

Retro-odontoid Synovial Cyst with Os Odontoideum and Atlantoaxial Instability

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We report the case of a patient with a retro-odontoid synovial cyst, coupled with os odontoideum and atlantoaxial instability. We observed the regression of the cyst after C1-2 transarticular screw fixation and posterior wiring without direct surgical excision of the cyst on the follow-up MR images, which were obtained two and three months postoperatively. The myelopathic symptoms and signs of this patient were resolved by degrees.

KEY WORDS : Synovial cyst · Os odontoideum · Atlantoaxial instability · C1-2 fixation · Spontaneous reduction.

Introduction

Intraspinous synovial cysts occurring in the C1-2 junction are extremely rare. Only 21 cases of synovial cysts have been reported to have occurred at the atlantoaxial joint^{1,4,7,8,17,18}. The symptoms of these patients were primarily attributed to compression of the spinal cord, as they complained of gait disturbance, paresthesia, pain in the neck or upper extremities, or motor weakness. The preferred option for the surgical resolution of synovial cysts located at the C1-2 junction is the direct excision of the cyst via transoral approach^{2,5,13}, posterolateral approach⁷, or laminectomy of the atlas^{1,2,8,15,19}. Here, we present a case of an intraspinal synovial cyst of the C1-2 junction, in a patient exhibiting os odontoideum and atlantoaxial instability.

Case Report

A 46-year old woman presented with posterior neck pain, paresthesia, and motor weakness in both hands, which had persisted for a period of several years. The patient's paresthesia and motor weakness had become aggravated over the recent months, but she had no definite history of trauma. The neu-

rological examination revealed hypesthesia on the C4, C5, and C6 dermatomes, as well as reduced muscle strength in both hands. The patient also complained of a mild gait imbalance.

Plain radiographs of the cervical spine indicated os odontoideum and C1-C2 instability (Fig. 1). Magnetic resonance (MR) images revealed a cyst which compressed the spinal cord, and was located on the dorsal side of the transverse atlantal ligament. These images also indicated a round cystic mass, which exhibited low signal intensity on T1-weighted images, high signal intensity on T2-weighted images, and peripheral rim enhancement on contrast enhanced T1-weighted images (Fig. 2).

We conducted the fixation and fusion via the C1-C2 transarticular screw and posterior wiring method with autologous bone.

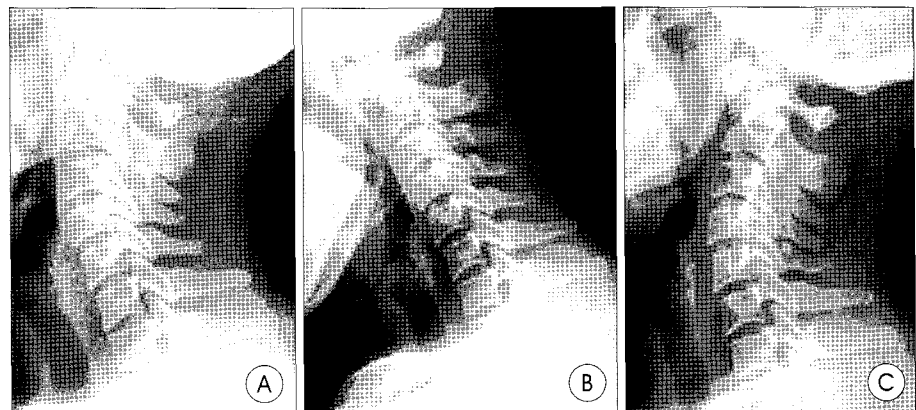


Fig. 1. Plain radiographs of the cervical spine indicating os odontoideum. Flexion (B) and extension (C) lateral radiographs of the cervical spine revealing atlantoaxial instability.

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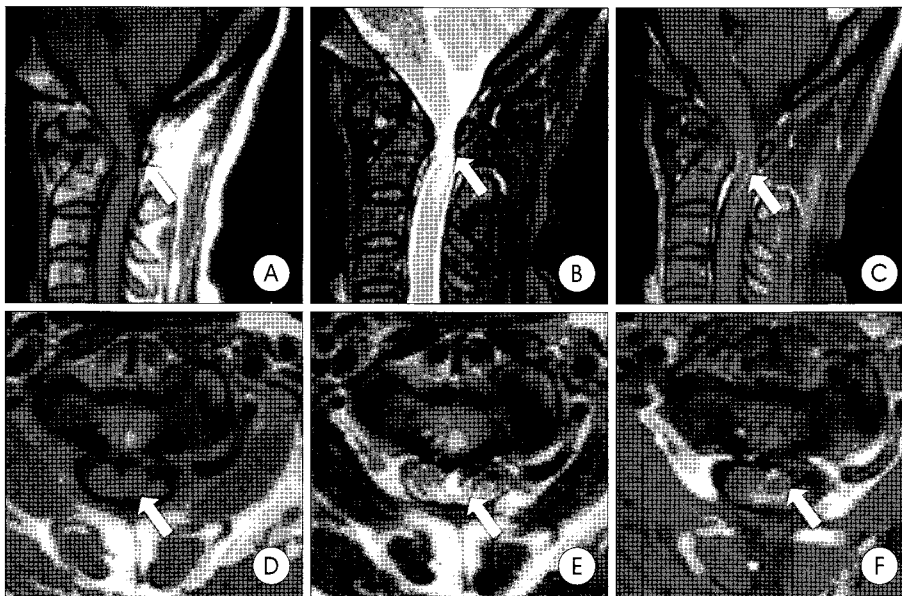


Fig. 2. A : Sagittal T1-weighted image indicating a round cystic mass (arrow) with low signal intensity, located at the transverse atlantal ligament. B : Sagittal T2-weighted image indicating a round cystic mass with high signal intensity. C : Contrast enhanced T1-weighted image indicating a peripheral rim enhanced cystic mass. D-F : Corresponding axial image revealing the same round cyst, compressing the spinal cord, which located at the transverse atlantal ligament.

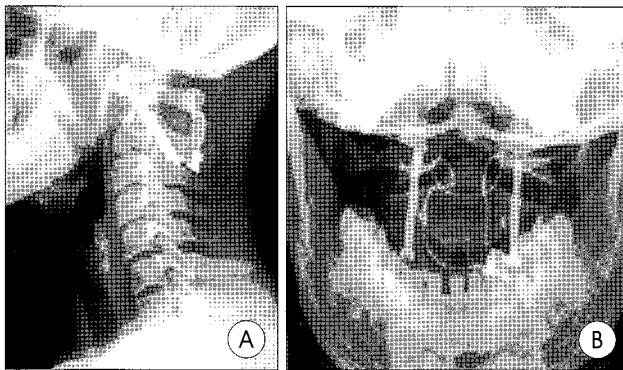


Fig. 3. Lateral (A) and open-mouth (B) radiographs of the cervical spine indicating solid fusion with transarticular screw fixation and posterior C1-2 wiring.

The MR images were obtained two and three months after the operation, in order to observe any changes in the cyst.

Three months after operation, we located a solid fusion appearing on the plain radiographs (Fig. 3). Follow-up MR images evidenced a spontaneous reduction in the size of the cyst (Fig. 4). The myelopathic symptoms and signs of the patient were then resolved by degrees. Neck disability index (NDI) improved from a preoperation value of 48, to 31 at two months and 29 at three months postoperation. The visual analog scale (VAS) also indicated pain reduction, with corresponding values of 5, 2, and 2.

Discussion

Synovial cysts are frequently observed disease of human joints, and they are often reported in the spine, particularly within

the lumbar spine. Spinal synovial cysts tend to be located principally in the spinal canal, adjacent to the facet joints of the lumbar spine^{9,14,16}. However, they only quite rarely occur in the cervical spinal canal^{3,10}. Only 21 cases of synovial cysts occurring at the atlantoaxial joint have been reported^{1,4,7,8,17,18}.

The pathogenesis of synovial cysts remains fairly unclear, but a host of theories have been advanced to elucidate the causes of spinal synovial cysts, such as degenerative changes in the facet joints, trauma, os odontoideum, metaplasia, the presence of developmental rests, excess stress inflicted at the facet joints coupled with the herniation of synovial tissue, and mucinous degeneration in the connective tissues^{15,20}. In some reports,

trauma has been theorized to result in cyst enlargement or hemorrhage into the synovial cyst cavity, which results in epidural compression of the spinal cord^{10,12}. In this case, the os odontoideum with atlantoaxial instability may constitute the underlying cause of the formation of the synovial cyst. The symptoms of such patients are primarily attributed to compression of the spinal cord, and they tend to complain of such symptoms as gait disturbance, paresthesia, pain in the neck or in the upper extremities, or motor weakness. In our case, the patient complained of posterior neck pain, paresthesia, motor weakness in both hands, and a mild gait imbalance.

Magnetic resonance (MR) imaging is the examination of choice for these synovial cysts. Cervical synovial cysts are generally reported to exhibit signal characteristics similar to those of a typical cyst, i.e., low to intermediate signal intensity on T1-weighted images, and high signal intensity on T2-weighted images. However, these cysts also sometimes display low signal intensity on T2-weighted images, which is generally attributed to hemorrhage and blood products within the cyst contents, or to calcium deposits^{11,21}.

Several options have been reported for the surgical treatment of synovial cysts at the C1-2 junction, and most of these involve the direct excision of the cyst, via the transoral approach^{2,5,13}, posterolateral approach⁷, or by laminectomy of the atlas^{1,2,5,8,15}.

Although satisfactory results have been reported, in which no further neurological injury resulted, the direct excision of the cyst, regardless of approach, is considered to be a dangerous procedure, and certainly carries with it the risk of serious neurological complications.

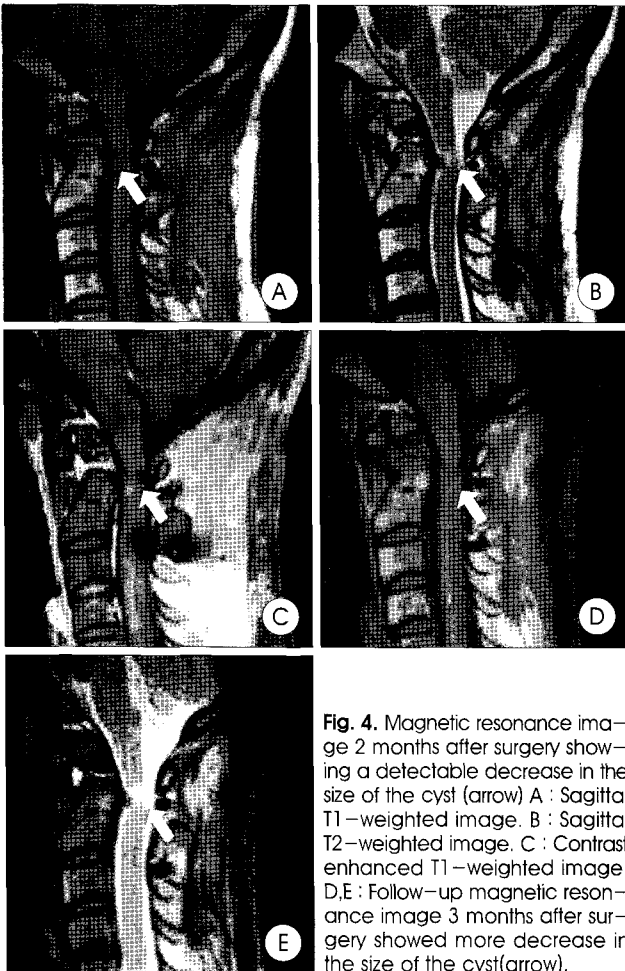


Fig. 4. Magnetic resonance image 2 months after surgery showing a detectable decrease in the size of the cyst (arrow) A : Sagittal T1-weighted image. B : Sagittal T2-weighted image. C : Contrast enhanced T1-weighted image. D,E : Follow-up magnetic resonance image 3 months after surgery showed more decrease in the size of the cyst (arrow).

Recently, C1-2 posterior fusion alone has been determined to result in the resorption of the synovial cyst, Chang⁴⁾ and Morio have associated this with clinical improvements¹⁷⁾. Doh et al.⁶⁾ reported that transarticular screw fixation was a superior surgical option for the management of atlantoaxial instability, with regard to both reduction loss and fusion rate. Therefore, we elected to use transarticular screw fixation and posterior wiring techniques, for better reduction and biomechanical stability. This may have caused the synovial cyst to shrink, or led to the resorption of the cyst. At two and three months after operation, the patient's paresthesia and myelopathic symptoms had improved, and we observed a reduction in the size of the synovial cyst on the follow-up MR images. We also observed the absence of C1-2 instability on the follow-up dynamic plain radiographs although the left screw was incomplete.

Conclusion

Retro-odontoid synovial cyst with atlantoaxial instability is an extremely rare condition, and many surgical options

exist for the treatment of such cysts. Among these options, we opted to use transarticular screw fixation and posterior C1-2 wiring techniques, without the direct surgical excision of the cyst, and achieved a spontaneous reduction in the size of the cyst, as well as marked clinical improvement.

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