

Transoral Surgical Approach for Treatment of Symptomatic Atlantoaxial Cervical Synovial Cysts

Semtomatik Servikal Atlantoaksiyal Sinoviyal Kistlerin Transoral Cerrahi Yaklaşım ile Tedavisi

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ABSTRACT

AIM: Synovial cysts are relatively common in the lumbar spine and very uncommon in the cervical spine. Several case reports and a few small series have been reported in the literature over the past four decades. There are two distinct cervical spine synovial cyst categories: atlantoaxial and the subaxial cervical spine. The surgical treatments are very different.

MATERIAL and METHODS: We report eleven patients undergoing transoral resection and posterior fusion for histologically confirmed symptomatic atlantoaxial synovial cysts. This represents a retrospective review over 18 years. The authors analyzed the literature of patients who underwent surgical treatment for symptomatic atlantoaxial synovial cysts.

RESULTS: There were four male and seven female patients with a mean age of 76 years (range 54-84 years). All patients presented with cervical myelopathy. Patients were neurologically assessed pre- and postoperatively and outcome reported using the Modified Rankin Outcome score. Mean follow-up period was 22 months (range 6-120 months). Ten of the eleven patients had improvement in their postoperative assessment and one patient remained unchanged.

CONCLUSION: Myelopathy is the presenting symptom in the vast majority of these patients. Surgical resection and decompression of the neural structures can be an effective treatment for symptomatic atlantoaxial synovial cysts.

KEYWORDS: Atlantoaxial, Cervical spine, Synovial cyst, Transoral, Myelopathy

ÖZ

AMAÇ: Lomber bölgede sinoviyal kistler sık görülse de servikal bölgede oldukça nadir görülür. Son kırk yıl içerisinde rapor edilmiş olan olgu sunumu ya da seri sunumu az sayıda olmuştur. Servikal bölgede görülen sinoviyal kistler iki ayrı kategoride incelenebilir, atlantoaksiyel ve subaksiyel olanlar.

YÖNTEM ve GEREÇLER: Ağız yolundan girilerek çıkarılmış olan 11 servikal sinoviyal kist olgusu sunulmaktadır. Bu olguların tamamı histopatolojik olarak sinoviyal kist olarak tanı almış semptomatik olgulardır. Bu inceleme 18 yıllık geriye dönük bir taramadır. Yazarlar semptomatik atlantoaksiyal sinoviyal kist nedeni ile ameliyat edilen olguları da literatürden araştırdılar.

BULGULAR: Toplam 11 hastanın 4'ü erkek, 7'si kadın ve ortalama yaş 76 olarak hesaplandı. (Yaş aralığı 54-84). Tüm hastaların temel bulgusu servikal miyelopatiydi. Tüm hastalar ameliyat öncesi ve sonrası değerlendirildi. Tüm bulgular Modifiye Rankin Sonuç ölçütlemesi ile hesaplandı. Hastalar ortalama 22 ay takip edildi (takip aralığı 6-120 ay). Hastalardan 10'u ameliyat öncesi döneme göre daha iyi, bir hastada ise değişiklik saptanmadı.

SONUÇ: Miyelopati, servikal sinoviyal kisti olan hastaların çoğunun başvuru sebebidir. Semptomatik atlantoaksiyal sinoviyal kistlerin cerrahi çıkarımı ve sinir dokularının basınç altından kurtarılması etkin bir tedavi yöntemidir.

ANAHTAR SÖZCÜKLER: Atlantoaksiyal, Servikal omurga, Sinovial kist, Transoral, Miyelopati

INTRODUCTION

Atlantoaxial synovial cysts are found adjacent to the transverse ligament and can produce symptoms of progressive myelopathy. These lesions are also reported in the literature as juxtafacet cysts (3,18,25,36,41,52,55), ganglion cysts (2,7,9,10,32,35,38,39,48,50), articular cysts (11), intraosseous cysts (34), epidural cysts (28) and ligamentum flavum cysts (1). Some investigators distinguish between synovial cysts, which are lined with pseudostratified columnar cells containing

clear fluid, versus ganglion cysts which have a connective tissue capsule without a mesothelial lining (30,33,36). The clinical presentation and surgical treatment of these lesions regardless of precise histopathological classification is essentially the same, the terminology seems less important. The most common term in the literature for these lesions is synovial cyst.

Synovial cysts have been reported at all levels of the spine. Most commonly, they appear in the lumbar spine and several

reports have described the clinical presentation, imaging characteristics and surgical treatment (1,2,6-10,12,18,24-26,28,31,32,35,36,39,43,50,52,54). Much less commonly found are synovial cysts of the cervical (11-16,20-23,30,33-38,41,42,45-49,51,52,55-59) and thoracic spinal segments (8,19,29,44,52). Subaxial cervical synovial cysts can present as myelopathy or radiculopathy (14-18,20,21,28,34,36,38,40-42,46,58) whereas C1/C2 synovial cysts present almost exclusively as myelopathy (11,13,23,30,37,45,48,49,51,57, 59). The surgical treatment of atlantoaxial synovial cysts versus subaxial synovial cysts varies considerably. The purpose of this review was to analyze the clinical features, surgical treatments and outcomes of atlantoaxial cervical synovial cysts in our series and those reported in the literature.

MATERIAL and METHODS

Between 1993 and 2010, 11 patients with atlantoaxial synovial cysts underwent surgical resection. Those patients with C1/C2 synovial cysts, for whom a minimum of six month follow-up evaluation was not possible or there was a lack of histopathological confirmation of a synovial cyst, were excluded from this analysis. Eight patients who underwent transoral resection of their presumed synovial cyst were excluded from this analysis. Seven of the patients did not have pathologic confirmation of a synovial cyst performed at the time of resection and one patient had less than six months of follow-up. We evaluated the clinical presenting symptoms, neuroimaging studies, location of the synovial cyst, surgical treatment and outcomes of the patient based upon a retrospective review of patient charts. This study was approved by the Institutional Review Board. The mean age of the patients was 76 years (range 54-84 years). There were four males and seven females. All patients presented with progressive myelopathy symptoms. No patients reported a history of cervical spine trauma or had previous surgery at the level of the synovial cyst. All patients underwent magnetic resonance (MR) imaging or, when unable to undergo MR imaging, computerized tomography (CT) myelography as part of their preoperative evaluation.

RESULTS

All patients underwent a transoral decompression, gross total synovial cyst resection and a posterior fusion, either concomitant or subsequent to the transoral resection. Histopathology confirming the presence of the synovial cyst

was performed in all 11 cases. Nine of the eleven patients underwent a planned tracheostomy prior to the transoral approach. No patients required a permanent tracheostomy. Three patients had an intraoperative cerebrospinal fluid leak which was repaired directly and treated with lumbar drainage. Based upon clinical evaluation, five patients underwent a postoperative swallowing study. Of these, two patients had documented swallowing dysfunction requiring temporary placement of a gastrostomy for postoperative dysphagia. Two patients required temporary placement of a gastrostomy for postoperative dysphagia. One patient had an infection with *Streptococcus viridians* treated with a course of intravenous antibiotics. All patients underwent neurological follow-up and the postoperative Rankin score was determined based upon that evaluation (Table I). The mean follow-up period was 22 months (range 6-120 months). It is important to note however, that one patient had an extensive follow-up period. When that patient is removed from the follow-up analysis the mean follow-up time was 12 months. All patients either stabilized (1 patient) or improved (10 patients) from their preoperative Rankin Outcome Score at follow-up. The patient characteristics are summarized in Table II.

DISCUSSION

Synovial cysts are recognized as a cause of radicular and myelopathic symptoms. The review of the literature on cervical synovial cysts identifies both atlantoaxial and subaxial locations. Magnetic resonance imaging has greatly increased the preoperative diagnostic accuracy of the lesions (5,8,24,29) (Figures 1-3). The etiology of synovial cysts is generally thought to be related to motion or trauma affecting the facet joint (26,43,50). This would correlate with the observation that the most common lumbar level for a synovial cyst is L4/L5 (28,43) and for the subaxial cervical level they are most often reported at C7/T1 (16,18,20,21,28,40-42,46,55). The vast majority of synovial cysts reported in the literature involve the lumbar spine (1,2,5-12,18,24-26,31,32,35-39,43,50-52,54). There are reports of synovial cysts affecting the thoracic spine, but these are much less common (8,19,29,44). Atlantoaxial synovial cysts most commonly arise in the retroodontoid space from the atlantodental joint and present as cervicomedullary compressive myelopathy (3,4,11,13,15,17,22,23,27,30,37,45,47-49,51,56-59). Reports of acute hemorrhage (31,43,54) or rapidly enlarging cysts (12,46) have been reported requiring urgent surgical

Table I: Modified Rankin Scale

0	No symptoms
1	No significant disability. Performs all usual activities, despite some symptoms
2	Slight disability. Able to look after own affairs without assistance, but unable to carry out all previous activities
3	Moderate disability. Requires some help, but able to walk unassisted
4	Moderately severe disability. Unable to attend to own bodily needs without assistance, and unable to walk unassisted
5	Severe disability. Requires constant nursing care and attention, bedridden, incontinent
6	Dead

Table II: Summary of Clinical Features of Patients Undergoing Surgery for Atlantoaxial Synovial Cysts

Age (yrs) Sex	Surgery	Cyst Resection	Rankin Score Preop Postop	F/U months	CSF Leak	Infection
54/F	TO/PF	Total	3 1	20	No	No
81/M	TO/PF	Total	2 1	9	Yes	No
76/F	TO/PF	Total	3 1	6	No	No
74/M	TO/PF	Total	2 1	8	No	No
80/M	TO/PF	Total	3 2	15	No	No
77/F	TO/PF	Total	3 2	10	Yes	Yes
80/M	TO/PF	Total	3 2	8	No	No
84/F	TO/PF	Total	3 3	15	No	No
79/F	TO/PF	Total	3 1	18	No	No
77/F	TO/PF	Total	2 1	10	Yes	No
75/F	TO/PF	Total	3 2	120	No	No

Abbreviations: TO/PF = transoral/posterior fusion, CSF = cerebrospinal fluid

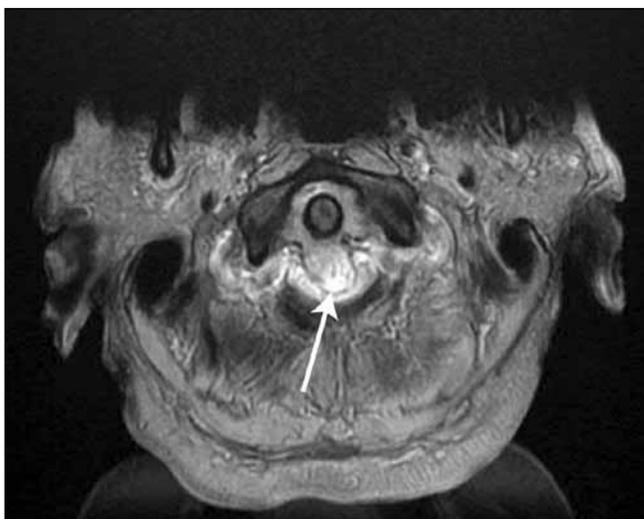


Figure 1: Axial T2-weighted MR demonstrating large synovial cyst (arrow) arising from the transverse ligament resulting in compression of the upper cervical spinal cord.



Figure 2: Sagittal T2-weighted MR demonstrating synovial cyst (arrow) arising from the transverse ligament resulting in severe compression of the medulla and upper cervical spinal cord.

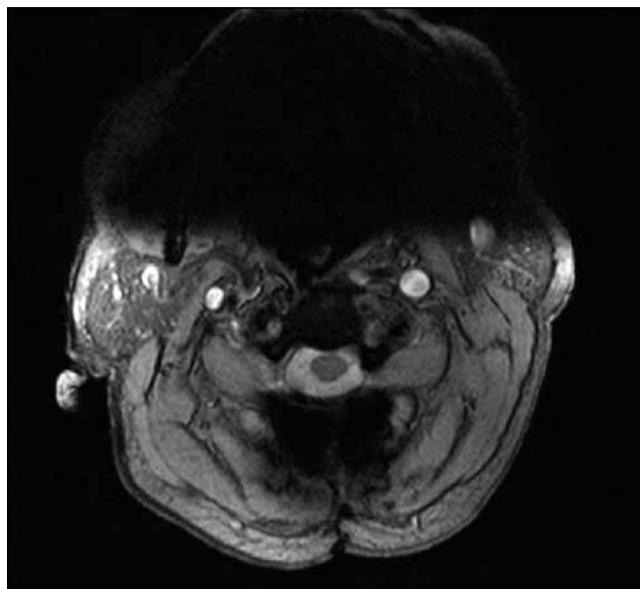


Figure 3: Axial T2-weighted MR shows postoperative changes showing resection of the synovial cyst and decompression of the upper cervical spinal cord.

intervention. The pathophysiologic etiology of synovial cysts and spinal segment frequency, remains a focus of debate in the literature. Generally, they are thought to arise from degenerative, congenital, inflammatory or traumatic causes (42). The upregulation of angiopoietin-1, basic fibroblastic growth factor, substance P, platelet-derived growth factor and interleukins at the site of mechanically stressed facet joints may result in synovial hyperplasia leading to the formation of the cyst (20).

The surgical approach can be via a transoral approach with or without a posterior fusion or a posterior laminectomy approach. Several authors have reported good success with a posterior C1/C2 decompressive laminectomy and resection of the synovial cyst (4,11,13,22,23,27,30,45,47,49,57,59). Zorzon

et al. reported on their two cases utilizing the posterolateral approach and subtotal resection of the cyst (59). Both patients did well at follow-up. Synovial cysts that are more lateral at the atlantoaxial junction are very successfully approached via a posterior approach (4,11,22,30,45,49,59). The posterior transdural approach to the cyst has been reported by some as a safe and effective procedure (3,27,59). Subtotal resection of the cyst wall was performed in some of these cases, albeit with good postoperative neurologic results (11,23,30,49). Vergne and colleagues reported on their patient who underwent a posterior laminectomy, but biopsy alone was performed because of concern for neurologic deficits (57). Birch et al. reported their series of four patients, two undergoing a transoral approach and two undergoing a posterior approach (11). The location of the cyst impacted the surgical approach chosen. The two patients with a more lateral cyst underwent the posterior approach and subtotal resection, whereas the two patients with midline cysts underwent a transoral approach and gross total resection. All four patients demonstrated improvement on postoperative neurologic evaluation. The presence of some residual cyst did not impact outcome (11). The risk of postoperative dysphagia with the transoral approach is not insignificant, but has also been reported with the posterior approach secondary to hypoglossal nerve injury (22).

Others have utilized the transoral approach (11,17,37). These authors reported gross total resections of the synovial cysts

with direct visualization via the transoral approach. There was one postoperative wound infection that was successfully treated (11). There are reports of fusion alone without resection of the synovial cyst resulting in good outcomes (4,13,47). These authors advocate consideration of fusion alone as an option when the resection of the cyst is considered to carry a significant neurologic risk. In addition, two reports of cyst regression with external immobilization have been reported (15,53). Percutaneous aspiration has been reported as a viable alternative to surgical intervention (56). A summary of the reports of surgical treatment of atlantoaxial synovial cysts is found in Table III.

There are several different surgical approaches reported in the literature for the treatment of atlantoaxial synovial cysts. The posterior approach of cervical laminectomy with or without partial suboccipital craniectomy can be advantageous especially with laterally located cysts. Gross total resection, subtotal resection, biopsy and no resection via the posterior approach have all been reported with good clinical outcomes. The transoral approach may be associated with increased risks of cerebrospinal fluid leak and dysphagia, but potentially has the advantage of better visualization of the cyst and less cervical cord retraction. The number of reported cases with the variety of surgical techniques employed does not yet lend itself for statistically significant meta-analysis and determination of the optimal approach. For those cysts that are more laterally located, the case reports suggest that the

Table III: Literature Summary of Reported Cases of Atlantoaxial Cervical Synovial Cysts

Authors & Year	Age(yrs) / Sex	Surgery	Cyst Resection	Pathology	Outcome
Miller et al. 1989	67/F	DL C1/C2	Total	Yes	Fair
Goffin et al. 1992	65/M	DL C1/C2	Total	Yes	Good
Choe et al. 1992	61/F	TO/PF	Total	Yes	Good
Kaufmann et al. 1996	52/M	TO	Total	Yes	Good
Vergne et al. 1996	64/F	DL C1	Total	Yes	Good
Birch et al. 1996	85/M	TO/C1/C2 PF	Total	Yes	Good
	84/F	DL C1/C2	Total	Yes	Good
	60/F	DL C1/C2	Subtotal	Yes	Good
	68/F	TO/C1/C2 PF	Total	Yes	Good
Fransen et al. 1997	75/F	DL C1/C2	Total	Yes	Good
Akiyama et al. 1999	51/F	DL C1/PF	Total	Yes	Good
Aksoy et al. 2000	61/M	DL C1/C2/AF	None	No	NR
Chang et al. 2000	45/M	C1/C2 PF	None	No	Good
Zorzon et al. 2001	84/F	DL C1/C2	Total	Yes	Good
	74/F	DL C1/C2	Total	Yes	Good
Eustacchio et al. 2003	75/M	DL C1	Subtotal	Yes	Good
Morio et al. 2003	71/F	C1/C2 PF	None	No	Good
Okamoto et al. 2004	72/M	DL C1/C2/PF	Subtotal	NR	Good
Elhammady et al. 2009	67/F	DL C1	Total	Yes	Fair

Abbreviations: DL = decompressive laminectomy; TO = transoral resection; PF = posterior fusion; AF = anterior fusion, NR = not reported

posterior approach offers good clinical outcomes and avoids the specific morbidity associated with the transoral approach. With the posterior approach, the reports in the literature suggest that gross total resection of the cyst or cyst wall is not always possible with more midline cysts without potentially increasing the risk of neurologic morbidity. Preoperative neurological functional status and location of the synovial cyst may be important factors in determining the best surgical approach in these patients. Our patients all underwent a transoral approach, gross total resection of the synovial cyst and posterior fusion. Our results compare favorably to those reported in the literature. The patient population is generally elderly as reflected in our series as well as those reported in the literature. The transoral approach does, in many patients, predispose them to a destabilizing procedure requiring a fusion. Careful surgical planning and proper counseling with patients can result in neurological stabilization and in most patients, improvement in neurological function.

CONCLUSIONS

Symptomatic atlantoaxial synovial cysts are rare. Reports in the literature describe a variety of surgical approaches that are effective treatment options. The transoral approach for atlantoaxial synovial cysts does have morbidity associated with it. No patient had any neurological deterioration with this approach. However, cerebrospinal fluid leak, dysphagia and infection are important risk factors to consider when planning the optimal surgical approach in these patients. Synovial cysts that are more lateral at the atlantoaxial junction may be better approached posteriorly. The results of our patients undergoing the transoral approach for resection of the synovial cyst and decompression of the upper cervical cord compare favorably with other reports, including posterior approaches. There is not enough surgical evidence to recommend one approach over another as the optimal treatment option. It does appear clear, however, that neural decompression can result in good functional outcomes in this cohort of patients. Magnetic resonance imaging is currently the optimal radiographic study to identify these lesions. Surgical intervention in these patients with progressive myelopathy is an effective treatment.

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