Diagnosis and surgery of ossification of posterior longitudinal ligament associated with dural ossification in the cervical spine

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Abstract Direct removal of the ossified mass via anterior approach carries good decompression to ossification of the posterior longitudinal ligament (OPLL) in the cervical spine. Ossification occasionally involves not only the posterior longitudinal ligament but also the underlying dura mater, which increased the opportunity of the cerebrospinal fluid (CSF) leakage or neurological damage. The surgeon was required to recognize the dural ossification (DO) and need more cautious manipulation. Hida et al. first described the computed tomography (CT) findings that indicated the association with DO, and suggest the double-layer sign appeared more specific for DO. This study reviewed 138 patients who received anterior cervical corpectomy and fusion (ACCF) for OPLL, and 40 patients were found in the association with DO during anterior procedure. Radiological studies revealed that the patients with severe OPLL (higher occupying rate and larger extent) have increasing opportunity of association with DO. The double-layer sign, as a specific indicator for association with DO was sensitive in the patients with mild OPLL, but less frequent in those with severe OPLL with DO. Two surgical techniques were used for the patients with DO in anterior decompression procedure. When the double-layer sign was observed on CT scans, the OPLL could be separated from

DO through a thin layer consisting a nonossified degenerated PLL to avoid CSF leakage. Otherwise, the entire ossified mass including OPLL and DO was removed completely. In this technique, the arachnoid membrane needed to be persevered with the aid of microscope to avoid a large area of membrane defect, resulting in uncontrolled CSF leakage. There was no significant difference in clinical results between the patients with DO and those without DO. Therefore, ACCF is meritorious for the patient with OPLL associated with DO, although more difficult manipulation and higher risk of CSF leakage.

Keywords Cervical spine · Ossification of the posterior longitudinal ligament · Dural ossification · Anterior cervical corpectomy and fusion

Introduction

Ossification of the posterior longitudinal ligament (OPLL) is a frequent cause of cervical myelopathy or radiculopathy in Asian population. Anterior decompression and direct removal of OPLL can achieve more satisfactory results even for severe multilevel OPLL [5, 12]. However, when the dural membrane is involved in ossification, the risks of complications such as cerebrospinal fluid (CSF) leakage and injury to spinal cord or nerve root increased. In this condition, the choice of surgical tactics is important to avoid unnecessary complications, and surgical manipulation for removal of the ossified mass must be very cautious. Therefore, identification of dural ossification (DO) associated with OPLL on imaging studies provide important information in considering the anterior approach preoperatively. Hida et al. [4] had described a single- and double-layer signs on computed tomography (CT) findings, which

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were associated with DO, and also suggested the typical double-layer sign was more pathognomonic for dural penetration and replacement by OPLL. The purpose of this study was to investigate the value of the double-layer sign to the diagnosis of DO and elucidate its significance to the patients with OPLL.

Materials and methods

Patient population

Between May 2002 and October 2006, a total of 138 consecutive patients (97 men and 41 women) received anterior cervical corpectomy and fusion (ACCF) for OPLL at our department. The mean age at operation was 56.3 years, ranging from 43 to 71 years. The patients presented moderate to severe spastic limb paresis, and the duration of symptoms lasted at least 12 months. Most of the patients decided to have a surgical decompression due to recent neurological aggravation and four among them needed anterior revision operation due to insufficient decompression after initial posterior laminoplasty.

Radiological evaluation

All the patients had pre- and postoperative plain radiographs, CT scans, and magnetic resonance images (MRIs). The diagnosis of OPLL was established for all the patients according to the results of radiological examination preoperatively, and more detailed characteristics of CT and MRI findings described as follows were investigated.

Computed tomography

The occupying rate (OR) was defined as the thickness of OPLL divided by the anteroposterior diameter of the bony spinal canal on the axial image. On the sagittal image, the extent of OPLL was investigated and OPLL was classified into four types: local-type, segmental-type, continuous-type and mixed type. The double-layer sign, characterized by anterior and posterior rims of hyperdense ossification separated by a central hypodense mass, was observed on axial images, which indicated the association with DO.

Magnetic resonance imaging

The cord flattening rate was defined as the minor axis length of the spinal cord divided by its major axis length at the level of maximal cord compression on axial T1-weighted MR images. This parameter has been used to

evaluate compression of the spinal cord without individual differences. When it is <0.4 after operation, surgical decompression is considered to be inadequate.

Surgical technique

Under general anesthesia, the patients were placed in the supine position with neck slightly extended. The cervical spine was exposed through a standard right-side anterior approach. The appropriate surgical level was confirmed by intraoperative radiography. After necessary discectomies, the vertebral bodies were partially removed using an appropriate rongeur. The residual vertebral bodies and OPLL were removed by high speed drill until the OPLL was thinned as much as we can. Then, the OPLL was separated from dural matter using a specialized microdissector. The head of this dissector was a hook with a narrow slot. It was inserted under the OPLL from the nonossified ligament, rotated and slightly lifted. The ligament was cut off by scalpel along the slot. After that, the OPLL was meticulously separated using the microdissector and removed by 1–2 mm Kerrison rongeur and microcurettes.

If the patient was found in the association with DO during anterior decompression procedure and there was a typical double-layer sign on CT scan, we tried to separate OPLL from DO through a thin layer consisting a nonossified degenerated PLL between the OPLL and the DO. This technique has been detailed by Mizuno et al. [8], and the ossified portion of the dural mater was carefully preserved to avoid dural defect and CSF leakage (Fig. 1). However, the OPLL was not always able to be resected from the ossified portion of dura mater, because the nonossified layer of the PLL was not found in those patients. In this condition, two surgical tactics could be selected. One was the floating method, in which the OPLL was only separated from the around vertebral wall, but remained together with DO. The entire ossified mass could gradually float anteriorly and did not compress the spinal cord eventually. Nevertheless, considering the decompressive effect by the floating method was limited or got very slowly at least, the entire ossified mass including OPLL and DO was removed completely in our patients if they could not be separated. This technique needed to be operated with the aid of microscope, and the arachnoid membrane was hoped to be preserved to avoid a large area of membrane defect, resulting in uncontrolled CSF leakage (Fig. 2). After decompression, the titanium mesh cage filled with autologous bone fragments or iliac crest strut together with anterior cervical plate was used to restore the stability of the involved segments. The procedure involved one-level corpectomy in 31 cases, two-level in 84 cases and three-level in 23 cases.

Fig. 1 A 53-year-old man developed numbness and weakness in his four extremities for 2 years, together with sphincter dysfunction for 1 week. Preoperative imaging studies showed a C4–C7 mixed-type OPLL. Double-layer sign was observed on preoperative axial CT scan, indicating association with DO. During operation, the OPLL was completely separated from the DO through the layer consisting a nonossified degenerated PLL and no dural tears occurred. **a** Preoperative radiographic image at neutral position; **b**, **c** preoperative

sagittal and axial CT scans demonstrating a C4–C7 mixed-type OPLL with double-layer sign; **d** preoperative MR image showed severe compression of the spinal cord; **e** postoperative radiographic image showing C4–C6 corpectomy and fusion; **f**, **g** postoperative sagittal and axial CT demonstrating complete resection of OPLL and floating of DO; **h** postoperative MR image showed sufficient decompression of the spinal cord

Fig. 2 **a** 47-year-old woman developed numbness in her four extremities for 7 years, aggravated to walking disturbance for 2 years. Preoperative imaging studies showed a C2–C3 local-type OPLL and C4–C7 continuous-type OPLL. No double-layer sign was observed on preoperative axial or sagittal CT scans. During operation, the OPLL could not be separated from the DO, and the whole ossified mass including OPLL and DO was removed completely. The arachnoid membrane was carefully preserved to avoid membrane defect. No CSF leakage developed after operation in this patient. **a** Preoperative radiographic image at neutral position; **b**, **c** preoperative sagittal and

axial CT scans demonstrating a C2–C3 local-type OPLL and C4–C7 continuous-type OPLL without double-layer sign; **d** preoperative MR image showed severe compression of the spinal cord at C4–C6 level; **e** postoperative radiographic image showing C4–C6 corpectomy and fusion; **f**, **g** postoperative sagittal and axial CT demonstrating complete resection of OPLL; **h** postoperative MR image showed sufficient decompression of the spinal cord; **i** intraoperative photo showed that the OPLL and DO was removed and arachnoid membrane was preserved

Clinical assessment

The Japanese Orthopedic Association (JOA) scoring system was used to evaluate the neurological status before and after the operation. An improvement rate (IR) was calculated as $IR = (\text{postoperative JOA score} - \text{preoperative JOA score} / 17 - \text{preoperative JOA score}) \times 100\%$. Surgical outcome was defined by the IR as follows: excellent ($IR \geq 75\%$),

good ($75\% > IR \geq 50\%$), fair ($50\% > IR \geq 25\%$), and poor ($IR < 25\%$).

Statistical analysis

For statistical analysis of results, SAS statistic software provided by Statistic Institute of Second Military Medical University was used. The Mann–Whitney *U* test and

Table 1 Radiological data the group with DO and the group without DO

	Group with DO (<i>n</i> = 40)	Group without DO (<i>n</i> = 98)	Total (<i>n</i> = 138)
Occupying rate (%)			
Mean	72.4 ± 8.2	41.2 ± 4.4	51.7 ± 4.2
Range	36–98	20–94	20–98
Extent of OPLL (no. of vertebrae)			
Mean	3.5 ± 0.4	2.8 ± 0.2	3.0 ± 0.2
Range	2–5	1–5	1–5
Type of OPLL [no. (%)]			
Local	5 (12.5)	34 (34.7)	39 (28.2)
Segmental	3 (7.5)	27 (27.6)	30 (21.7)
Continuous	15 (37.5)	18 (18.4)	33 (23.9)
Mixed	17 (42.5)	19 (19.4)	36 (26.1)
Double-layer sign [no. (%)]			
Yes	22 (55.0)	3 (3.1)	25 (18.1)
No	18 (45.0)	95 (96.9)	103 (81.9)
Preoperative cord flattening rate			
Mean	0.20 ± 0.06	0.26 ± 0.05	0.24 ± 0.05
Range	0.10–0.34	0.16–0.32	0.10–0.34
Postoperative cord flattening rate			
Mean	0.48 ± 0.12	0.45 ± 0.10	0.46 ± 0.08
Range	0.37–0.63	0.35–0.56	0.35–0.63

Fisher's exact test were performed. Levels of significance reaching 95% or more were accepted.

Results

Forty (29.0%) patients were found in the association with DO during the anterior decompression procedure. The patients were grouped into the group with DO (*n* = 40) and the group without DO (*n* = 98). No difference was found in age and sex between the two groups and the radiological characteristics and clinical results were compared as follows.

Radiologic findings

Mean OR of the group with DO reached to 72.4 ± 8.2% significantly higher than 41.2 ± 6.4% of the group without

DO (*P* < 0.001). OPLL of the group with DO extended 3.5 ± 0.4 vertebrae, which was also significantly more than 2.8 ± 0.2 vertebrae of the group without DO (*P* < 0.05). Furthermore, most OPLL of the group with DO was distributed as continuous- or mixed type. The double-layer sign was preoperatively observed in 25 patients and DO was intraoperatively found in 22 patients. All the patients had postoperative MRI examination in 1 week postoperatively. The result revealed that anterior decompression provided a significant increase in the cord diameter for all the patients (*P* < 0.01), and no significant difference in postoperative cord flattening rate was found between two groups (Table 1).

Diagnostic value of double-layer sign

In this study, the sensitivity (Se) and specificity (Sp) were calculated to evaluate the value of the double-layer sign to the diagnosis of association with DO, and the results show that the Se was 55% and the Sp was 96.9% for all the patients (Table 1). To evaluate its value in different subgroup patients, the patients were stratified according to their severity of the OPLL in the Table 2. The patients with OR < 60% were grouped as mild OPLL, and those with OR more than 60% as severe OPLL. Consequently, the Se' was 81.0% and the Sp' was 96.9% for the subgroup patients with mild OPLL, and the Se'' was 26.3% and the Sp'' was 97% for the subgroup patients with severe OPLL.

Clinical results

In this series, all the patients were followed-up for a mean time of 18 months (12–36 months). Generally, the neurological status was significantly improved. The mean JOA score increased to 14.1 ± 0.6 (7–16) points at the last follow-up, significantly higher than 8.8 ± 1.4 (4–14) points before surgery (*P* < 0.01). The IR of neurological function ranged from 12.5 to 87.5%, with a mean of 60.3 ± 12.5%. The surgical outcome was excellent in 28 (20.3%) patients, good in 75 (54.3%) patients, fair in 22 (15.9%) patients and poor in 13 (9.4%) patient. The clinical results compared between two groups were detailed in Table 3. Although both pre- and postoperative JOA score of the group with DO were lower than those of the group without DO (*P* < 0.05), there was no significant difference

Table 2 Susceptibility of double-layer sign to dural ossification

	Mild OPLL (OR < 60%)		Severe OPLL (OR ≥ 60%)	
	Single-layer sign	Double-layer sign	Single-layer sign	Double-layer sign
Dural ossification [no. (%)]	4 (2.9)	17 (12.3)	14 (10.1)	5 (3.6)
No dural ossification [no. (%)]	63 (45.7)	2 (1.4)	32 (23.2)	1 (0.7)

Table 3 Surgical outcome of the group with DO and the group without DO

	Group with DO (<i>n</i> = 40)	Group without DO (<i>n</i> = 98)	Total (<i>n</i> = 138)
Preoperative JOA score			
Mean	8.7 ± 1.6	9.3 ± 1.4	8.8 ± 1.4
Range	5–12	4–14	4–14
Postoperative JOA score			
Mean	13.2 ± 1.3	14.4 ± 0.7	14.1 ± 0.6
Range	11–16	7–15	7–16
Improvement rate (%)			
Mean	63.2 ± 15.2	59.5 ± 12.7	60.3 ± 12.5
Range	22.2–87.5	12.5–77.8	12.5–87.5
Surgical outcome [no. (%)]			
Excellent	8 (20.0)	20 (20.4)	28 (20.3)
Good	20 (50.0)	55 (56.1)	75 (54.3)
Fair	9 (22.5)	13 (13.3)	22 (15.9)
Poor	3 (7.5)	10 (10.2)	13 (9.4)

in the IR of neurological function, and the surgical outcome was comparable between two groups.

CSF leakage and other complications

Cerebrospinal fluid leakage was the main postoperative complication in this series, and 18 patients experienced CSF leakage after operation. Among them, 15 patients had significant sign of DO. If CSF leakage occurred, drainage tube was pulled out in 12 h after operation, and continuous pressure to the wound was performed. To our experience, CSF leakage usually stopped after 3–5-day conservative treatment of local pressure, although three patients experienced a CSF pseudocyst. Dural tear healed and the pseudocyst gradually disappeared after 2–3 months of repeated puncturation and aspiration. Other complications included bilateral C5 palsy in two cases and hematoma in two cases. C5 palsy developed at 8–24 h postoperatively, and the strength of related deltoid and biceps muscles decreased to grade 1 in manual muscle test (MMT). The conservative treatment was used including neurotrophs drugs, high-pressure oxygen therapy and functional exercises. Strength of the paralyzed muscles recovered to four in MMT after 3 months. The patients who experienced hematoma recovered in neurological function after an emergency operation.

Discussion

Ossification of the posterior longitudinal ligament is also one of the major diseases in Chinese population, in which cervical myelopathy or radiculopathy develops. Because

the dural membrane is sometimes involved in ossification, having the capability to anticipate association with DO when multilevel anterior cervical corpectomy with fusion (ACCF) is performed for OPLL would facilitate operative planning. One of the important functions of dural mater is protection of the nervous tissues from epidural harm. Nevertheless, once this membrane is ossified in the association with OPLL, it becomes very difficult to separate the PLL from the DO. CSF leakage through the defect portion of the dura mater is one of the common complications encountered when using anterior approach for direct removal of cervical OPLL in this condition, and the increased opportunity for spinal cord or nerve root damage may cause disastrous neurological deterioration [2, 6, 10]. Therefore, recognizing dural penetration is very important to anterior cervical direct decompression for OPLL. The patients should be informed with higher risk in preoperative education, and the surgeon was alerted to need more cautious manipulation to avoid unnecessary complications including CSF leakage and injury to the nervous tissues.

Although the precise mechanism of DO has not been elucidated, the characteristics of DO associated with OPLL have been documented by several authors. Hida et al. [4] first described two CT findings that indicated the association with DO. The single-layer sign, consisting of the demonstration of a single homogeneous ossified OPLL mass, was found in only one of nine patients. The double-layer sign, characterized by anterior and posterior ossified rims separated by hypertrophied PLL, appeared more specific for DO, because it found in 10 of 12 patients. Mizuno et al. [9] and Min et al. [7] agreed that the ossified mass presented with double-layer sign on CT scans has the increased possibility of DO than single-layer sign (or en bloc type) in Japanese and Korean population. However, Epstein [3] reported that a kind of C sign in addition to the current modified single-layer sign, defined by an irregular hook-like angled mass of OPLL, served to predict DO with OPLL with moderate accuracy in the North American population.

In this study, the significance of double-layer sign was evaluated to identification of DO in Chinese population. The Sp was 96.9%, but the Se was only 55% for all the patients. Because the Chinese patients usually had a longer history of the disease with more severe OPLL when they decided to receive operation, we also investigated the significance of double-layer sign in different subgroup patients with mild or severe OPLL. The results showed that the double-layer sign was more sensitive in the patients with mild OPLL, and it was less frequent in those with severe OPLL associated with DO. The fashion of the OPLL development could be responsible for this result. The ossification of mild OPLL begins from the superficial as well as the deep layer of the PLL, which is separated by a

hypertrophied portion of the ligament, forming a double-layer sign and cause dural penetration. However, the layer which consisted a nonossified degenerated PLL could disappear with the increasing of the OPLL severity. The results in this study alerted Chinese surgeon to be more cautious when removing severe OPLL from dural mater, although there was no double-layer sign on preoperative CT scan.

Mizuno et al. [8] has described a surgical technique for DO associated with OPLL, in which the OPLL was separated from DO through a thin layer consisting a nonossified degenerated PLL, resulting in the avoidance of dural defect and CSF leakage. However, it was not that there was a nonossified PLL layer in all the patients with DO, as showed by our results that the double-layer sign is uncommon in the patients with severe OPLL. In our technique for these patients, the entire ossified mass including OPLL and DO was removed completely. This technique needed to preserve the arachnoid membrane with the aid of microscope to avoid a large area of membrane defect, resulting in uncontrolled CSF leakage. In fact, several patients in our series using this technique for removal of OPLL and DO had CSF leakage postoperatively. However, because the area of membrane defect was limited, suture or fascia graft reported in the literature seemed to be unnecessary because CSF leakage could be cured just by conservative treatment to our experiences [1, 11].

In the anterior cervical procedure for our patients with OPLL, the ossified PLL mass was completely removed with DO or without DO, and there was no significant difference in clinical results between the group with DO and the group without DO. The MR studies in this study also showed that anterior approach provided sufficient decompressive effect for the patients with DO, which was similar to the patients without DO. Therefore, ACCF is meritorious for the patients with OPLL associated with DO, although more difficult manipulation and higher risk of CSF leakage.

Conclusion

Although the double-layer sign on preoperative CT scan is a relatively specific indication for the association with DO, there were a considerable number of the patients with DO presenting with single-layer sign, especially those with severe OPLL, according to the result of this study. This alerted the surgeon to the increased possibility of DO when they encounter a double-layer sign in the patients with mild OPLL as well as those with severe OPLL. Removal of the ossified PLL mass with DO or without DO could provide a similar decompressive effect with the patients without DO through anterior cervical approach, which was simultaneously accompanied with more difficult manipulation and higher risk of CSF leakage.

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