Reminder of important clinical lesson

Cervical degenerative intraspinal cyst: a case report and literature review involving 132 cases

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Summary

Intraspinal and extradural cysts in the cervical spine are rare disorders that may cause myelopathy or radiculopathy. A synovial cyst or ganglion derived from the facet joint and that from a ligamentum flavum have been reported. We report a surgical case of degenerative intraspinal cyst, causing cervical myelopathy. MRI of a case revealed cystic lesion at C4–5. Spinal cord was compressed by cyst and symptoms of myelopathy were also observed. The patient with cervical spinal canal stenosis underwent laminoplasty and excision of the cyst. The patient recovered well immediately after the surgery. Literature review showed that 133 patients have been reported, including the present case. Previous reports indicated that most cysts occurred in old patients and at the atlanto–axial or C7–T1 junction, and laminectomy or laminoplasty with excision of the cyst gave good results in most cases.

BACKGROUND

Degenerative intraspinal or juxtafacet cysts cause various neurological symptoms. They are often seen at lumbar facet joints but rare at cervical facet joints. We report a surgical case of degenerative intraspinal cyst, causing cervical myelopathy. We also review 66 published articles reporting 132 degenerative intraspinal cysts in the cervical spine, and discuss the characteristic clinical features, surgical results and the choice of surgical treatment and diagnostic tools.

CASE PRESENTATION

A 56-year-old man experienced sudden numbness in the upper extremities and gait disturbance for 3 weeks. He complained of severe pain radiating through his lower back and buttocks when his neck was extended. Neurological examination showed hyper-reflexes of the deep tendons in the extremities. Plain radiographs of the cervical spine showed narrowing of the disc space at C4–C5 and C5–C6. MRI of the cervical spine showed canal stenosis at C4–C5 and C5–C6, a cystic lesion at the left C4–C5 epidural space and marked degenerative change of the left C4–C5 facet joint (figures 1A,B). The axial CT myelogram showed a mass compressing the spinal cord ventromedially at the left C4–C5 level and degeneration of the left C4–5 facet.

TREATMENT

The patient underwent laminoplasty between C3 and C7. At the C4–C5 facet joint, there was a cyst in the ventral side of the ligamentum flavum (figure 1C). It was adhering to the dural surface but was easily detached from the dura. When the capsule of the lesion was incised, a thick yellow mucosal fluid flowed out. The cyst was excised with the pericystic ligamentum flavum. Histopathological examination showed the cyst to be composed of non-specific degenerative tissue and no synovial lining cells.

OUTCOME AND FOLLOW-UP

The patient showed good recovery in ambulation and severe pain radiating through his lower back and buttocks. Follow-up MRI revealed a decompressed spinal cord and no recurrence of the cyst. He has no symptom two years after surgery.

DISCUSSION

Intraspinal extradural cysts have occasionally been seen in lumbar spine since MRI was introduced in clinical examination. But those are still rare in the cervical spine. They have been reported as arachnoid, synovial and ganglion cysts. Kao *et al*¹ grouped synovial and ganglion cysts together because they considered that both types of cyst arise at the joint capsule of the facet synovial joint and proposed the term ‘juxtafacet cysts’ to represent both of them. Hatem *et al*² used the term ‘intraspinal cervical degenerative cyst’ in the report of three cases of cervical cysts including a case of ligamentum flavum cyst because cysts of the ligamentum flavum, defined as those arising inside this ligament, are differentiated from the juxtafacet cyst. Shima *et al*³ proposed the term ‘degenerative intraspinal cyst’ as a more accurate description of these cysts in the spinal canal that arise from degenerated spinal structures including not only facet joints, but also ligaments and disc. In the review of the past reports of intraspinal extradural cysts in the cervical spine, five cysts are considered to arise from the ligamentum flavum.^{2 4–6} We use the term ‘degenerative intraspinal cyst’ as we agree with Shima *et al* for the point that some cysts in the spinal canal arise from degenerated spinal structures including not only facet joints, but also ligaments.

To the best of our knowledge, the total number of degenerative intraspinal cysts that have ever been reported is 67 articles, in 133 patients, including the present case (table 1). Although the cysts occur over a wide age range (8–86 years), they are more commonly encountered in

Figure 1 (A) Sagittal T2-weighted MRI shows canal stenosis at C4–C5, C5–C6 and a high-intensity extradural mass at C4–C5. (B) Axial MRI shows a high-intensity extradural mass in the spinal canal adjacent to the left C4–C5 facet and marked degenerative change of the C4–C5 facet. (C) Intraoperative image shows extradural cyst in the spinal canal adjacent to the left C4–C5 facet and quite adherent to the dura.

older persons (mean age 67.3 years). The reported case included 65 men, 64 women and unknown 4 cases among the total 133 cases. The level of the cyst was the atlanto-axial junction in 40 cysts (30.1%), C3–C4 in 13 (9.8%), C4–C5 in 10 (7.5%), C5–C6 in 10 (7.5%), C6–C7 in 9 (6.8%) and C7–T1 in 51 (38.3%). Bilateral cysts have been reported in two cases, and both of them were located at C7–T1.^{7 8} A higher incidence of cysts was seen at levels C1–C2 and C7–T1 levels. The degenerative diseases like as disc herniations and canal stenosis usually occur at middle cervical spine and lower lumbar spine. The degenerative intraspinal cysts are often seen at the lower lumbar spine as well as degenerative diseases. But they are not often seen at middle cervical spine. There might be different

occurrence mechanism of degenerative intraspinal cysts between cervical and lumbar spine. Krauss *et al*⁸ postulate that the C7–T1 level was a common location for juxtafacet cysts because this level is immediately superior to the relatively immobile thoracic spine. In his comment Cooper disagreed with Krauss *et al* because the C7–T1 level is less mobile than the three superior motion segments and is infrequently the site of disc herniations and spondylosis, which occur most frequently at the sites of most movement. Hatem *et al*² mentioned that the presence of the paradoxical motion without discogenic spondylosis at the C7–T1 level and the loss of the mobility in the higher two levels observed in older individuals can explain preferential localisation of these cysts at this level.

Table 1 Reported cases of degenerative intraspinal cysts in the cervical spine

No	Authors	Year	Age (years)	Gender	Location of cyst	Treatment
1	Kao <i>et al</i>	1974	52	M	C6–C7 facet joint	C6–C7 laminectomy
2	Cartwright <i>et al</i>	1985	41	M	C7–T1 facet joint	C6–T1 laminectomy
3	Jarbe <i>et al</i>	1987	60	M	C6–7 facet joint	C6–T1 laminectomy
4	Onofrio and Mih	1988	73	M	Odontoid process	C1–C2 laminectomy
5	Patel & Sanders	1988	42	F	C4–C5 facet joint	C4 hemilaminectomy
6	Miller <i>et al</i>	1989	67	F	odontoid process	C1–C2 laminectomy
7	Nijensohn <i>et al</i>	1990	58	M	C4–C5,C5–C6 facet joint	C4–C6 posterior spinal fusion
8	Takano <i>et al</i>	1992	72	M	C3–C4 ligamentum flavum	C3–C6 laminectomy
9	Quaghebeur & Jeffree	1992	82	M	C1–C2 facet joint	C1–C2 laminectomy
10	Goffin <i>et al</i>	1992	65	M	C2quadrate ligament	C1–C2 laminectomy
11	Choe <i>et al</i>	1993	61	F	odontoid process	C1 laminectomy
12	Weymann <i>et al</i>	1993	72	F	C1–C2 facet	C1 laminectomy
13	Epstein & Hollingsworth	1993	47	F	C7–T1 facet joint	C7–T1 hemilaminectomy
14	Freiberg <i>et al</i>	1994	(–)*	(–)*	C7–T1 facet joint	C7–T1 laminectomy
15	Vergne <i>et al</i>	1996	64	F	Odontoid process	C1 laminectomy
16	Birch <i>et al</i>	1996	68	F	behind dens	C1–C2 transoral fusion
17	Birch <i>et al</i>	1996	78	F	behind dens	observe
18	Birch <i>et al</i>	1996	60	F	behind dens	C1–C2 laminectomy
19	Birch <i>et al</i>	1996	85	M	behind dens	transoral fusionC1–C2
20	Birch <i>et al</i>	1996	84	F	behind dens	C1–C2 laminectomy
21	Chan LF, <i>et al</i>	1996	45	F	C7–T1 ligamentum flavum	C7–T1 laminectomy
22	Kaufmann <i>et al</i>	1996	52	M	Periodontoid	Removal of anterior atrantal arch, odontoid process
23	Sabo <i>et al</i>	1996	(–)*	(–)*	C4–C5 facet joint	(–)*
24	Sabo <i>et al</i>	1996	(–)*	(–)*	C7–T1 facet joint	(–)*
25	Sabo <i>et al</i>	1996	(–)*	(–)*	C7–T1 facet joint	C7–T1 laminectomy
26	Fransen <i>et al</i>	1997	75	F	odontoid process	C1–C2 hemilaminectomy
27	Kotilainen & Marttila	1997	64	M	bilateral C7–T1 facet joint	C7 laminectomy
28	Kayser <i>et al</i>	1998	74	M	C4–C5 facet joint	(–)*
29	Krauss <i>et al</i>	1998	61	M	C7–T1 facet joint	No29–No41All patients underwent partial
30	Krauss <i>et al</i>	1998	69	M	C7–T1 facet joint	hemilaminectomy
31	Krauss <i>et al</i>	1998	79	F	C7–T1 facet joint	Two patients underwent posterior cervical fusions
32	Krauss <i>et al</i>	1998	68	M	C7–T1 facet joint	
33	Krauss <i>et al</i>	1998	53	F	C6–C7 facet joint	
34	Krauss <i>et al</i>	1998	62	M	C3–C4 facet joint	
35	Krauss <i>et al</i>	1998	70	M	C4–C5 facet joint	
36	Krauss <i>et al</i>	1998	58	F	C7–T1 facet joint	
37	Krauss <i>et al</i>	1998	59	M	C7–T1 facet joint	
38	Krauss <i>et al</i>	1998	76	M	C7–T1 facet joint	
39	Krauss <i>et al</i>	1998	74	M	C7–T1 facet joint	
40	Krauss <i>et al</i>	1998	65	M	bilateral C7–T1 facet joint	
41	Krauss <i>et al</i>	1998	64	F	C7–T1 facet joint	
42	Cudlip <i>et al</i>	1999	61	M	C7–T1 facet joint	C7–T1 laminectomy
43	Cudlip <i>et al</i>	1999	61	M	C7–T1 facet joint	C7–T1 laminectomy
44	Cudlip <i>et al</i>	1999	75	M	C3–C4 facet joint	C3–C5 laminectomy
45	Lunardi <i>et al</i>	1999	58	M	C7–T1 facet joint	C7–T1 laminectomy
46	Akiyama <i>et al</i>	1999	51	F	Craniocervical	C1 laminectomy and foramen magnum decompression
47	Chang <i>et al</i>	2000	45	M	C1–C2 transverse ligament	C1–C2 posterior fusion alone
48	Aksoy <i>et al</i>	2000	61	M	os odontoideum C2 body	C1–C2 posterior fusion & laminectomy
49	Stoodley <i>et al</i>	2000	65	M	C7–T1 facet joint	C6–T1 laminectomy
50	Chaoui <i>et al</i>	2000	74	M	C5–C6 facet joint	C5–C6 hemilaminectomy
51	Ito T <i>et al</i>	2000	8	M	C1–C2 facet joint	C1–C2 posterior fusion alone
52	Zorzon <i>et al</i>	2001	74	F	C1–C2 facet joint	C1–C2 hemilaminectomy
53	Zorzon <i>et al</i>	2001	84	F	C1–C2 facet joint	C1–C2 hemilaminectomy
54	Yamamoto A <i>et al</i>	2001	81	F	C3–C4 ligamentum flavum	C3–C7 laminectomy
55	Yamamoto A <i>et al</i>	2001	65	M	C3–C4 ligamentum flavum	C3–C7 laminectomy
56	Hatem <i>et al</i>	2001	65	F	C3–C4 facet joint	C3–C4 hemilaminectomy
57	Hatem <i>et al</i>	2001	77	M	C7–T1 facet joint	C7–T1 hemilaminectomy
58	Hatem <i>et al</i>	2001	73	M	C4–C5 ligamentum flavum	C4–C5 laminectomy
59	Shima <i>et al</i>	2002	66	M	C7–T1 facet joint	C3–C6 laminolpasty
60	Shima <i>et al</i>	2002	68	M	C7–T1 facet joint	C7 laminectomy
61	Shima <i>et al</i>	2002	72	F	C7–T1 facet joint	C7 laminectomy
62	Eustacchio S <i>et al</i>	2003	75	M	Craniocervical	C1 hemilaminectomy
63	Morio <i>et al</i>	2003	71	F	C1–C2 facet joint	C1–C2 posterior fusion alone
64	Fonoff <i>et al</i>	2004	64	M	C3–C4 facet joint	C3 hemilaminectomy
65	Miwa <i>et al</i>	2004	74	F	C7–T1 facet joint	C7–T1 hemilaminectomy

Continued

Table 1 Continued

No	Authors	Year	Age (years)	Gender	Location of cyst	Treatment
66	Cheng YY <i>et al</i>	2004	59	M	C6–C7 facet joint	removal of the cyst
67	Okamoto <i>et al</i>	2004	72	M	C1–C2 facet joint	C1–C2 hemilaminectomy
68	Cho BY <i>et al</i>	2004	80	M	C7–T1 ligamentum flavum	C7 laminectomy
69	McGuigan <i>et al</i>	2005	86	M	C4–C5 facet joint	C4–C6 cervical decompression
70	Gazzeri R, <i>et al</i>	2005	66	F	C7–T1 ligamentum flavum	C7–T1 hemilaminectomy
71	Colen CB	2006	58	F	C7–T1 facet joint	spontaneous resolution
72	Song JK, <i>et al</i>	2006	74	M	C7–T1 facet joint	C7–T1 hemilaminectomy
73	Cheng WY, <i>et al</i>	2006	58	F	C6–C7 facet joint	C6–C7 laminectomy
74	Kostanian VJ, <i>et al</i>	2007	66	F	C6–C7 facet joint	CT guided aspiration
75	Yahara Y, <i>et al</i>	2009	63	F	C4–C5 ligamentum flavum	C4–C5 laminectomy & C4–C5 posterior fusion
76	Nojiri H, <i>et al</i>	2009	61	M	C7–T1 ligamentum flavum	C5–C7 laminoplasty & T1 laminectomy
77	Nojiri H, <i>et al</i>	2009	77	F	C2–C3 facet joint	C3–C7 laminectomy
78	Costa F, <i>et al</i>	2010	84	M	C7–T1 facet joint	C7–T1 hemilaminectomy
79	Mendes–Araújo L	2010	51	F	C1–C2 facet joint	Refused surgery
80	Weng C, <i>et al</i>	2010	13	F	odontoid process	O–C2 posterior fusion
81	Muzii VF, <i>et al</i>	2010	60	M	C4–C5 ligamentum flavum	C4–C5 laminectomy
82	Aizawa T, <i>et al</i>	2010	78	M	C1–C2 facet joint	C1–C2 hemilaminectomy
83	Moon HJ, <i>et al</i>	2010	74	M	C5–C6 facet joint	C4–C5 anterior discectomy fusion & C5–C6 anterior foraminotomy
84–118	Lyons MK, <i>et al</i>	2011	Mean 69 (33–82)	M:24F:11	facet joint	No84~No118All patients underwent gross total resection of the cyst. Twenty-one patients underwent a hemilaminectomy. Fourteen patient underwent a fulldecompressive laminectomy
	Lyons MK, <i>et al</i>	2011	Mean 79	2 cases	C2–C3 facet joint	
	Lyons MK, <i>et al</i>	2011	Mean 72	5 cases	C3–C4 facet joint	
	Lyons MK, <i>et al</i>	2011	Mean 65	8 cases	C5–C6 facet joint	
	Lyons MK, <i>et al</i>	2011	Mean 71	3 cases	C6–C7 facet joint	
	Lyons MK, <i>et al</i>	2011	Mean 70	17 cases	C7–T1 facet joint	
119	Sivakumar W, <i>et al</i>	2011	67	F	C7–T1 facet joint	C7–T1 hemilaminectomy & C5–T2 posterior fusion
120	Found E, <i>et al</i>	2011	47	F	C7–T1 facet joint	C7–T1 hemilaminectomy & C6–T1 posterior fusion
121	Lyons MK, <i>et al</i>	2011	54	F	C1–C2 facet joint	transoral resection and posterior fusion
122	Lyons MK, <i>et al</i>	2011	81	M	C1–C2 facet joint	transoral resection and posterior fusion
123	Lyons MK, <i>et al</i>	2011	76	F	C1–C2 facet joint	transoral resection and posterior fusion
124	Lyons MK, <i>et al</i>	2011	74	M	C1–C2 facet joint	transoral resection and posterior fusion
125	Lyons MK, <i>et al</i>	2011	80	M	C1–C2 facet joint	transoral resection and posterior fusion
126	Lyons MK, <i>et al</i>	2011	77	F	C1–C2 facet joint	transoral resection and posterior fusion
127	Lyons MK, <i>et al</i>	2011	80	M	C1–C2 facet joint	transoral resection and posterior fusion
128	Lyons MK, <i>et al</i>	2011	84	F	C1–C2 facet joint	transoral resection and posterior fusion
129	Lyons MK, <i>et al</i>	2011	79	F	C1–C2 facet joint	transoral resection and posterior fusion
130	Lyons MK, <i>et al</i>	2011	77	F	C1–C2 facet joint	transoral resection and posterior fusion
131	Lyons MK, <i>et al</i>	2011	75	F	C1–C2 facet joint	transoral resection and posterior fusion
132	Brotis AG, <i>et al</i>	2012	82	F	C3–C4 ligamentum flavum	C3 bilateral laminectomy
133	Authors	2012	56	M	C4–C5 facet joint	C3–C7 laminoplasty

These degenerative intraspinal cysts of the cervical spine were usually diagnosed by MRI, which show an oval-shaped cystic lesion at the anterior of the spinal canal at the atlanto–axial junction and posterolaterally in the mid-cervical and lower cervical regions. The signal intensity of the cyst was low to intermediate on T1-weighted images and high on T2-weighted images. Contrast MRI may reveal an enhancing cystic rim. Synovial cysts may also contain gas. MRI is also useful postoperatively to observe the recurrence of the cysts. CT often reveals degenerated facet joints. Calcification or air in the cyst can also be seen in CT.

In the treatment of degenerative intraspinal cysts, most of patients underwent laminectomy and excision of the cysts. Four cases underwent atlanto–axial arthrodesis without excision of the cysts.^{9–12} One case was reported that spontaneous resolution of a C7–T1 synovial cyst was seen.¹³ Laminectomy or laminoplasty and excision of the

cyst are recommended for cases with cervical spinal canal stenosis. If there is no concomitant canal stenosis, minimal laminectomy with excision of the cyst is enough. There have been no reports of recurrence of cyst in the cervical spine after surgery. Stoodley mentioned that, because recurrence after partial excision is rare, it is preferable to leave cyst remnants attached to the dura than to excise and patch the dura, which carries the risk of cerebrospinal fluid leakage.¹⁴ In our case, no recurrence was observed after partial resection of the cyst. We also think it is not necessary to remove the cyst wall adhering to the dura completely. Intraoperative ultrasonography was reported to be very useful in some cases. The border between the cyst and the dura was well visualised by ultrasonography. Intraspinal cysts in the cervical spine are uncommon. When they are diagnosed adequately and treated with appropriate surgery, the clinical outcome is generally good.^{15–20}

Learning point

- A case of cervical intraspinal cyst with myelopathy was diagnosed by MRI and treated with laminoplasty. The patient recovered well postoperatively. Previous reports indicated that most cysts occurred in old patients and at the atlanto-axial or C7-T1 junction, and laminectomy or laminoplasty with excision of the cyst gave good results in most cases.

Competing interests None.

Patient consent Obtained.

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