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Case Report

Unstable os odontoideum contributing to cervical myelopathy and obstructive sleep apnea

Abolfazl Rahimizadeh, Zahed Malekmohammadi, Mona Karimi, Ava Rahimizadeh, Naser Asgari

Pars Advanced and Minimally Invasive Medical Manners Research Center, Pars Hospital, Iran University of Medical Sciences, Tehran, Iran.

E-mail: *Abolfazl Rahimizadeh - a_rahimizadeh@hotmail.com; Zahed Malekmohammadi - dr.malekmohammadi@gmail.com; Mona Karimi - Kmmonik20@gmail.com; Ava Rahimizadeh - Ava.rahimizadeh@hotmail.com; Naser Asgari: nvasgari@gmail.com

***Corresponding author:**
Abolfazl Rahimizadeh,
Pars Advanced and Minimally Invasive Medical Manners Research Center, Num. 10, Rastak St., Keshawarz Boulevard, Tehran, Iran.

a_rahimizadeh@hotmail.com

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ABSTRACT

Background: Sleep apnea is characterized by repetitive cessation of breathing during sleep. It may be attributed to obstructive, central, or mixed pathologies close to the upper airway resulting in a decreased diameter of the oropharyngeal tract.

Case Description: A 36-year-old male with progressive cervicomedullary myelopathy/quadriparesis exhibited obstructive sleep apnea (OSA) attributed to an anteriorly displaced os odontoideum (OO). Atlantoaxial screw-rod stabilization resulted in improvement of both neurological function and OSA.

Conclusion: A symptomatic unstable OO may contribute to suboccipital pain, progressive quadriparesis, vertebrobasilar insufficiency, and OSA. Appropriate operative intervention utilizing atlantoaxial screw-rod stabilization may help to resolve these deficits.

Keywords: Cervical myelopathy, Obstructive sleep apnea, OS odontoideum

INTRODUCTION

Obstructive sleep apnea (OSA) is defined by momentary, cyclical cessation in breathing that is not severe enough to cause significant arterial hypoxemia and/or hypercapnia.[3,4] There are three categories of OSA: the obstructive type (e.g., OSA due to narrowing or a partial upper airway obstruction), the central subtype (CSA) secondary to cervicomedullary compromise (e.g., caused by deregulation of the respiratory center), and the mixed type (e.g., due to a combination of both OSA and CSA).[16]

Os odontoideum (OO), a traumatic or congenital abnormality of the second cervical vertebrae, is characterized by a separate bony segment with a smooth circumferential margin.[9,10,12] Here, the authors describe a 36-year-old male who developed OSA and a progressive quadriparesis attributed to an unstable OO. Following reduction and stabilization, both his quadriparesis and OSA improved.

CASE REPORT

A 36-year-old male exhibited a progressive spastic quadriparesis (hyperactive reflexes, positive Hoffmann signs, and an equivocal plantar reflex bilaterally) of 6-month duration. He also had developed OSA over the past 2 years, documented by two overnight polysomnography studies.

Radiographic confirmation of OO

The dynamic lateral cervical X-rays showed a free-floating OO that compromised the oropharyngeal airway [Figure 1b]. The cervical lordosis was 65 in extension with 27° of atlas angulation [Figure 1a]. With neck flexion, the cervical lordosis decreased to −5° and the atlas angulation to 15° [Figure 1b].

The cervical magnetic resonance imaging (MRI) showed atlantoaxial dislocation attributed to anterior displacement of the OO resulting in marked AP diameter canal narrowing at the cervicomedullary junction [Figure 2a]. The T2-weighted MRI showed a high intramedullary cord signal at the index level, consistent with myelomalacia versus edema [Figure 2b].

Computed tomography (CT) studies (coronal and sagittal images) also confirmed anterior displacement of the OO. The sagittal CT showed the characteristic jigsaw pattern [Figure 3a]. In addition, the area between the anteriorly displaced OO and the axis was occupied with soft connective tissues which, in combination with OO and the anterior atlas ring, were in close proximity to the upper airway tract. Further, the posterior airway space was about 4 mm [Figure 3b and c].

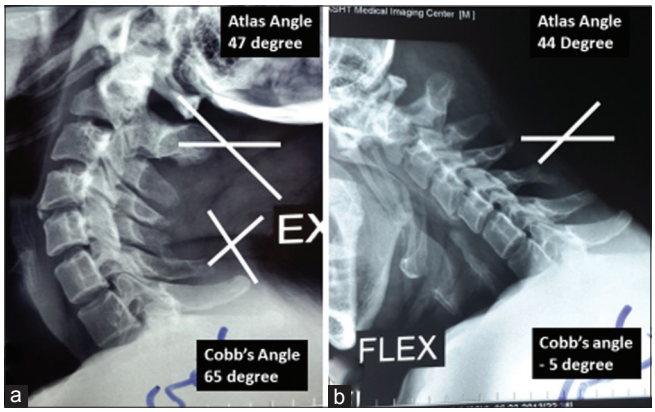

Figure 1: Dynamic lateral cervical radiographs: (a) in extension, Cobb and atlas angles are 65° and 47°, respectively. (b) In flexion, Cobb and atlas angles are decreased to −5° and 44°, respectively. Since atlas is loose with respect to axis, changes in atlas angle are invaluable in os odontoideum.

Figure 2: (a) T1-weighted sagittal magnetic resonance imaging (MRI) of cervical spine shows an os odontoideum (OO) with atlantoaxial dislocation and narrowing of the cervicomedullary junction. The OO and atlas ring are engulfed in a soft tissue extending from os to axis. The posterior airway space is quite narrow in the MRI taken in neuter position (white dash). (b) T2-weighted sagittal MRI shows myelopathy at the cervicomedullary junction.

Surgery

The patient underwent a C1-C2 posterior fusion [Figure 4a]. Postoperatively, his neurological examine markedly improved, and the OSA disappeared completely. Five years later, he still has experienced no recurrence of OSA [Figure 4b].

DISCUSSION

Frequency and etiology of OSA and rarity with OO

OSA is a relatively common but highly morbid condition that affects middle-aged adults (e.g., 9.1% of males and 4% of

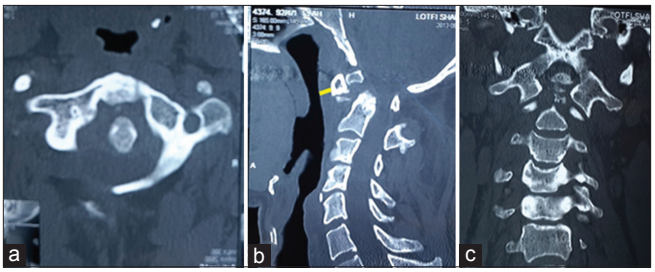

Figure 3: Computed tomography scan of the cervical spine, (a) axial view shows atlantoaxial dislocation. (b) Reconstructed sagittal view shows displaced os odontoideum (OO) - atlas ring forward displacement in jigsaw pattern; note posterior airway space is very narrow with the neck in neuter position. (c) Reconstructed coronal view shows the OO.

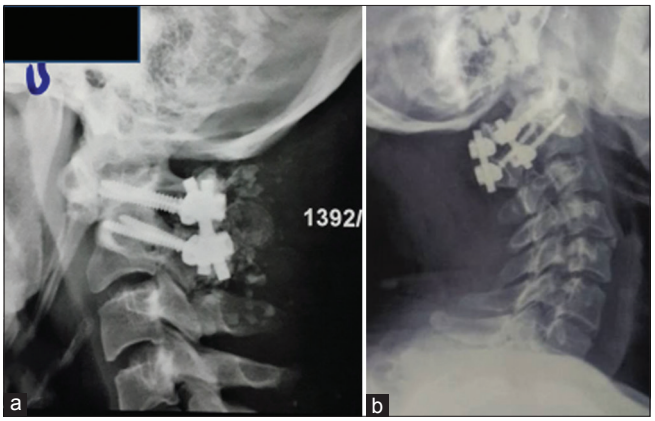

Figure 4: Postoperative lateral cervical X-ray shows C1-C2 screw rod fixation (a) a few days after surgery. (b) Five years after surgery.

females).[3,4,14] Intraluminal pathologies contributing to upper airway obstruction/OSA most typically include cancers of tongue base, pharynx or supraglottic larynx, an enlarged osteophyte at the level of C2-C3, and diffuse idiopathic skeletal hyperostosis/osteochondroma of the atlas.[2,5,6,8,11,13,15] Notably, OO is one of the least frequent causes of OSA; in fact, the authors could find only one previously reported example of this in the medical literature.[7]

Surgery for OO

The surgical management of unstable OO requires C1-C2 reduction and fusion/fixation best achieved with the classic Harms technique.[1] Alternative surgical options include C2-C1 transarticular fixation, application of a translaminar C2 screw instead of a C2 pedicle screw, and the use of an atlas hook instead of a C1 lateral mass screw.[9,10,12] Adequate management of instability with relief of C1-C2 cord/brain stem compression may allow for symptoms of OSA to resolve.

CONCLUSION

A symptomatic os odontoideum may contribute to suboccipital pain, progressive quadriparesis, vertebrobasilar insufficiency, and OSA. Appropriate operative intervention may help resolve these deficits.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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