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Case study

Combined upper cervical canal stenosis and cervical ossification of the posterior longitudinal ligament resulting in myelopathy: A case series and literature review

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ABSTRACT

Combined upper cervical canal stenosis and cervical ossification of the posterior longitudinal ligament (OPLL) is an under-recognized disorder. The objective of the present study was to investigate the radiological manifestations and surgical outcomes of this disease combination. Between May 2011 and July 2014, we studied the radiological manifestations of 18 cases of combined upper cervical canal stenosis and cervical OPLL. Appropriate decompression procedures were performed and the clinical outcomes were evaluated using a visual analog scale (VAS) and the Japanese Orthopedic Association (JOA) scoring system. Radiological outcomes, including the space available for the spinal cord (SAC) at the cephalad-adjacent level, occupying ratio of OPLL, and cervical sagittal alignment, were measured. We found that the etiologies of upper cervical canal stenosis included craniovertebral junction deformity, atlantoaxial subluxation, and OPLL extending to the C2 level. The radiological features of OPLL varied. Postoperatively, all patients showed evidence of improvement in their VAS and JOA scores. The radiological results were satisfactory in terms of the SAC at the cephalad-adjacent level, occupying ratio of OPLL, and cervical alignment. We found that the radiological manifestations of combined upper cervical canal stenosis and cervical OPLL varied among patients. Satisfactory results can be achieved by applying appropriate decompression techniques.

1. Background

Cervical canal stenosis is a common degenerative disorder associated with narrowing of the cervical spinal canal that frequently affects elderly patients [1,2]. The condition is often associated with herniation or bulging of intervertebral discs, osteophyte production, and ossification of the posterior longitudinal ligament (OPLL). As the canal diameter is relatively wider at inferior levels, spinal stenosis is rarely encountered in the upper cervical segment [3]. To date, only a few cases of upper cervical canal stenosis triggering myelopathy have been reported [4,5]. OPLL is characterized by ectopic bone formation within the posterior longitudinal ligament, and can trigger cervical canal stenosis. In most cases of cervical OPLL, ossification is observed at levels C4 and C5, and is often thickest at level C5 [6]. Simultaneous presentation of upper cervical canal stenosis and cervical OPLL in the same patient is very rare. Therefore, both the radiological manifestations and the surgical outcomes remain unclear.

Here, we report our case series of combined upper cervical canal stenosis and cervical OPLL triggering myelopathy. In addition, we systematically reviewed the literature to define the overall radiological manifestations and surgical outcomes of the combined disease. Our objectives were to (1) clarify the radiological manifestations of upper cervical canal stenosis combined with cervical OPLL and (2) evaluate the surgical outcomes in terms of symptom relief, changes in the space available for the spinal cord (SAC) at the cephalad-adjacent level, occupying ratio of OPLL, and cervical alignment.

2. Methods

Between May 2011 and July 2014, we operated on 18 patients with combined upper cervical canal stenosis and cervical OPLL. The inclusion criteria were demonstration of cervical OPLL via X-ray and computed tomography (CT); an upper cervical canal sagittal diameter <14 mm; and a minimum of 18 months of

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Table 1
Case series of patients of combined upper cervical canal stenosis and cervical OPLL.

Case	Group	Age(year)/Gender	Combined morbidity	Disease duration (month)	FU (month)	Upper cervical stenosis	OPLL			VAS		JOA		OR of OPLL (%)		SAC (mm)		Cervical alignment (°)	
							extent	classification	thickest level	Pre	Final	Pre	Final	Pre	Post	Pre	Post	Pre	Final
1	A	63/M	diabetes mellitus	6	27	basilar invagination	C4/C5, C5/C6	circumscribed	C4/C5	8	3	9	10	63.3	27.02	7.1	15.8	18.1	19.6
2	A	77/M	hypertension	18	19	basilar invagination	C3–C5, C6	mixed	C5	7	2	11	16	50.2	46.1	11.1	19.8	10.9	10.2
3	A	71/F	Down syndrome	30	26	atlas hypoplasia	C3–C7	mixed	C5	6	2	10	10	56.7	25.2	10.3	20.2	14.6	18.6
4	A	72/M	hypertension	120	21	atlas hypoplasia	C2–C7	continuous	C3	6	2	7	15	64.8	34.3	12.9	21.3	6.7	9.2
5	B	56/F	–	48	26	atlantoaxial subluxation	C3–6	mixed	C5	5	2	10	16	31.3	19.2	12.3	20.9	18.7	20.5
6	B	45/M	rheumatoid arthritis	24	18	atlantoaxial subluxation	C3–6	continuous	C5	6	1	10	15	49.3	19.6	13.3	19	19.5	16.8
7	B	49/F	–	36	24	atlantoaxial subluxation	C3–5, C6	mixed	C4	7	2	14	17	43.6	28.9	11.2	20.8	12.8	8.9
8	B	68/F	rheumatoid arthritis	72	25	atlantoaxial subluxation	C3, C4, C7	segmental	C4	6	2	12	16	66.4	20.3	13.6	20.1	20.4	15.6
9	B	45/M	diabetes mellitus	60	31	atlantoaxial subluxation	C3–C5, C6	mixed	C5	5	1	11	14	51.6	27.1	9.9	19.9	16.2	14.5
10	C	41/F	–	18	24	OPLL	C2–C5	continuous	C4	4	1	10	11	48.5	20.6	17.6	18.1	−5.8	−1.5
11	C	36/F	chronic kidney disease	6	18	OPLL	C2–C6	continuous	C5	5	2	11	15	42.3	17.7	16.5	18.3	−8.3	−10.4
12	C	44/M	–	60	30	OPLL	C2–C5	continuous	C5	4	1	12	12	61.2	25.6	16.3	17.9	4.9	6.9
13	C	68/M	–	1	21	OPLL	C2–C4, C6	mixed	C3	5	0	14	15	76.5	27.5	17.3	18.5	10.9	7.9
14	C	88/M	coronary heart disease	84	24	OPLL	C2–C6	continuous	C4	6	2	15	16	47.3	22.9	16.9	19.1	−7.6	−5.5
15	C	72/F	–	18	18	OPLL	C2–C3, C5	mixed	C4	5	1	12	15	56.2	23.6	14.3	20.3	2.2	−5.8
16	C	49/M	–	24	24	OPLL	C2–C6	mixed	C5	4	0	7	17	65.7	19.0	18.1	21.3	−6.1	−5.6
17	C	60/M	–	42	25	OPLL	C2–C4, C6	mixed	C3	7	3	10	16	70.2	21.7	16.4	16.9	15.4	10.7
18	C	59/M	hypertension	36	24	OPLL	C2–C4, C5	mixed	C4	5	1	11	15	42.2	19.8	17.2	19.8	14.6	12.3

FU Follow-up, OPLL ossification of the posterior longitudinal ligament, JOA Japanese Orthopedic Association, OR occupying ratio, SAC space available for the spinal cord, Pre preoperative, Post postoperative, M male, F female.

follow-up. The exclusion criteria were any history of previous trauma or surgery on the cervical region and incomplete radiological results. Eleven males and seven females of mean age 59.1 ± 14.4 years (range, 36–88 years) were included (Table 1). The study was approved by the Ethics Committee of the Second Affiliated Hospital, School of Medicine, Zhejiang University, and all patients provided written informed consent.

2.1. Radiological manifestations

We reviewed the radiological features of upper cervical canal stenosis on CT and X-ray images. By reference to the lateral radiography and sagittal CT scans, OPLL was classified as continuous, segmental, mixed, or circumscribed in line with the suggestions of the Investigation Committee on OPLL of the Japanese Ministry of Health, Labor, and Welfare [7]. The level at which the OPLL with greatest thickness was also determined.

2.2. Surgical methods

Patients were divided into three groups based on the radiological features of upper cervical canal stenosis, and appropriate decompression procedures were performed. **Group A** patients had craniovertebral junction deformities triggering upper cervical canal stenosis (n = 4). Two with basilar invaginations underwent transoral decompression, posterior occipitoaxial fusion, and laminoplasty (Fig. 1). Two cases exhibiting hypoplasia of the posterior arch of the atlas underwent resection of the C1 posterior arch, with laminoplasty (Fig. 2). **Group B** patients exhibited atlantoaxial subluxation triggering upper cervical canal stenosis (n = 4) and underwent atlantoaxial fixation, fusion laminoplasty, as the atlantoaxial joint could not be reduced in the extension position (Fig. 3). **Group C** patients exhibited OPLL extending to the C2 level and triggering upper cervical canal stenosis (n = 9). These patients underwent C2 dome-like laminoplasty combined with expansive lower-level open-door laminoplasty (Fig. 4).

2.3. Clinical and radiological outcomes

Pain intensity was assessed using a visual analog scale (VAS). Neurological recovery was scored as recommended by the Japanese Orthopedic Association (JOA). The recovery rate was also used to assess surgical outcomes, and was calculated as follows: Recovery rate (%) = (Final JOA score − preoperative JOA score)/(17 − preoperative JOA score) × 100%, and was ranked as excellent (75–100%), good (50–74%), fair (25–49%), poor (0–24%), and worse (<0) at the final follow-up.

On CT axial imaging, the preoperative and postoperative occupying ratios of OPLL (%) were measured as the anteroposterior diameters of the OPLL with greatest thickness divided by the anteroposterior diameters of the bony spinal canal at the same level. The SAC at the cephalad-adjacent level was defined as the distance from the ventral edge of the posterior ring of the atlas or foramen magnum to the dorsal aspect of the odontoid process, as described by Guo et al. [8]. The sagittal cervical alignment was defined by the Cobb angle between C2 and C7 in profile of neutral, and lateral plain radiographs.

2.4. Statistical analysis

We used SPSS software version 17.0 (IBM, Armonk, New York) for all statistical analyses. Paired *t*-tests were employed to analyze differences among the preoperative, postoperative, and final parameters. Data are presented as means ± standard deviation and a *P* value <0.05 was considered to reflect statistical significance.