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Neglected Case of C1/C2 Instability- How it can be Treated After 16 Years of Injury?

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Authors' contributions

This work was carried out in collaboration between both authors. Author MS wrote the first draft of the manuscript and proposed the study design. Author SZ performed statistical analysis and searched the literature. Both authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Even in the era of modern technology, odontoid fractures can be easily overlooked and undertreated due to the specific mechanism of injury and anatomical features.

We present a case of odontoid fracture in young sportsman followed by atlanto-axial dislocation. The surgical treatment occurred 16 years after injury due to low compliance and lack of significant symptoms during 16 years.

This case was challenging regarding proper classification, diagnosis and treatment options. However, final result was satisfactory.

Keywords: Odontoid fracture; missed spine fracture; delayed diagnosis; C1/C2 fusion.

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ABBREVIATIONS

ADI	: Atlas- Dens Interval
C1	: First Cervical Vertebra (Atlas)
C2	: Second Cervical Vertebra (Axis)
ASIA score	: American Spinal Injury Association
CT	: Computed Tomography
MRI	: Magnetic Resonance Imaging
SAC	: Space Available for Cord
VAS	: Visual Analogue Scale

1. INTRODUCTION

Cervical spine fractures are very common, with incidence from 118/100 000 (in Norwegian) [1] to 65/100,000 in China [2]. There are two peaks of incidence regarding age of life. The highest incidence is in the group from 15-45 years and the second peak is in the group from 65-80, and this group is especially challenging regarding the treatment options [3,4]. Motor vehicle associated injuries, fall from the height, age <40, pelvic fracture, Injury Severity Score (ISS) >15 are strong predictors of cervical spine injury [5]. Among cervical spine fractures odontoid fractures remain a great challenge for diagnosis and treatment due to the unique features of C2 anatomy and potentially devastating complications, even life threatening [6,7]. Odontoid fractures currently account for 9–15% of all adult cervical spine fractures with type II fractures accounting for the 60% of these injuries [8]. Despite state of the art equipment and very good trauma protocols, even today there is a significant incidence of missed cervical spine injuries [9], especially in less developed countries and in patients with lack of compliance. The proper diagnosis of upper cervical spine fractures in unconscious patients remains a great problem [10,11]. In our case, the young sportsman sustained his injury 16 years before treatment and we were faced with the problem of proper diagnosis, classification and treatment of this unusual condition.

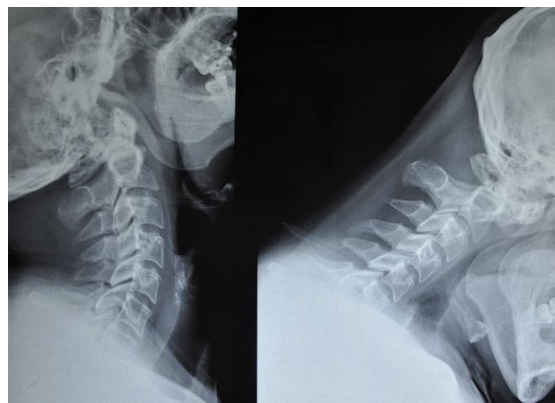
2. PRESENTATION OF CASE

A 35 year-old-man fell on his head during a football match in the year 1999. At the moment of the injury, he complained about temporary tetraparesis with duration of about 1 min. He did not undergo any medical examination, since he had no major complications. According to his statement, he had for two years after injury positive Lhermitte`s phenomenon (electrical sensation that runs down the back and into the

limbs by bending the head forward). In 2002, he sustained another head injury, again after he fell down during a football game. Again he had a temporary tetraparesis, but this time with vegetative disorders for 6 months (dyspnea, tachycardia) and this was diagnosed and treated as depression. In September 2015, he accidentally hit his head against the wall and he developed progressive tetraparesis (American Spinal Injury Association grade C - ASIA C). X-rays, computed tomography scans (CT scans) and magnetic resonance imaging (MRI) revealed pseudoarthrosis after C2 type 2 fracture with posterior and vertical dislocation of the tip of the dens with type 1 C1/C2 instability by Fielding and Hawkins [12] (Pictures 1, 2, 3, 4, 5). Neurological examination revealed hyper-reflexia in the upper extremities bilaterally and a positive Hoffman's sign bilaterally.



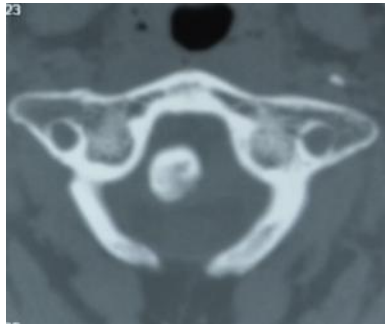
Picture 1. Preoperative X-ray



Picture 2a and 2b. Preoperative flexion/extension X-rays



Picture 3. Sagittal CT scan showed increased ADI (atlas-dens interval) and decreased SAC (space available for cord)



Picture 4. Axial CT scan revealed atlanto-axial dislocation



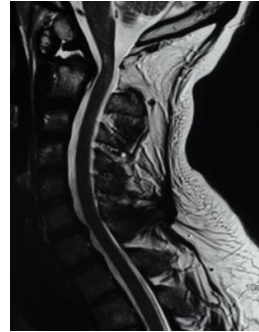
Picture 5. Preoperative coronal CT reconstruction shows nonunion of the odontoid

The patient underwent surgical treatment with C1 lateral mass and C2 pedicle screw (Harms), with

correction of C1/C2 instability and fusion with tricortical iliac graft (Pictures 7, 8).

After surgical treatment he improved his neurological condition, especially in hands, but he complains of a shoulder pain.

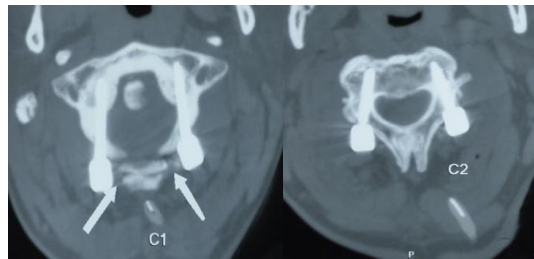
The preoperative neck pain measured by visual analog scale score (VAS) was $4,8 \pm 1,3$ and improved significantly to $1,9 \pm 0,9$ after surgery ($p < .001$).



Picture 6. Preoperative MRI shows compression of spinal cord



Picture 7. Postoperative x-ray with satisfactory position of the screws



Picture 8a and 8b. Postoperative CT scans shows correct screw position

3. DISCUSSION

Despite progressive improvement of diagnostic tools and surgical technique (anterior fixation and posterior fusion), the management of odontoid fractures (especially type II), remains a great challenge for spine surgeons. It is well known that odontoid fractures generally are not associated with neurologic deficit, hence the misdiagnosis can occur. Missed odontoid fracture leads to non-union and progressive myelopathy which require surgical decompression and fusion [13,14].

Koivikko revealed that factors associated with nonunion are fracture gap (> 1 mm), posterior displacement (> 5 mm), delayed start of treatment (> 4 days) and posterior re-displacement (> 2 mm) [15].

There is a great problem in the countries undergoing economy and cultural transition regarding patient compliance as we have shown in our case [16]. It is very surprising that the young sportsman did not undergo any medical examination, despite temporary tetraparesis.

On the other hand, we had the situation where patient underwent examination, but the proper diagnosis was not established and the patient had been treated as depression [9,17].

In our case we had a combination of odontoid fracture and atlanto-axial instability. Atlanto-axial junction is a very mobile joint and its stability is provided by several important ligaments: the transverse ligament, the apical ligament and alar ligament. The instability can be pure ligamentous or osteo-ligamentous. According to Dickmann, the integrity of transversal ligament is the key of the atlanto-axial stability [18].

Patient in our case had type II odontoid fracture (according to Anderson and D'Alonzo classification), and according to the literature this type of fractures is highly associated with non-union [19,20].

There are only few case reports similar to our case that we found in the literature. Moreau et al. achieved good stabilization by closed skeletal reduction and pedicle screw fixation [21]. Bu et al. also succeed to achieving good position by closed reduction [22]. Due to the fact that the symptoms in our patient began 16 years ago, we

did not try to perform closed reduction by skeletal traction, hence we performed open reduction and posterior stabilization. There are two main C1/C2 fusion techniques: trans-articular screw and screw-rod constructs. We performed screw-rod construct with C1 lateral mass screws and C2 pedicle screws [23-25]. We found in one meta-analysis slightly higher rates of fusion and less risk of injury to the vertebral artery in screw rod constructs [26].

We found an interesting case report where the patient underwent transoral resection of the odontoid and on the same day posterior occipito-cervical arthrodesis [27].

4. CONCLUSION

Nonunion and C1-C2 instability of odontoid fractures usually results from delayed diagnosis and inappropriate treatment. However, the available treatment options for odontoid fractures remain controversial. As in our case, diagnosis and treatment is usually initiated at the onset or progression of late neurologic symptoms. After the analysis of this case remain open questions regarding the classification of this injury pattern and whether adequate surgical treatment has been carried out.

CONSENT

As per international standard or university standard, patient's written consent has been collected and preserved by the authors.

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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