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CASE REPORT

Retro-odontoid synovial cyst resected via an anterolateral approach without fusion

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Abstract

Purpose Retro-odontoid synovial cysts are rare and attributable to degenerative changes in the atlantoaxial joints. An anterolateral approach facilitates access to lesions located anterior to the craniocervical junction without harming the atlantoaxial joints, and can also treat small lesions in the ventral mid-portion of the craniocervical junction without compression of spinal cord.

Methods We present herein the case of a 70-year-old man with a retro-odontoid synovial cyst. A ventral midsection mass was present at the level of the atlantoaxial joint. The compressed anterior medulla led to neurological deficits. Slight atlantoaxial instability was radiologically present. An intradural cyst resection without fusion was performed via the anterolateral approach. The diagnosis of a synovial cyst was histologically confirmed.

Results The patient was followed up for 3 years and exhibited improvements in the neurological deficits. There were no recurrence and postoperative deterioration of atlantoaxial instability.

Conclusions The anterolateral approach for the retro-odontoid synovial cyst had little effect on C1–2 instability and yielded neurological improvements.

Keywords C1–2 · Synovial cyst · Anterolateral approach · Atlantoaxial instability

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Introduction

Retro-odontoid synovial cysts are rare, can be attributed to degenerative changes in the atlantoaxial joints, and tend to affect older populations. The natural course of a retro-odontoid synovial cyst remains unknown, although clinical deterioration has been reported over periods of weeks to months [1–13]. Treatments for retro-odontoid synovial cysts include transoral resection with posterior fusion, posterior resection with or without posterior fusion, posterior fusion only, and aspiration; however, the ideal treatment method remains unknown [1–19].

Motion at the C1–2 articulation plays a role in cyst formation and growth [20]. Although atlantoaxial joint instability appears to be a causative factor, there have been no reports of a relationship between typical atlantoaxial instability between the flexion and extension positions and retro-odontoid synovial cysts. An anterolateral approach facilitates access to lesions located anterior to the craniocervical junction without harming the atlantoaxial joints [21–23]. An anterolateral approach can also be used to treat small lesions in the ventral mid-portion of the craniocervical junction without spinal cord compression.

We report herein the resection of a retro-odontoid synovial cyst via the anterolateral approach without fusion, which yielded neurological improvements and no deterioration of the atlantoaxial instability during a 3 year follow-up period.

Case presentation

A 70-year-old man who had gradually developed a gait disturbance was referred to our hospital. Neurological examinations revealed myelopathy in his right upper

Fig. 1 Cervical roentgenograms at the flexion (**a**) and extension positions (**b**). The mass presented in the ventral midsection at the C1–2 level. The spinal cord was severely dorsally compressed (**c**). T2-weighted imaging revealed hyperintense right intramedullary signals at the C1–2 level (**d**)

extremity and exaggerated deep tendon reflexes in his right upper and bilateral lower extremities. His cervical spondylosis severity score, which was evaluated using the Japan Orthopedic Association (JOA) scoring system, was 10. Routine laboratory examinations yielded normal results. The patient was non-diabetic and non-rheumatoid.

A cervical roentgenogram revealed an almost normal lordotic alignment. The atlanto-dens intervals were 3.0 mm at flexion and 2.0 mm at extension (Fig. 1a, b). Slight atlantoaxial instability was radiologically present. Computed tomography (CT) angiography revealed a normal vertebral artery. Magnetic resonance imaging (MRI) revealed a mass in the ventral midsection at the atlantoaxial joint level and canal stenosis at the C5–C6 level. The mass exhibited isointensity on T1-weighted images (T1WI), hyperintensity on T2-weighted images (T2WI), and slight rim enhancement. The spinal cord was severely dorsally compressed at the C1–C2 level in the midline (Fig. 1c). T2WI revealed hyperintense right intramedullary signals at the C1–C2 level (Fig. 1d).

Surgical procedure and pathological findings

Our routine anterior approach was via the right side. The patient was placed in the lateral position with his neck extended and chin turned 10° leftward. The head vertex was slightly downward. A curved skin incision was made transversely over the proximal sternocleidomastoid (SCM) muscle, distally along the anterior border of this muscle,

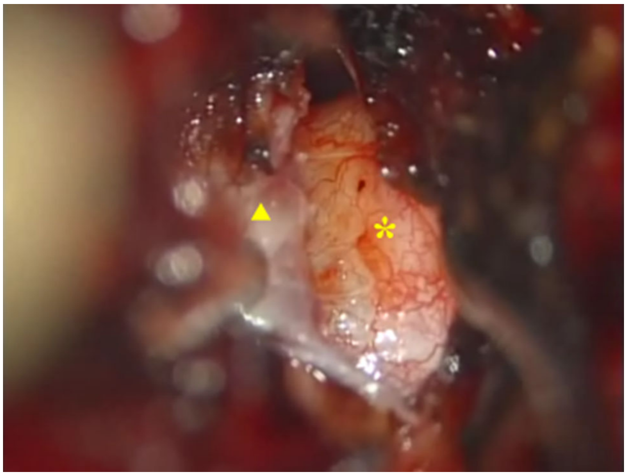

Fig. 2 Intraoperative photograph. The dura (*triangle*) was opened and an inward bulging of the ventral dura was identified as a synovial cyst (*asterisk*)

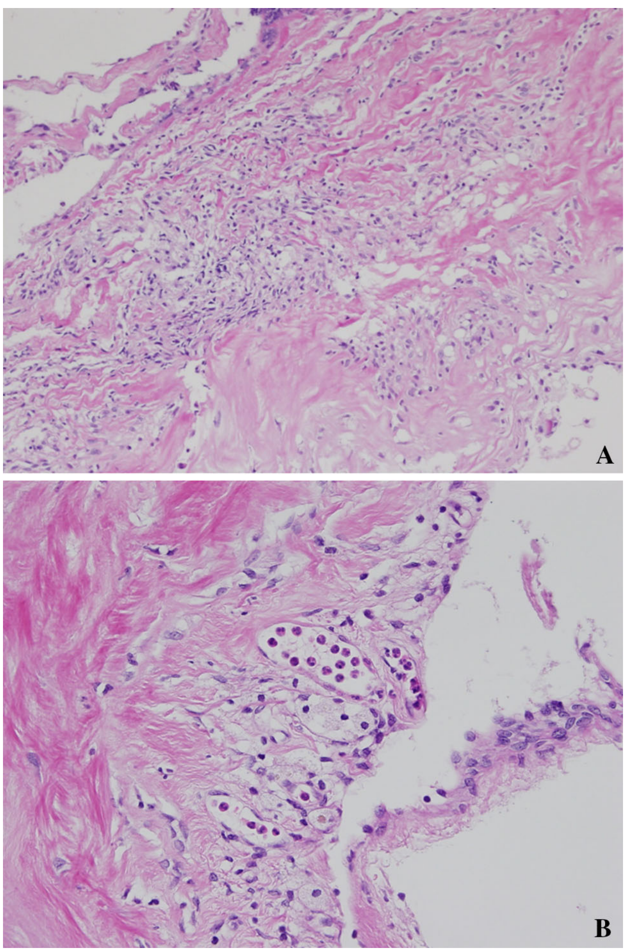

Fig. 3 Hematoxylin and eosin staining revealed a lining of synovial cells in the fibrocartilaginous capsule. **a** Lower magnification; **b** higher magnification

and posteriorly to curve across the mastoid process. We dissected medially to the SCM muscle and laterally to the carotid sheath. The greater auricular nerve underneath the platysma muscle was dissected for immobilization. The insertions of the SCM muscle and the splenius capitis muscle were divided at the base of the skull. We could identify the internal jugular vein and spinal accessory nerve. The tip of the transverse process of C1 could be identified by palpation. After incising the deep fascia from the tip of the C1 transverse process, we could identify the suboccipital triangle.

To expose the vertebral artery and C1, the superior oblique muscle, rectus capitis posterior major muscle, and inferior oblique muscle were divided from the attachment of the C1 transverse process and the occipital bone. After the vertebral artery sheath was exposed in the C1–C2 portion and above C1, the C1 posterior arch and C1 lateral mass were partially drilled and resected. The C2 nerve root that crossed laterally to the vertebral artery was cut. Venous bleeding was controlled with direct bipolar coagulation and hemostats. Vertebral artery transposition was not performed. Using microscopy, we opened the right lateral side of the dura mater and identified an inward bulging of the ventral dura at the anterior aspect of the spinal cord (Fig. 2). This round cystic mass, which contained mucinous material, was resected. Other than the lesion, there was no communication to the extradural space between the tectorial membrane and dura mater. A histological examination detected a lining of synovial cells in the fibrocartilaginous capsule (Fig. 3a, b). The dura was closed with a GORE-TEX suture and patched with muscle pieces and fibrin glue.

Postoperative course

The patient wore a Philadelphia collar for 1 week, followed by a soft neck collar for another week. There was no postoperative cerebrospinal fluid leakage. A postoperative neurological examination revealed improvements in the gait disturbance and myelopathy. The patient had no C2 numbness or neck pain. There were no major or minor complications. The patient received a JOA score of 15. Postoperative CT revealed the drilled and resected lateral part of the posterior atlas arch (Fig. 4c). The postoperative cervical roentgenogram revealed an almost normal lordotic alignment and no deterioration of the atlantoaxial instability after a 3-year follow-up (Fig. 4a, b). MRI revealed the decompressed mass in the ventral midsection at the atlantoaxial joint level at 3 years postoperation (Fig. 4d).

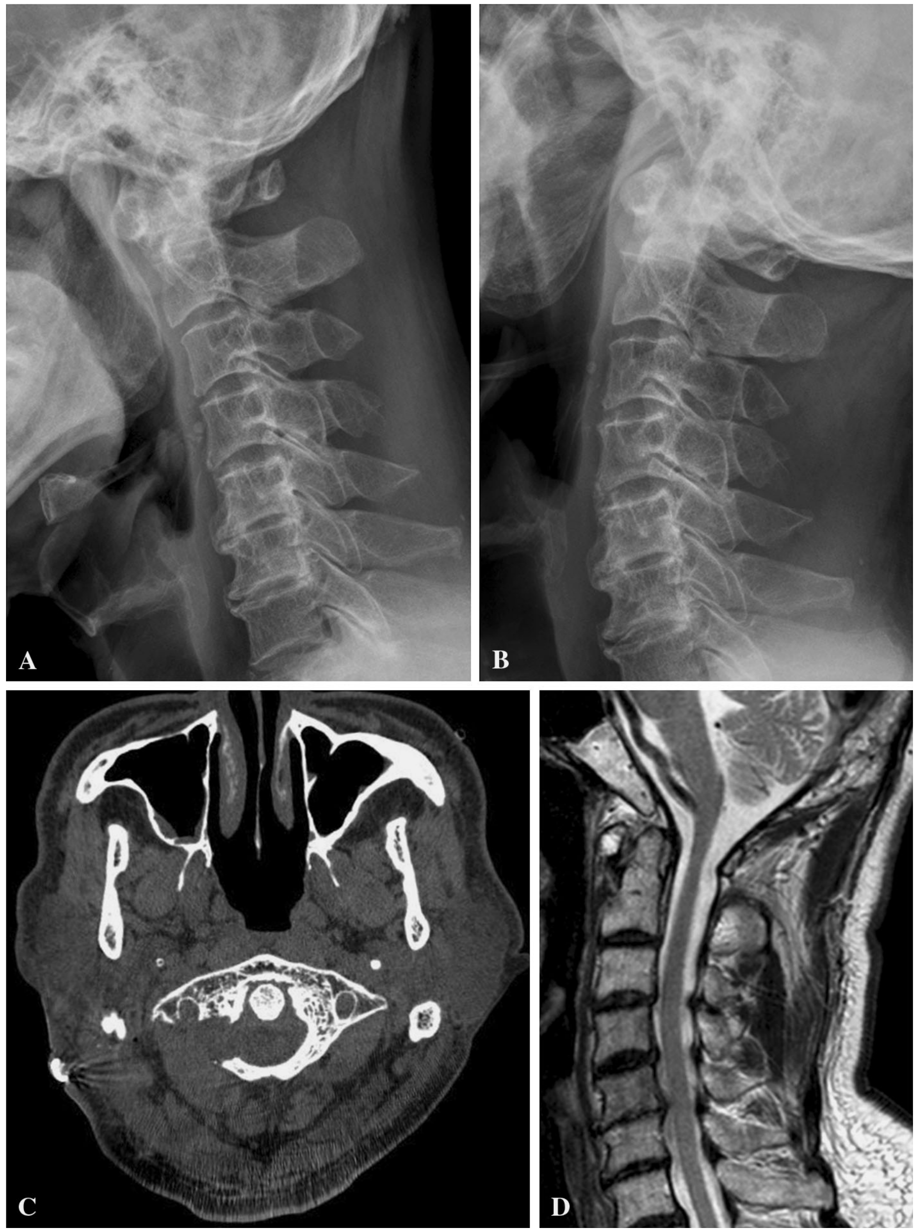

Fig. 4 Postoperative cervical roentgenograms revealed no deterioration of atlantoaxial instability during a 3-year follow-up period (**a**, **b**). Postoperative computed tomography revealed the resected lateral part of the posterior atlas arch (**c**). Magnetic resonance imaging revealed the decompressed mass in the ventral midsection at the C1–2 level at 3-year post-operation (**d**)

Discussion

Retro-odontoid synovial cysts are rare; to date, there have been 13 histologically confirmed reports of these lesions [1–13] (Table 1). The mean age of the affected patients was 71.9 ± 8.9 years (range 51–85 years), and the population included 12 men and 13 women. Thirteen patients were treated via a transoral approach with posterior fusion. One patient was treated via a transoral approach only. Eleven patients were treated via a posterior approach. There have also been five histologically non-confirmed reports of retro-odontoid synovial cysts [14, 18, 19, 24, 25]. Two and three patients were treated via the posterolateral approach and with conservative therapy, respectively. One patient underwent C1–2 fusion.

Motion of the atlantoaxial articulation has been suggested to play an important role in cyst formation and growth. C1–2 posterior fusion studies have demonstrated regression in cases of both C1–2 small and focal synovial cysts and C1–2 cystic deterioration [6, 14, 20]. In contrast, posterior fusion requires specific instrumentation, decreases the neck rotation range, and introduces the possibility of progressive upper cervical compression.

A transoral approach subsequent to posterior fusion is a reasonable approach for large C1–2 synovial cysts. Transoral decompression of C1–2 synovial cysts was

Table 1 Clinical data of 13 histologically confirmed reports of retro-odontoid synovial cysts

Author (year)	Age (sex)	Symptoms	Operations
Onofrio and Mih [9]	73 (M)	Arm weakness	Occipital craniotomy, laminectomy of C1/C2
Miller et al. [7]	67 (F)	NP, gait disturbance	Occipital craniotomy, laminectomy of C1/C2
Goffin et al.[5]	65 (M)	Arm/leg sensory disturbance, tetraparesis	Occipital craniotomy, laminectomy of C1/C2
Choe et al. [2]	61 (F)	NP, arm/leg numbness/weakness	Transoral approach, O–C fixation
Weymann et al. [12]	72 (F)	NP, arm numbness, gait disturbance	Laminectomy of C1
Birch et al. [13]	85 (M)	Arm numbness	Transoral approach, C1–2 fixation
	84 (F)	NP	Laminectomy of C1/C2
	60 (F)	Arm weakness	Laminectomy of C1/C2
	68 (F)	Quadriparesis	Transoral approach, C1–2 fixation
Eustacchio et al. [3]	75 (M)	Tetraparesis and ataxia	Occipital craniotomy, hemilaminectomy of C1
Fransen et al. [4]	75 (F)	NP	Occipital craniotomy, hemilaminectomy of C1/C2
Akiyama et al. [1]	51 (F)	Headache, leg numbness/weakness	Transcondylar approach, O–C fixation
Tobenas-Dujardin et al. [10]			Transoral approach, C1–2 fixation
Okamoto et al. [8]	72 (M)	NP, arm/leg weakness	Hemilaminectomy of C1/C2, C1–2 fixation
Kirk and Pik [6]	74 (M)	NP, arm sensory disturbance	Transoral approach, C1–2 fixation
Van Gompel et al. [11]	80 (M)	Arm weakness, myelopathy, dysphonia	Transoral approach, O–C fixation
	79 (F)	NP, arm/leg numbness, myelopathy	Transoral approach, O–C fixation
	75 (F)	Finger numbness	Transoral approach, O–C fixation
	81 (M)	NP, dysarthria, myelopathy, dysphagia	Transoral approach, O–C fixation
	74 (M)	NP, arm numbness/weakness	Transoral approach, O–C fixation
	80 (M)	NP, arm numbness/weakness, dystonia	Transoral approach, O–C fixation
	77 (F)	Arm/leg hemiparesis, myelopathy	Transoral approach, O–C fixation
	54 (F)	Arm/leg weakness/numbness	Transoral approach, O–C fixation
	76 (F)	Finger numbness, hand weakness	Transoral approach, O–C fixation
	78 (M)	NP	Transoral approach
Present case	70 (M)	Arm numbness/weakness, gait disturbance	Anterolateral approach

NP neck pain, *O–C* occipitocervical

reportedly accompanied by few complications [6, 10, 11, 13]. Patients who have undergone transoral decompression must rely on nasogastric tube feedings to allow pharyngeal wound healing. This postoperative procedure might confer an infection risk upon the patients.

Percutaneous aspiration surgery is a less invasive surgical alternative [18]. However, we believe that this is an option for patients who are otherwise not good surgical candidates because of the likelihood of recurrence associated with this type of aspiration surgery. Conservative therapy is another option for patients who bear a high surgical risk, those with mild neurological syndromes, and those who refuse surgery [13, 22, 24]. However, the conservative strategy requires careful clinical and radiological monitoring.

Synovial cysts that arise from the lateral aspect of the transverse ligament are treated via a posterior and posterolateral approach. The posterolateral approach can also be used to treat C1–2 synovial cysts at the ventral midsection with slight cord retraction [19]. The transoral and anterolateral approaches are appropriate for C1–2 synovial cysts at the ventral midsection. The most appropriate anterolateral approach was proposed by George and colleagues [21–23]. The anterolateral approach facilitates exposure and control of the V3 segment of the vertebral artery and access to lesions located anteriorly and laterally to the craniocervical junction. This approach can treat small lesions in the ventral mid-portion of the craniocervical junction without spinal cord compression. Surgeons should exercise caution for lesions that are harder and less malleable. With this approach, the operation field is deep and narrow, and therefore it is difficult to achieve a watertight closure of the dura mater. The indications for this approach are tumors located in the extradural spaces or bony structures of the anterior and anterolateral parts of the craniocervical junction, as well as control of the vertebral artery V3 segment.

In this case, we treated a C1–2 focal synovial cyst that presented at the ventral midsection via an anterolateral approach. There was no postoperative deterioration of atlantoaxial instability. We intradurally confirmed and resected the mass lesion. However, we did not dissect and treat the tectorial membrane and cruciform ligament in addition to the lesion. The synovial cyst between the dens and the transverse ligament of atlas had progressed medially and compressed the tectorial membrane and dura mater. The inflammation and mechanical stress of the synovial cyst might have brought these elements together to generate a cyst wall that blocked the connection to the extradural space. There was no postoperative cerebrospinal fluid leak.

Conclusion

Retro-odontoid synovial cysts are rare and attributable to degenerative changes in the atlantoaxial joints. The anterolateral approach facilitates access to lesions located anteriorly to the craniocervical junction without harming the atlantoaxial joints. In this case, we performed surgery to resect a retro-odontoid synovial cyst via an anterolateral approach without fusion. During a 3-year follow-up period, we observed neurological improvement with no deterioration in the C1–2 instability. The postoperative atlantoaxial instability depends more on the patient's age and preoperative mobility than on the operative procedure. Therefore, the indication of an anterolateral approach without fusion for a retro-odontoid synovial cyst requires deliberate preoperative consideration.

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Conflict of interest The authors report no conflicts of interest concerning the materials or methods used in this study or the findings specified in this paper.

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