Case Report

Complete Spontaneous Regression of the Intraspinal Synovial Cyst at the C1-C2 Junction Following with Atlantoaxial Fusion of Non-Union Odontoid Fracture: A Case Report

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The author reported an extremely rare case of intraspinal synovial cyst at C1-C2 junction, located posteriorly to the odontoid process, compressing the upper cervical spinal cord. The patient had a history of spinal trauma 40 years ago without prior definitive treatment, presenting with two months of left leg and arm numbness with neck pain. Imaging studies including plain radiographs, computed tomography (CT), and magnetic resonance imaging (MRI) of the cervical spine showed a large synovial cyst associated with atlantoaxial instability from non-union fracture of the odontoid process. The atlantoaxial fusion was performed without a direct excision of the synovial cyst, the patient showed significant improvement of neck pain and numbness of all extremities. In addition to the fracture stabilization, the follow-up post operative MRI revealed spontaneous regression of the intraspinal synovial cyst after atlantoaxial fusion alone.

Keywords: *Synovial cyst, Atlantoaxial instability, Odontoid fracture*

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The intraspinal synovial cyst at the C1-C2 junction is extremely rare condition[1-7]. Many reports used the terms 'ganglion cyst'[8-11] or 'articular cyst'[7] and most of them were related to degeneration of the C1-C2 joints[3,5,7,9,10,12]. Later publications frequently reported the cases of synovial cyst-associating with the instability of atlantoaxial joints[1-3,6,11,14,15] and treating with variety of surgical options, including lateral atlantoaxial joint puncture alone[14], cystic excision with atlantoaxial fusion[1,15] and atlantoaxial fusion alone[2,3,6]. The current report presented a case of an intraspinal cyst at the C1-C2 junction connecting to atlantoaxial facet joint and atlantodental synovial cyst. The cyst associated with atlantoaxial instability was spontaneously regressed after atlantoaxial fusion without manipulation of the cyst.

Case Report

A 68-year old man presented with two-month numbness of left arm and leg and occasional morning neck pain. The numbness had progressed to the right hand and arm within a month; nevertheless, there were no other symptoms of spinal cord compression syndrome. The patient had a history of sudden paralysis from a car accident 40 years ago but did not undergo spinal imaging due to the complete recovery of weakness within a few days.

A neurological examination revealed only slightly increased deep tendon reflex, but the muscle tone and strength of all extremities were normal. In addition to normal touch and pinprick sensation, the neck active range of movements was without any limitations.

Radiographic films showed a non-union odontoid fracture type II on open-mouth film, and atlantoaxial instability on flexion-extension lateral cervical spine films. Magnetic resonance imaging (MRI) of the cervical spine showed a large cystic mass which compressed the spinal cord, located behind vertebral body of the axis, just below the level of fracture odontoid process (Fig. 1). The images showed iso-intensitysignal on T1-weighted image, high signal intensity on T2-weighted image in the lesion, and rim enhancement around the lesion on gadolinium enhanced T1-weighted

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image. The upper part of the intraspinal cyst continued with the synovial cyst of left atlantoaxial facet joint and also communicated to the atlantodental synovial cyst through the non-union odontoid fracture (Fig. 2). The patient's diagnosis was a non-union odontoid fracture associated with intraspinal synovial cyst and severe cord compression. Due to the very mild symptom of clinical cord compression syndrome, the author planned to perform a surgical fusion of the atlantoaxial joint without resection the cystic mass or spinal cord decompression.

The pre-operative surgical planning with computed tomography (CT) showed a corticalised fracture of the odontoid process, very small left pedicle of the axis from very high ridding of left vertebral artery (Fig 3). The author, therefore, decided to perform

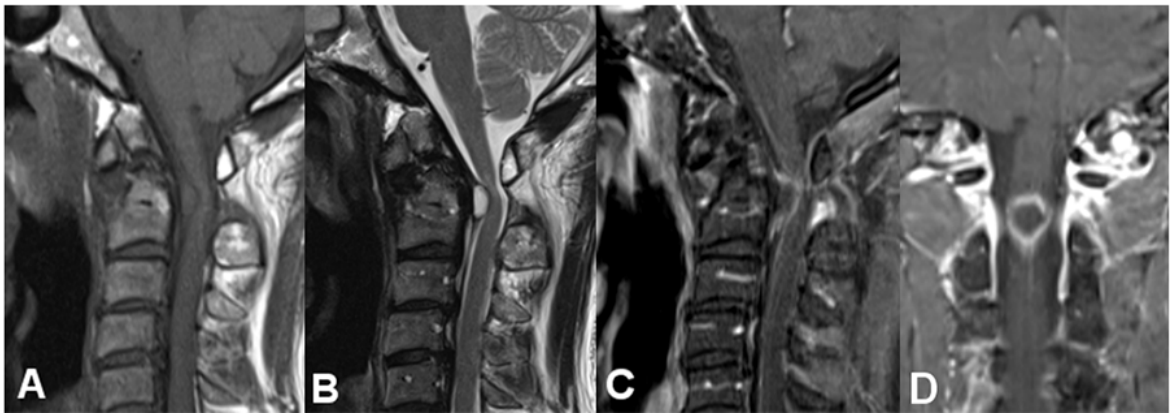

Fig. 1 Mid-sagittal and coronal magnetic resonance images (MRI) revealed a large intraspinal cyst mass of C1-2 junction with severe cord compression. A) Sagittal T1-weighted image showed iso-intensity signal in the lesion. B) Sagittal T2-weighted image revealed high signal intensity in the cystic mass. C & D) Sagittal and coronal post-gadolinium T1-weighted image displayed rim enhancement surround the cyst.

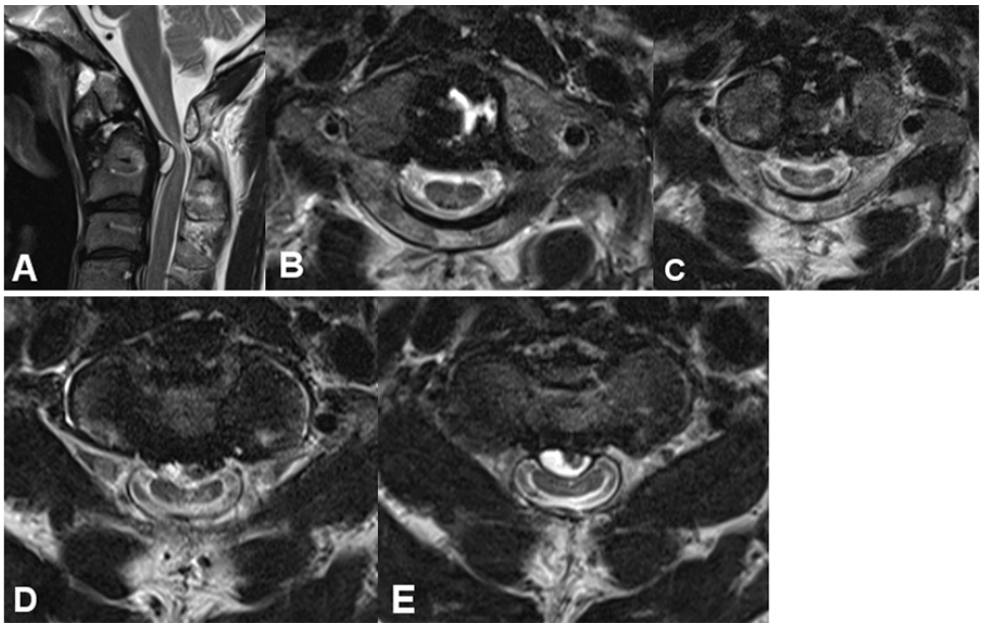

Fig. 2 Slightly left side off mid-sagittal T2-weighted image (A), and axial T2-weighted images (B-E) showed intraspinal cyst connected together with atlantodental cyst through the nonunion odontoid fracture and synovial cyst of left atlantoaxial facet joint.

posterior atlantoaxial fixation with lateral mass C1, translaminar C2 screw fixation technique (S4®Cervical System, B Braun) to avoid the risk of left vertebral artery injury, combining with posterior spinal fusion (Fig. 4).

The numbness of arms and legs was immediately improved on the day of surgical fixation, and the patient was discharged from the hospital on the fourth Postoperative day with full neurological

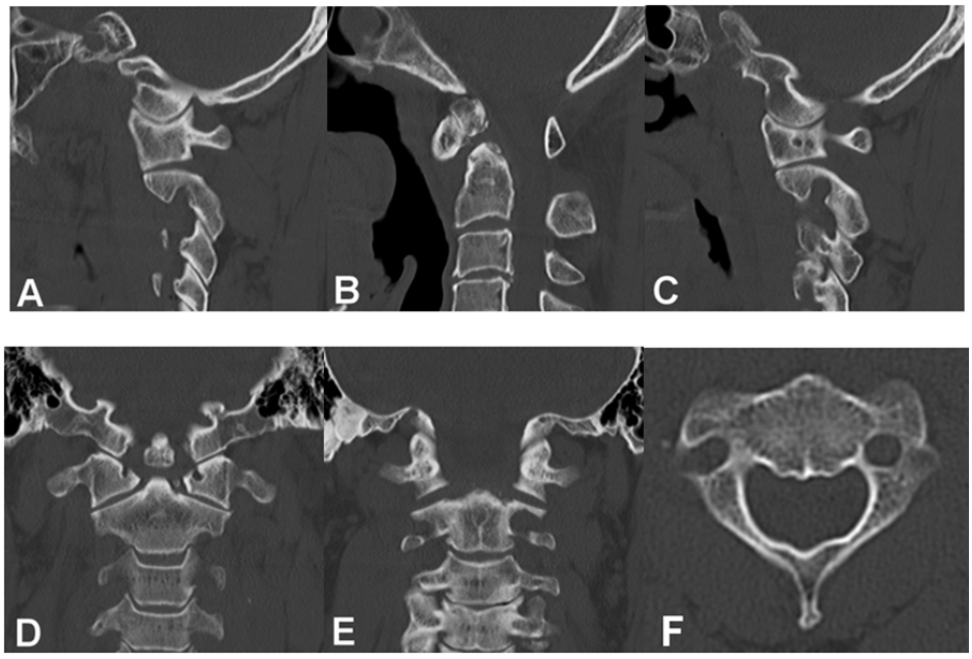

Fig. 3 Bone window computed tomography (CT) revealed corticalised odontoid fracture on mid-sagittal (B) and coronal (D) views. A, Sagittal CT showed slightly small size of the right pars. C, Sagittal CT and E, Coronal CT showed very thin of left pars from medial located and high ridding of left vertebral artery. F, Axial CT of the axis showed very short and narrow of left pedicle from medial located of vertebral artery.

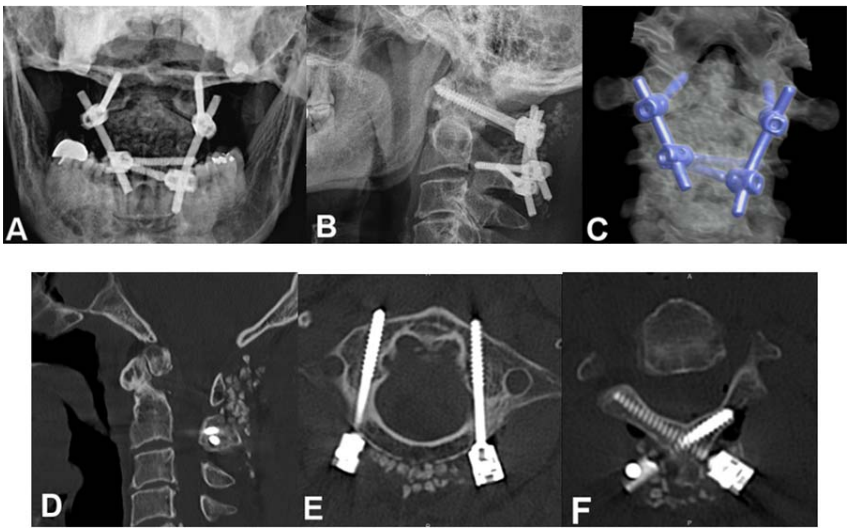

Fig. 4 Postoperative atlantoaxial fusion, open mouth (A), lateral cervical spine (B) radiographs, 3D-CT (C), and Sagittal CT scan (D) demonstrated the polyaxial screw and rod system fixed lateral mass C1 screws (E) to translaminar C2 screws (F) in proper sagittal and coronal alignment combined with posterior spinal fusion.

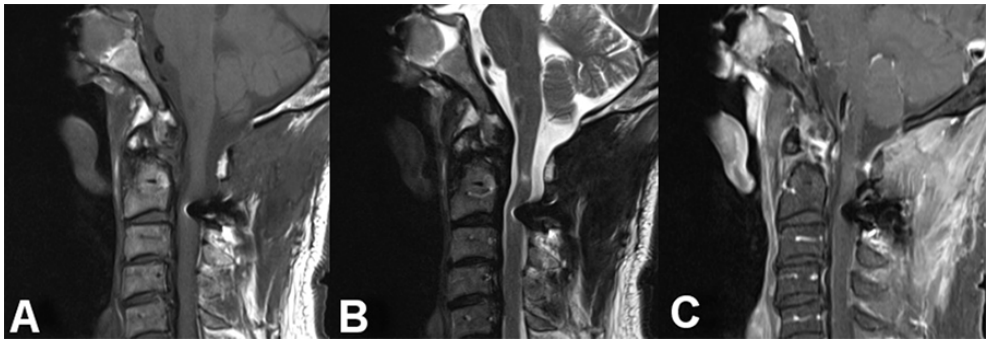

Fig. 5 The 5th month-postoperative MRI, Sagittal T1-weighted (A), T2-weighted (B), and post-gadolinium T1-weighted (C) images showed disappear of the intraspinal synovial cyst at C1-C2 junction.

function. However, the patient had a routine follow-up of clinical, radiographs and MRI to evaluate the instrumentation, fusion condition and the size of synovial cyst. On the five-month-MRI revealed complete spontaneous regression of intraspinal synovial cyst at C1-C2 junction (Fig. 5) and CSF was around the spinal cord in the area of pevious cord compression.

Discussion

The intraspinal synovial cysts of the cervical spine mainly arise from cervical facet joint in the lower cervical spine[12,16,17] and transverse (quadrate) ligament of the odontoid process at the C1-C2 junction or atlantodental joint[2,3,5,7,9,14,15]. Moreover, only a few reports could demonstrate the cyst from atlantodental joint communicated to lateral atlantoaxial joint[14] and facet joint[6,18] corresponding to the current case report. The pathogenesis of synovial cyst of previous reports can be divided into two categories: the first category is degenerative change of atlantoaxial joint[5,7,9,10,13,19], and the other is instability of atlantoaxial joint[1-4,6,11,14,15,18].

Some authors[7,9] proposed to use the term "atlantoaxial degenerative articular cyst" instead of "synovial cyst" at the C1-C2 junction for degenerative category, because the cyst has an anatomical connection with articular structure and the pathogenesis is related to degenerative changes of the atlantodental joint[7,9]. Whenever the clinical of spinal cord compression from degenerative articular cyst is expressed, the cystic mass is inevitable to resect or decompress. There were a few cases operated via a transoral approach to remove the cyst followed with spinal instrumentation and fusion[7,10]. However, most of the authors preferred a posterolateral approach or laminectomy C1-C2 with transdural resection[5,7,9,13,18,19] in order to avoid an operation of spinal fusion, in which this technique also had favorable outcomes.

For another category of pathogenesis of synovial cyst, the term of intraspinal synovial cyst at C1-C2 junction is replaced by retro-odontoid synovial cyst[3,14] for the cystic mass that associated with old fracture odontoid process or os odontoideum with atlantoaxial instability. Many treatment options were reported with successful outcomes, including conservative treatment with Philadelphia collar[11], aspiration of lateral atlantoaxial joint without spinal fusion[14], different methods to excision or decompress the cyst followed by atlantoaxial fusion[1,4,15], and atlantoaxial fusion alone[2,3,6].

Almost all of the previous reports were direct excision of the cyst through transoral[7,10], posterolateral approach[7,13] or laminectomy C1-C2[5,9]. Since the first report of atlantoaxial fusion alone by Chang et al[2] in 2000 showed spontaneous regression of the cyst, many reports were later frequently published[3,4,6]. However, the surgical resection or decompression of the cyst followed with C1-C2 fusion was also reported due to concerns over the long cyst-spontaneous-regression period[1,14,15].

In the current case with the very mild symptoms and non-urgent conditions for spinal cord decompression, rather than performing atlantoaxial fusion for non-union fracture odontoid process, the patient was treated with spinal fusion by avoiding cystic removal. Although surgical anatomy was difficult to apply instrumentation, the author denied occipitocervical fusion to preserve motion function of occiput-C1 joint. The polyaxial screw-rod system was fixed to lateral mass C1 and laminar C2 because of two reasons; first, the C2 laminar screw is superior strength than C2 pars screw and the second reason is C2 laminar screw minimizes the vertebral artery injury[20] than

pedicle or pars screw insertion. The surgical fixation and fusion achieved the satisfied instrumentation and immediate, clinical improvement, and the post operative MRI was postponed to the fifth month. The MRI showed disappearance of the intraspinal synovial cyst of the C1-C2 junction, consistently with the concept of "atlantoaxial fusional one is a successful method for spontaneous regression of the intraspinal synovial cyst-associated with atlantoaxial instability".

Conclusion

The intraspinal synovial cyst of the C1-C2 junction associated with atlantoaxial instability with mild clinical symptoms of spinal cord compression can be treated with atlantoaxial fusion alone. The spontaneous regression of the cyst and clinical improvement were achieved by atlantoaxial joint stabilization. However, the clinical follow-up and post operative MRI are very important and necessary to evaluate the post operative successful outcomes.

What is already known on this topic?

The surgical management of the intraspinal synovial cyst at C1-C2 junction from degenerative change of atlantoaxial joint is the direct excision of the cyst. The cyst-associated with atlantoaxial instability has been treated successfully by the atlantoaxial fusion alone, whereas many authors are concerned about the spontaneous regression period of the cyst who, therefore, prefer a direct cystic resection or decompression following with C1-C2 fusion.

What this study adds?

This study reported the intraspinal synovial cyst at C1-C2 junction or retro-odontoid cyst communicated to the cyst from atlantodental joint via lateral atlantoaxial joint and the synovial cyst from facet joint in the case of non-union fracture odontoid process. This study also demonstrates a successful method for spontaneous regression of the intraspinal synovial cyst without direct decompression of the cyst by the atlantoaxial fusion using the lateral mass screw and rod system fixation.

Potential conflicts of interest

None.

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ถุงน้ำจากไขข้อกระดูกสันหลังที่คอข้อที่ 1 และ 2 ภายในโพรงกระดูกสันหลังยุบหายเองได้ในผู้ป่วยกระดูกสันหลัง odontoid process หักที่ได้รับการผ่าตัดเชื่อมกระดูกสันหลัง

ธีระ ตั้งวิริยะไพบูลย์

รายงานผู้ป่วยรายนี้เป็นผู้ป่วยที่เคยได้รับอุบัติเหตุกระดูกสันหลังที่คอประมาณ 40 ปีก่อน ไม่เคยได้รับวินิจฉัยและผ่าตัดรักษามาก่อน มาตรวจด้วยอาการปวดต้นคอและแขนขาซ้ายชาเป็นเวลา 2 เดือน จากการตรวจพิเศษทางรังสีวิทยา ตรวจพบถุงน้ำจากไขข้อในโพรงกระดูกสันหลังที่เกิดจากกระดูกสันหลังที่คอข้อที่ 1 และ 2 ซึ่งเป็นกรณีที่พบได้ยากมาก ๆ และกดไขสันหลังจากทางด้านหน้าร่วมกับมีภาวะกระดูกสันหลัง odontoid process หักเก่าที่ยังไม่ติดกันและมีภาวะไม่มั่นคงของกระดูกสันหลังข้อที่ 1 และ 2 ผู้ป่วยได้รับการรักษาด้วยวิธีการ ผ่าตัดยึดกระดูกสันหลังข้อที่ 1 และ 2 ด้วยโลหะ ร่วมกับการเชื่อมกระดูกสันหลังโดยไม่ได้ผ่าตัดเอาถุงน้ำออกโดยตรง ผลการรักษาดีมากทั้งในแง่อาการปวดต้นคอ การชา และถุงน้ำที่กดไขสันหลังยุบหายเองได้ สรุปได้ว่าการผ่าตัดแก้ไขสาเหตุภาวะไม่มั่นคงของกระดูกสันหลังข้อที่ 1 และ 2 สามารถรักษาภาวะถุงน้ำจากไขข้อกระดูกสันหลังที่คอข้อที่ 1 และ 2 ภายในโพรงกระดูกสันหลังโดยไม่จำเป็นต้องผ่าตัดเอาถุงน้ำออกโดยตรง