

**ATLANTOAXIAL DISLOCATION IN SPONDYLOARTHRITIS: 8 NEW CASES**

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**ABSTRACT**

The C1-C2 dislocation is little known in spondyloarthritis while it can cause severe neurological manifestations. The aim of this work was to study clinical and radiographic features of C1-C2 subluxation in a sample of Tunisian spondyloarthritis (SA) patients. We report a series of 8 SA patients with atlanto-axial dislocation. We diagnosed anterior dislocation in 7 cases. It was associated with C1-C2 rotatory subluxation in one case. One patient displayed an isolated rotatory dislocation with lateral erosive arthritis of the lateral masses of the atlas. A peri-odontoid pannus was objectified in 6 cases. Among them two displayed a lysis of odontoid. One patient had a vertical dislocation of C2 associated with a forward subluxation. No spinal cord compression was noted in all cases. We believe that the proliferation of systematic studies of the cervical spine before any ankylosing spondylitis evolved is essential.

**KEYWORDS:** Spondyloarthritis, cervical spine, dislocation.**BACKGROUND**

Spondyloarthritis is a chronic inflammatory disease evolving progressively towards ankylosis, the spine and the sacroiliac are preferentially affected. The cervical spine in SA offers a wide variety of lesions. Among them C1-C2 dislocation which is also described in rheumatoid arthritis RA. But unlike the latter, the C1-C2 subluxation during AS benefited from little research. The possibility of severe neurological complications leads us to present 8 cases diagnosed over the last eight years.

**METHODS**

A retrospective case series study conducted at the rheumatology department of the National Institute of Orthopedics. Patients with cervical spine dislocation and spondyloarthritis between 2006 and 2014 were identified. Data were then collected from their clinical case records. The presence of pain in the neck, occipital, temporal or retro-orbital areas, and radicular pain in the upper extremities was noted. Radiographs of the cervical spine included lateral views taken in flexion and neutral positions were analyzed. A diagnosis of forward dislocation was made if the distance between the anterior aspect of the dens and the posterior aspect of the anterior arch of atlas was more than 3 mm during flexion. Radiographs, computed tomographic (CT) scans and magnetic resonance imaging (MRI) scans were reviewed and relevant findings such as site and type of dislocation noted. Demographic data, neurological status on admission, functional outcome and complications were noted from the clinical case records for all patients.

**RESULTS**

During 2008 to 2014, eight (4 men and 4 women) AS patients were recruited. Even though patients were negative for HLA-B27, the clinical manifestations of their disease were sufficient to meet the AMOR and the ESSG classification criteria for axial and peripheral AS.<sup>[1]</sup> Their Age ranged from 22 to 59 years (Mean 34.5). The ages of onset and disease duration were respectively 26.83 years and 7.66 years on average (ranging respectively between 15-50 years and 1-22 years). There were no trauma history. The mean BASDAI was 56%, the mean BASFI was 55%, the mean the average BASRI was 5.29. Two patients were receiving NSAIDs. Two others were taking Methotrexate at a 17.5 mg per week dose, three other patients were on TNF $\alpha$  blockers (Etanercept) and one patient on sulphasalazine at a 2 grams per day dose. All patients presented with inflammatory cervical pain lasting for 1 to 10 years before diagnosis (Mean 5.5 years). The range of motion of the cervical spine was decreased in all cases, its movements were accompanied by tenderness. The muscle strength of the upper extremities was unaffected. On neurological examination, we displayed quadripyramidal syndrome in 5 cases with no motor nor sensory deficit. Biologically an inflammatory syndrome was objectified in all cases with a mean CRP at 76 mg / l and an erythrocyte sedimentation rate at 70 mm.

The pelvic X-ray indicated the presence of bilateral sacroiliitis in all cases and coxitis in 5 cases (bilateral coxitis in 2 cases). The cervical spine X-ray showed that there was a 55 mm anterior displacement of the cranio-

atlantal complex on the axis in **6 cases**. Dynamic imaging showed that there was no significant instability. C1-C2 forward dislocation was the most common pattern of cervical spine injury in SA occurring in 7 patients. The MRI of the cervical spine with and without contrast indicated the presence of anterior displacement of C1 in 7 cases. It was associated with C1-C2 rotatory subluxation in one case. One patient displayed an isolated rotatory dislocation with lateral arthritis of C1. A peri-odontoid pannus was objectified in 6 cases. Among them two displayed a lysis of odontoid. One patient had a vertical dislocation of C2 associated with a forward

subluxation. No spinal cord compression were noted in all cases. Surgical treatment were proposed but not retained due to the absence of neurological impact in three cases. In the remaining cases, patients refused surgery. All patients had neck braces with triple support: chin, occipital and sternal.

Five of them have had corticosteroids boluses with regression of the pyramidal irritation signs. All patients are followed to date with clinical and radiological stability.

The following table summarizes the main features of cervical dislocations in our patients.

	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7	Case 8
Neck pain	+	+	+	+	+	+	+	+
Pyramidal syndrom	+	-	+	-	+	+	-	+
Plain cervical radiographs C1-C2 diastasis (mm)	5	6	7	-	5	6	5.5	5.5
MRI findings	Forward Dilocation	+	+	+	-	+	+	+
	Rotatory dislocation	-	+	-	+	-	-	-
	Peri-odontoid pannus	+	+	+	-	+	+	-
	Vertical dislocation	-	-	-	-	-	-	+

## DISCUSSION

C1-C2 subluxation is an unusual phenomenon in spondyloarthritis. To date, there were no study indicating the exact prevalence of this complication among Tunisian patients. The transverse ligament, which is the primary C1/C2 stabilizer of the anterior arch of the atlas and the odontoid process, provides strong support to the atlanto-odontoid joint. A mean force of 84 kg is needful for rupturing the transverse ligament, and causing therefore a 3–5 mm anterior displacement of C1 on C2.<sup>[2]</sup> Incompetence of the transverse ligament of C1 vertebra has generally been accepted as the cause of forward dislocation of C1 on C2 vertebra.

Theories of the mechanism includes enthesitis of the transverse ligament<sup>[2,3]</sup> and peri-odontoid pannus formation.<sup>[4]</sup> First, weakness of this ligament may result in instability of the atlanto-odontoid joint, leading to the atlas slipping anteriorly when the head is flexed. Second it is possible to explain it based on analogous events observed in rheumatoid and psoriatic arthritis, which have been attributed to pannus formation. The inflammatory process that resulted in pannus formation can extend to the surrounding tissues causing destruction of the skeletal and ligamentous support. MRI findings in six of our eight cases demonstrating a soft tissue mass at the peri-odontoid area may support this mechanism.

The clinical course in our patients tended more to peripheral arthritis than axial SA. This was observed with increased frequency in existing SA patients with persistent peripheral arthritis.<sup>[3]</sup> In the treatment of peripheral arthritis in SpA, DMARDs such as methotrexate and sulphasalazine, though ineffective in spinal inflammation<sup>[8-10]</sup>, are accepted as the initial treatment of peripheral arthritis in SpA. Anti-TNF

therapy, a second line agent for arthritis refractory to DMARDs, may have additional advantage in the treatment of AAS in SpA due to its effectiveness in axial spinal disease and enthesitis<sup>[9-10]</sup>, the other postulated mechanisms for AAS in SpA.

Chronic neck pain is usually the result of greater occipital nerve impingement and C1/C2 arthrodesis usually eliminates the pain.<sup>[11,12]</sup> Although there are no established guidelines, surgery is usually indicated in patients who have neurological features and intractable neck pain. Surgery in asymptomatic patients with radiographic evidence of cervical instability without significant pain or any neurological deficit remained controversial. Patients with posterior atlanto-dental interval (PADI) that is lesser than 14 mm on lateral neck flexion radiographs, and decreased space around the cord lesser than 13 mm on MRI are recommended for prophylactic arthrodesis.<sup>[11,12]</sup>

## CONCLUSION

While the cases discussed above highlighted the features of cervical dislocations in SA. C1-C2 subluxation should be considered in patients with persistent neck pain. Early recognition may allow for early medical intervention, and potentially avert the need for C1/C2 spinal fusion surgery.

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