevalence of 7.4% [19]. Cervical rheumatoid involvement, which seems rare in black Africans, is frequent in some Westerners; This difference in prevalence may be explained by ethnic variations. However, Arnold's neuralgia in rheumatoid arthritis remains poorly documented regardless of the study population [4]-[8] [20]. Studies of the rheumatoid cervical spine have focused more on atloido-axoid subluxation and rarely on damage to péri-vertebral structures, including Arnold's nerve [1] [21]-[23]. Our case report presents certain diagnostic limitations, particularly regarding the structural assessment of nerve involvement. Indeed, most studies rely on magnetic resonance imaging (MRI), which provides better visualization of the inflammatory pannus and the affected nerve structures.

4. Conclusion

The originality of our case lies in the rarity of cervical spinal involvement in Africa. On the other hand, the fact that Arnold's neuralgia was discovered remains exceptional, regardless of the study population. This observation highlights the importance of enhanced clinical and radiological monitoring of the cervical re-

gion during rheumatoid arthritis, in order to enable early detection and appropriate management of rheumatoid cervical spine involvement.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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