

Spontaneous Atlantoaxial Rotatory Fixation in Old Age After Cerebral Infarction

Case Report

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Study Design. Case report of spontaneous Fielding and Hawkins Type I atlantoaxial rotatory fixation in a 78-year-old man after hemiplegia and homonymous hemianopsia caused by cerebral infarction.

Objectives. To describe a case of spontaneous atlantoaxial rotatory fixation in old age and review previous adult cases of atlantoaxial rotatory fixation without fracture.

Summary of Background Data. Atlantoaxial rotatory fixation in adults is a relatively rare finding and is mainly caused by trauma. To the author's knowledge, there has been no previous report of spontaneous atlantoaxial rotatory fixation in old age.

Methods. The patient's head was fixed in a 40° left-rotated position. Left hemiplegia and homonymous left-side hemianopsia developed due to cerebral infarction. Computed tomography of the cervical spine clearly showed rotatory fixation of the atlas on the axis.

Results. Successful reduction was obtained after 1 day of skull traction.

Conclusions. It was hypothesized that repeated left-rotational stress due to homonymous hemianopsia loaded to the atlantoaxial joint caused abnormal laxity of the joint. [Key words: atlantoaxial rotatory fixation, elderly, hemianopsia] *Spine* 2000;25:2137–2140

Atlantoaxial rotatory fixation (AARF) was first reported by Bell in 1830.² It is more common in children than in adults, and adult AARF is usually caused by trauma.^{1,14,15} According to Fielding and Hawkins,⁵ AARF can be classified into four types when the atlas-to-dens interval is taken into account. Type I is rotatory fixation with no anterior displacement and the odontoid acting as the pivot. Type II is rotatory fixation with anterior displacement of 3 to 5 mm and one lateral articular process acting as the pivot. Type III is rotatory fixation with anterior displacement of more than 5 mm. Type IV is rotatory fixation with posterior displacement.⁵ The current case involved a Fielding and Hawkins Type I AARF of nontraumatic origin in an elderly man.

Case Report

A 78-year-old man visited the authors' clinic with painful left-side, fixed torticollis of sudden onset. He exhibited left hemiplegia caused by cerebral infarction for which he had received

rehabilitation for 1.5 months in the authors' hospital. There was no history of cervical trauma before this episode. His head was fixed in a 40° left-rotated position (Figure 1). He was unable to tilt or rotate his head to the right because of severe neck pain. The right sternocleidomastoid was spastic and resistant to an attempt to correct the deformity. The pain was located at the major occipital nerve area. In addition to left hemiplegia, left homonymous hemianopsia due to cerebral infarction was noted. Left upper and lower extremities had approximately four-point muscle weakness detected by manual muscle testing, with no evidence of neurologic deterioration after this episode. Laboratory data showed weak inflammatory reaction of elevated C-reactive protein (3.6 mg/dL) and erythrocyte sedimentation rate (35 mm/hr). Radiography of the cervical spine showed that the head was held in a position of left rotation (Figure 2). The standard anteroposterior open-mouth view of C1–C2 could not be obtained because of severe neck pain. A computed tomographic (CT) scan of the cervical spine clearly showed the rotatory fixed atlas (C1) and axis (C2) with C1 held in a 40° left-rotated position without anterior displacement (Figure 3). Fielding and Hawkins type I AARF was diagnosed.

Eight days after the onset, skull traction with a 6-kg weight was initiated, and weight was gradually added to reach 12 kg temporarily. Reduction was obtained on the same day. The patient was kept on a bed under 2 kg continuous skull traction for 2 more days, and allowed to walk with a collar thereafter.

After reduction, the patient reported treatment-related pain in the left shoulder for a few days, but no neurologic deterioration occurred. Radiographs and CT showed good reduction (Figures 4–6). There was no asymmetry of the articular facets of C1 in relation to the dens on the open-mouth view (Figure 5). Retropharyngeal swelling was found after reduction (Figure 4). Neither recurrence of AARF nor neurologic deterioration was observed within 5 months after reduction.

Discussion

The precise pathologic features of this condition were first documented by Fielding and Hawkins in 1977.⁵ Based on 17 cases of irreducible atlantoaxial rotatory fixation, they described the diagnostic features by physical abnormalities and roentgenography. According to the anteroposterior displacement of the atlas, they classified the condition into four types, which provided valuable guidance concerning the prognosis and treatment. Standard roentgenographic studies are often inconclusive, and AARF is difficult to diagnose, because the position of the head may cause overlap of adjacent bone structures making interpretation of the abnormal relations of the at-

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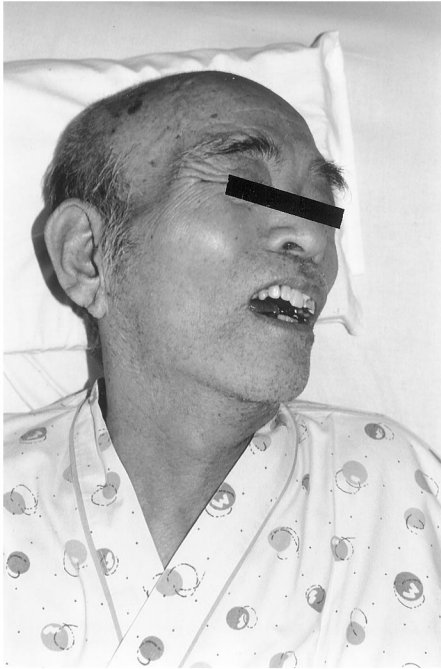


Figure 1. Photograph of patient at admission showing the head held in a left-rotated position.

las, axis, and occiput difficult. The CT scan was indispensable in making the diagnosis in the present case.

The atlantoaxial joint has characteristic anatomic structures compared with other spinal segments including the existence of the synovial joint of the odontoid. Transverse and alar ligaments play a major part in stabilization of the atlantoaxial joint. Injuries of these ligaments may cause dislocation of the atlantoaxial joint, but another factor is required to produce rotatory fixation.

In children with AARF, idiopathic origin and causes such as trauma and upper respiratory infection have been indicated. The higher incidence of AARF in children may be related to certain anatomic structures that



Figure 2. Initial radiograph of cervical spine.



Figure 3. Computed tomogram before reduction clearly showed the rotatory fixed atlas (C1) and axis (C2) with C1 held in a 40° left-rotated position without anterior displacement.

are peculiar to children.¹² The dens–facet angle of the axis is steeper in children than in adults. Meniscus-like synovial folds are found in the C0–C1 and C1–C2 facet joint of children but not in those of adults. In contrast, AARF is a very rare finding in adults, especially in the elderly. The cause of AARF in adults (including old age) is considered to be trauma and rheumatoid arthritis; other origins are idiopathic. The authors reviewed the reported cases of AARF in adults^{3,5,6,8,10,11,13,16–19,21,22,25} (Table 1), and AARF in adults caused by trauma, especially high-energy trauma such as traffic accidents and falls, was seen but no previous cases of spontaneous AARF in the elderly were found.

Of the various theories regarding the cause of AARF, several are related to the capsular laxity of the atlanto-



Figure 4. Radiograph made after reduction. Retropharyngeal swelling was found.

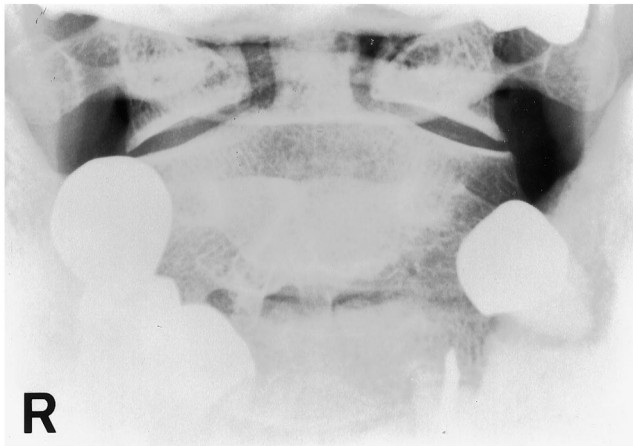


Figure 5. Radiograph made after reduction. Open-mouth antero-posterior view shows no asymmetry of the articular facets of C1 in relation to the dens.

axial joint. Wittek²⁴ attributed the onset to an intra-articular factor causing effusion in the synovial joint that resulted in ligament distension. Coutts⁴ proposed the theory that synovial fringes, when inflamed or adherent, may block reduction.⁴ Wortzman and Dewar²⁵ reported that AARF was due to impingement of the articular capsule. Other investigators have suggested several extra-articular factors as the cause of AARF. Grisel⁹ suggested that muscle contracture after an upper respiratory tract infection is a factor. Fioranni-Gallotta and Luzzatti⁷ reported that the deformity was due to rupture of one or both alar ligaments and the transverse ligament. Watson-Jones²³ hypothesized that inflammation in the neck or pharynx caused decalcification of the bone at the side of ligamentous attachments, resulting in loosening of the ligaments of the upper cervical region. Fielding and Hawkins⁵ suggested a theory combining intra- and extra-articular factors. They suggested that the reduction

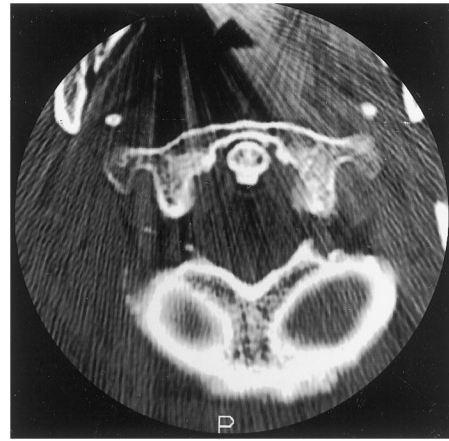


Figure 6. Computed tomogram made after reduction showing improvement in the alignment of C1 and C2.

of the rotational deformity is obstructed in the early stages by swollen capsular and synovial tissues and by associated muscle spasm. Sustained spasm caused ligament and articular capsule contracture resulting in joint fixation. Recently Tucker and Taylor²⁰ thought that the mechanism of AARF could be explained by rigid bony interlocking of the facet joint, reproducible in dry bones.²⁰

In the current case, the patient had some characteristic features. He had homonymous left-side hemianopsia due to cerebral infarction. Although he could walk with a walking device, he spent most of the day in bed watching television. The television was located at the left side of his bed, and his head was therefore held in a left-rotated position for long periods. The homonymous left-side hemianopsia enhanced the tendency. The authors hypothesized that repeated stress loaded to the atlantoaxial while watching television provoked laxity of the capsule and/or ligaments of the atlantoaxial joint and excess movement of this joint.

Table 1. Summary of Previous Reported Cases of AARF Without Fracture in Adult

Case	Age	Gender	Cause	Treatment	Literature	
1	18	F	idiopathic	injection	Wortman ²⁵	1968
2	32	M	trauma	traction	Wortman ²⁵	1968
3	28	M	trauma	traction	Wortman ²⁵	1968
4	68	F	RA	traction	Fielding ⁵	1977
5	27	F	idiopathic	traction → posterior fusion	Fielding ⁶	1978
6	46	M	trauma	traction	Rinaldi ¹⁶	1979
7	67	M	trauma	traction → anterior fusion	Kobayashi ¹³	1983
8	19	F	angiography*	manipulation → posterior fusion	Shoji ¹⁷	1984
9	16	F	trauma	traction → posterior fusion	Clark ²	1986
10	48	F	trauma	traction	Van Holsbeeck ²¹	1989
11	22	F	minor trauma	traction	Van Holsbeeck ²¹	1989
12	18	M	trauma	traction	Van Holsbeeck ²¹	1989
13	24	F	unknown	unknown	Suzuki ¹⁸	1992
14	73	M	infection	unknown	Harata ¹⁰	1995
15	49	F	trauma	traction → anterior fusion	Fujii ⁸	1995
16	75	M	trauma	traction	Higuti ¹¹	1996
17	22	F	trauma	traction → posterior fusion	Takano ¹⁹	1996
18	17	M	trauma	traction → halo vest	Waegeneers ²²	1997

* angiography of carotid.

AARF = atlantoaxial rotatory fixation; RA = rheumatoid arthritis.

■ Conclusions

This was a case of Fielding and Hawkins Type I AARF in a 78-year-old man who had no history of cervical trauma before this episode. A CT scan was very useful in the diagnosis of AARF. Laxity of the capsule and/or ligaments of the atlantoaxial joint due to repeated head rotation in the same direction was considered to be the cause.

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