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의학석사학위논문

Radical excision of lumbosacral lipoma

:An early experience of 'followers'

요천추 지방종의 근치적 절제

: '후발주자'의 초기 경험

2018년 2월

서울대학교 대학원

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이 논문을 의학석사학위논문으로 제출함

2018 년 2월

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정 상 준

정상준의 석사학위논문을 인준함

2018 년 1월

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Abstract (English)

Introduction:

Indication, timing and method for surgical treatment of lumbosacral lipoma is controversial. Studies of the natural history of asymptomatic lumbosacral lipoma report that the deterioration rate over 10 years is approximately 35%~45%. Along with this, incidence of neurological or urological complications associated with surgery in the asymptomatic lumbosacral lipoma is about 5%. Radical resection of the lumbosacral lipoma and complete reconstruction of the placode is supported in that better long-term outcome can be achieved with low complication rate. We analyzed the outcomes of lumbosacral lipoma treated with the aggressive untethering and radical excision of fat in the 5 years of initial learning phase.

Methods and Results:

From November 2009 to December 2014, there were 81 fresh cases of lumbosacral lipoma with dorsal, transitional and chaotic types and true lipomyelomeningocele (LMMC). Caudal and filar types were excluded. Complete untethering was accomplished in 79 cases (98%). Radical excision of the lipoma was attempted in all cases and achieved in 67 cases (83%). Postoperative neurological complication was observed in 8 cases (10%). Out of 8 patients, 4 patients were showed no abnormality on the preoperative examinations. Group of lipoma types (dorsal + transitional vs. chaotic + true LMMC) and availability of radical lipoma excision turn out to be factors related to neurological outcomes in univariate analysis ($p<0.001$ and $p=0.027$, respectively). Group of lipoma types, availability of radical excision and postoperative cord/sac ratio are related factors in multivariate analysis ($p=0.025$, $p=0.049$ and $p=0.031$).

Conclusions:

As a follower of aggressive untethering and radical excision of fat, careful consideration is required to plan the surgical treatment of lumbosacral lipoma on account of the ‘underestimated’ complication rate. Type of the lipoma is the important factor determining the surgical outcome.

Keywords: lumbosacral lipoma, neurological outcome, radical excision, complications, untethering

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Introduction

Lumbosacral lipomatous malformation is commonly called as 'lumbosacral lipoma' though it is not a true neoplasm. It is classified into dorsal, transitional, caudal, filar, and chaotic types according to the anatomical features which are associated with different surgical procedures and outcome (1-3). The literature on natural course rates 33-47.3% of symptomatic progression (4-6) and wide range of symptomatic relief after untethering surgery which led to debates on the value of prophylactic surgery in asymptomatic cases. Moreover, timing and the method of operation are varied according to surgeons. In 1997, the Paris group (7) published an article on worse outcome of partial resection compared to natural course in the lumbosacral lipoma and suggested conservative policy in asymptomatic patients. In 2009, however, Pang et al. (3, 5) reported an excellent outcome after radical resection of lipoma with voluminous dural reconstruction in asymptomatic patients. The surgery on the lumbosacral lipoma is not without risk whether the extent of lipoma resection is partial or radical, and the postoperative neurological (including urological) complication rates were reported 4.2%-7.9% (3, 7).

Currently the radical resection of lipoma has been gradually spread among the pediatric neurosurgeons although surgical indications in asymptomatic patients are still in debate. Since the operative technique requires experience and systematic intraoperative monitoring is required, there is a limit to be implemented in many centers. In addition, although pioneer has reported excellent progression free

survival with overwhelming stability, it is necessary to confirm whether this invasive method can safely deliver the same results to other pediatric neurosurgeons as compared with conventional partial resection. It is an issue that must be verified in order to judge whether this treatment can be widely practiced.

Our group has introduced policy of radical resection of lipoma with intraoperative electrophysiological monitoring (IOM) in 2009. Thereafter we experienced a learning curve in the aspects of lipoma surgery and IOM. Because the previously reported outcomes after radical resection of lipoma are from the pioneers and world-leaders in this field, we present our postoperative early neurological outcome data as one of the 'followers' of radical resection surgery with IOM to share the information on the phase of learning curve with those who started this method later than the leaders. We investigated on the rates of complete untethering, radical resection of lipoma and postoperative complication at postoperative 6 months and analyzed the factors influencing outcome.

Material and Methods

Patient Population

We retrospectively reviewed 81 patients who were surgically treated with a fresh lumbosacral lipoma excluding caudal and filar types in our institution from November 2009 to December 2015. There were 40 male patients and 41 female

patients. The median age of patients was 3 months, ranging from 1 month to 42 years (Table 1).

Table 1. Demographic and preoperative clinical characteristics of the surgically treated 81 lumbosacral lipoma patients

Median Age	3MO (1MO-42YO)
Sex (M:F)	40:41
Abnormality on evaluation	
Abnormal	40 (49%)
Free	41 (51%)
Lipoma type	
Dorsal	20 (25%)
Transitional	44 (54%)
Chaotic	4 (5%)
True LMMC	13 (16%)
Syrinx	
Present	36 (44%)
Absent	45 (56%)

MO: months old, YO: years old, M: male, F: female, LMMC: lipomyelomeningocele

The indication for surgery was one of the following cases: 1) new appearances of neurological symptoms including voiding or defecation problems or exacerbations of preexisting symptoms (n=40), 2) abrupt increases in the size of an extraspinally herniated sacs that may aggravate tethering (n=5), 3) abrupt increases in the size of

intraspinal lipomas that compress the spinal cord (n=7), 4) cases in which syringes develop or aggravate during follow-up (n=15), 5) weak cord pulsations on ultrasonography (US) which suggest tethering (n=5), and 6) difficulty in careful inspection to detect changes of symptoms by caregivers (n=9).

Because of the small spinal canal and postoperative enlargement of residual lipoma in young infants, surgery was performed after 3 months of age unless early surgery is indicated. If neurological symptoms, signs or abnormalities in electrophysiological and urodynamic studies (UDS) were detected, especially when they were progressive, or if remarkable enlargement of the syrinx or extraspinally herniated sac was noted, early surgery was indicated (8). Slit-like structures less than 2 mm in the spinal cord were considered dilated or persistent central canal rather than syrinx (9).

The preoperative evaluation included neurological examination, electromyography (EMG), nerve conduction test, and urologic evaluation including UDS. For the radiological evaluation, US was used as a screening test and spine magnetic resonance imaging was taken to confirm the diagnosis, to evaluate the lesion, and to make a surgical plan. Electrophysiological and UDS were performed after 2 months of age. In the electrophysiologic study, evidence of new or ongoing activity of deterioration was checked. In urological evaluation including UDS, the following items were judged to be abnormal: 1) occurrence and aggravation of urinary symptoms such as incontinence, urgency, frequency or dribbling continued even

after the end of bowel training 2) Repeated urinary tract infections, 3) increase in postvoid residual urine volume, 4) an underactivity or hyperactivity of a detrusor by neurogenic cause that consistently and persistently occurs, 5) the presence of detrusor-sphincter dyssynergia, 6) presence of vesicoureteral reflux or 7) The presence of hydronephrosis.

Of the 55 (68%) patients who showed no symptoms or signs at the time of surgery, 15 (15/55=27%) patients had preoperative abnormalities in the electrophysiological study or UDS. Therefore, 40 (49%) patients had no apparent abnormalities in the preoperative evaluation, and 41 (51%) patients had abnormal findings in the preoperative examination. In the present article, 'neurological symptoms/signs/complications' include urologic and bowel problems as well as motor and sensory dysfunctions.

The lipomas were classified as dorsal, transitional, chaotic or true lipomyelomeningocele (true LMMC) (Figure 1). Caudal and filar lipomas were not included in this study. Dorsal and transitional followed classical Chapman's classification, and chaotic followed Pang's classification (2, 3). True LMMC is defined as a lesion in which the distal spinal cord and cerebrospinal fluid (CSF) space are herniated out of the spinal canal through the lamina defect. According to these criteria, there were 20 cases (25%) of dorsal type, 44 cases (54%) of transitional type, 4 cases (5%) of chaotic type and 13 cases (16%) of true LMMC (Table 1). A syrinx was found in 36 (44%) cases before the surgery.

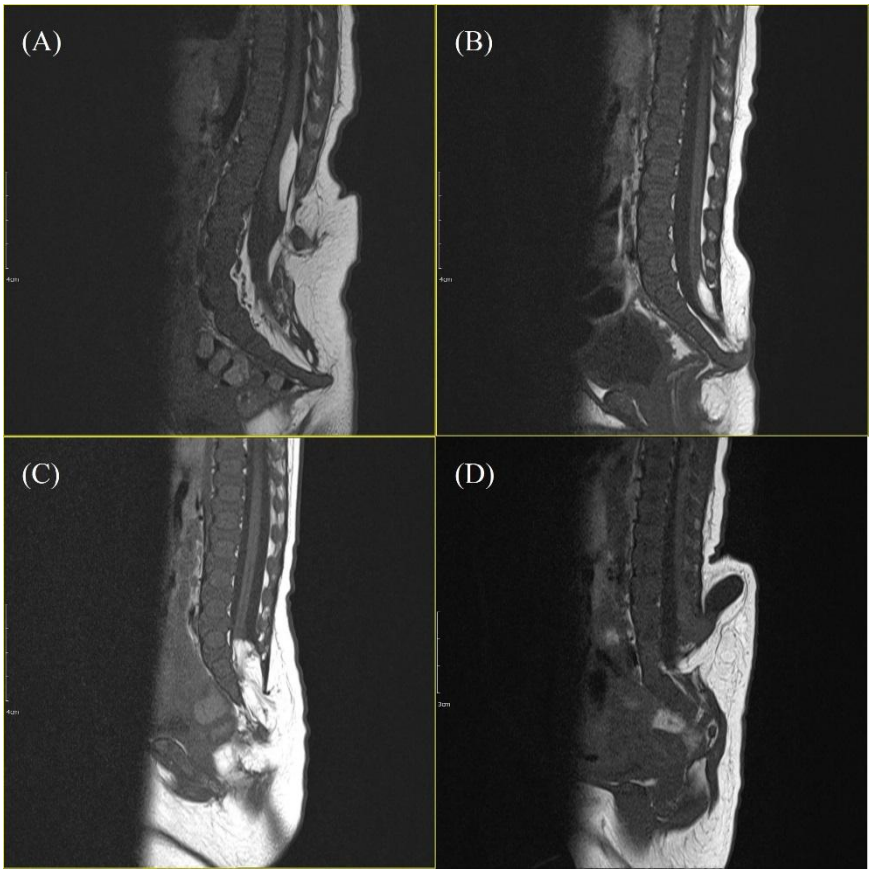

Figure 1. Classification of lumbosacral lipomas. (A) Dorsal lipoma: Lipoma is attached only to the dorsal aspect of the spinal cord. Conus medullaris is not affected. **(B) Transitional lipoma:** Lipoma is attached to both the dorsal part and the caudal part including conus medullaris of the spinal cord. **(C) Chaotic lipoma:** The caudal part of the lipoma is also present on the ventral side of spinal cord and encases not only conus medullaris but also nerve roots including ventral roots. **(D) True lipomyelomeningocele:** Spinal cord, lipoma, and dural sac containing cerebrospinal fluid are herniated from the spinal canal through laminal defects.

Operative Techniques

All the cases underwent untethering and attempted radical resection of the lipoma. When only the spinal cord, the fibrous tissue between the fat and the cord (so called 'white plane'), functioning roots with minimal amount of surrounding fat are remained, we regarded it radical resection (Figure 2). For radical resection of lipoma, microscissors or small sharp scissors were commonly used. However, if the interface between the lipoma and spinal cord is of irregular shape or too much concave, we used an ultrasonic aspirator (10, 11). After radical resection of the lipoma, neurulation was done through pial reconstruction. If necessary, duraplasty was done with dural substitute made of porcine intestinal submucosa (SurgiSIS, Cook Biotech Inc, West Lafayette, Ind). In cases where the range of exposure included more than two levels of the lumbar spine, a laminotomy rather than laminectomy was performed and the reconstructive laminoplasty was done afterward.

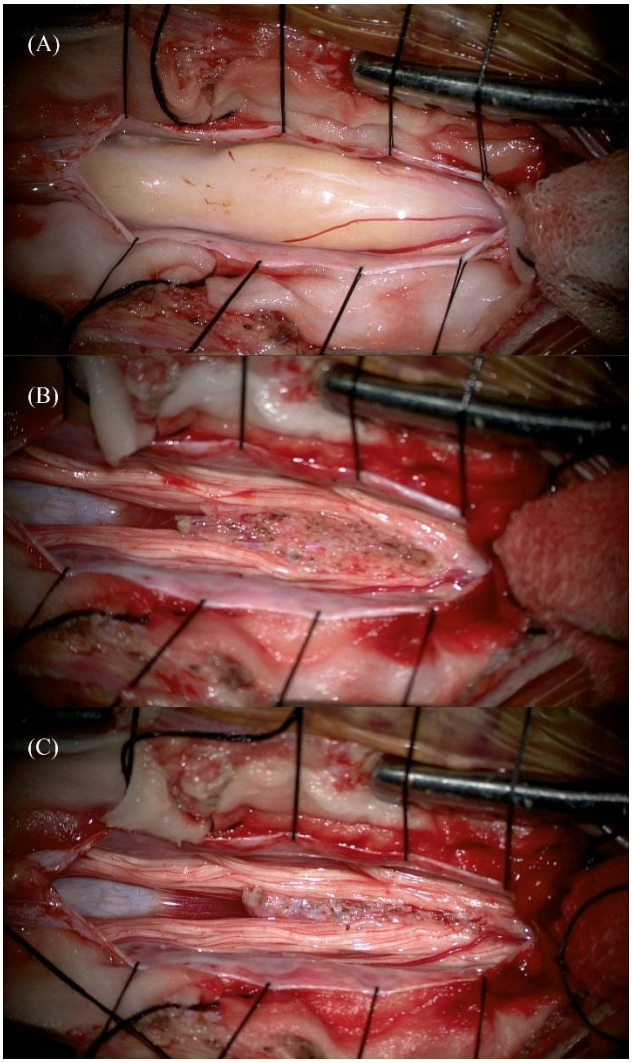

Figure 2. Surgical photographs of radical excision of lumbosacral lipoma during untethering. (A) Yellow bulky lipoma is attached dorsal side of the spinal cord. **(B)** After radical excision of lipoma, white plane of the spinal cord is exposed. **(C)** Neurulation is accomplished through pial reconstruction by suturing the pial membrane at the bilateral edge of the removal site.

Regarding IOM, until October 2009 gross motor responses on stimulation by a nerve stimulator were visually checked and the monitoring of the anal sphincter was not possible. In November 2009 when the first case of the present series was operated on, monitoring and mapping by electromyographic responses including anal sphincters became possible. Concentric electrodes were used for stimulation. In May 2012, a specialized IOM team started to support surgery with additional monitoring of somatosensory and motor evoked potentials, and bulbocavernous reflex (12). IOM was performed using the NIM-Eclipse nerve monitoring system (Medtronic Xomed Inc.).

Outcome Analysis

The rates of complete untethering and radical resection of lipoma were calculated.

The neurological outcome of surgical treatment was determined by evaluating motor, sensory, voiding, and defecation at postoperative 6 months. If the patient complains new symptoms or progression of symptoms compared to the preoperative state and shows evident worsening in neurological, electrophysiological and urological examinations, it was regarded as neurological deterioration. The factors affecting the surgical outcome including gender, age, presence of preoperative symptoms and signs, type of lipoma, presence of preoperative syrinx, operator, performance of radical resection of lipoma, pial reconstruction or duraplasty, and

postoperative cord/dural sac (C/D) ratio were analyzed. Fisher exact test was used for univariate analysis of categorical data, and multivariate analysis was performed using logistic regression analysis. Statistical analyses were performed using SPSS 22 (IBM Corp., Armonk, NY, USA).

Results

Rates of complete untethering and radical resection of lipoma

Complete untethering was possible in 79 (98%) cases except two of chaotic type. Radical resection of lipomas was possible in 67 (83%) cases. Of the 14 patients who did not undergo radical resection, two patients were chaotic type. In the remaining 12 cases, radical resection was unaccomplished because of decreased intraoperative motor evoked potentials (MEP, N=2) or presence of EMG responses on electrical stimulation with low amplitude of 0.5 mA at the lipoma near the root exit zone (N=10). These 12 cases were operated on in the early period of this study.

Neurological outcomes

Postoperative neurological outcome of the patients is summarized in Table 2. Of the 41 patients who had no neurological abnormality in the preoperative evaluation, 37 (90%) patients remained normal but 4 (10%) were worsened. Of the 40 patients

who had abnormalities in the preoperative examination, 20 (50%) were improved, 16 (40%) remained static, and 4 (10%) became worse. Among the 81 total patients, 8 (10%), of whom 4 had preoperative neurological abnormalities and 4 had not, showed postoperative deterioration. In detail, there were 3 patients with both weakness and voiding problems. Another 3 had only weakness and the remaining 2 had voiding problems with or without defecation problems. Other surgical complications occurred in 4 cases: 2 cases of wound dehiscence, 1 of CSF leakage which needed revision and 1 of wound infection.

Table 2. Neurological outcome of the patients

Normal	37 / 41 (90%)
Improved	20 / 40 (50%)
Static	16 / 40 (40%)
Worsen	8 / 81 (10%)
Preoperatively free	4 / 41 (10%)
Preoperatively abnormal	4 / 40 (10%)

Both of the dorsal and the transitional types showed a low neurological complication rate of 5% and 2%, resepectively. However, the neurological complication rates of the chaotic type and true LMMC were 50% and 29%, respectively (Table 3). When the factors affecting the neurological outcome were examined, it was found that there was a correlation between lipoma types and the outcome among preoperative factors. The difference in neurological complication rates according to the type was statistically significant (p=0.002), more pronounced

when the dorsal and transitional types were considered low risk group, whereas the chaotic type and true LMMC as high risk group (p=0.001) (Table 4). Among the operative factors, availability of the radical excision was associated with neurological outcome (p=0.027). These factors were also found to be statistically significant in multivariate analysis (p=0.025 and p=0.049, respectively). In addition, the postoperative C/D ratio turned out to be associated with postoperative neurological outcomes.

Table 3. Neurological outcome according to lipoma types

	No worsening	Worsening	Total No. of patients
Dorsal	19 (95%)	1 (5%)	20
Transitional	42 (98%)	1 (2%)	43
Chaotic	2 (50%)	2 (50%)	4
True LMMC	10 (71%)	4 (29%)	14
Total	73	8	81

Analysis of the correlation between perioperative factors and the group of lipoma types (high risk vs. low risk) showed a correlation between the group of lipoma types and age group at operation, preoperative syrinx and completeness of pial reconstruction (Table 5). In the high risk lipoma group, 72% of the patients required surgical treatment at early infant younger than 3 months of age, whereas in the low risk lipoma group only 28% (p=0.002) did so. Preoperative syrinx was observed in 52% of patients in the low risk group, but only in 17% of patients in the high risk group (p=0.008). Finally, complete pial reconstruction was performed in 89% of

patients in the low risk group, but only in 67% of the high risk group (p=0.034).

Table 4. Univariate and multivariate analysis of the preoperative or operative factors on neurological outcome.

Variables		Reference	Univariate[a]			Multivariate[b]		
			OR	95% CI	*p* value	OR	95% CI	*p* value
Pre-operative factors	Gender	male	1.713	0.381-7.701	0.712	12.182	0.567-261.821	0.110
	Age group	<3mo	0.352	0.078-1.591	0.253	0.010	0.000-1.622	0.467
	Lipoma group	low risk	15.250	2.742-84.810	0.001*	161.716	1.912-13680.733	0.025*
	Syrinx	absent	0.155	0.018-1.325	0.070	0.197	0.005-8.210	0.393
	Neurological status	normal	1.028	0.239-4.425	1.0	0.070	0.003-1.463	0.086
Operative factors	Radical excision	non-radical	0.159	0.034-0.739	0.027*	0.005	0.000-0.988	0.049*
	Pial reconstruction	incomplete	0.265	0.055-1.283	0.113	0.075	0.003-1.919	0.117
	Duraplasty	not done	1.098	0.244-4.954	1.0	0.047	0.001-1.539	0.086
	Postop. C/D ratio	<50%	0.422	0.074-2.418	0.297	0.001	0.000-0.517	0.031*

OR: odds ratio, 95% CI: 95% confidence interval, mo: months old, Postop. C/D ratio: postoperative cord/dural sac ratio

a: Fischer's exact test, b: logistic regression, * $p < 0.05$

Table 5. Factors related to the group of lipoma types

Factors	Reference	OR	95% CI	*p* value
Gender	male	1.290	0.450-3.697	0.790
Age group	<3mo	0.166	0.052-0.532	0.002*
Preoperative syrinx	absent	0.182	0.048-0.691	0.008*
Preoperative abnormality	normal	2.929	0.995-8.623	0.059
Radical excision of lipoma	non-radical	0.291	0.085-0.994	0.071
Completeness of pial reconstruction	incomplete	0.250	0.071-0.878	0.034*
Duraplasty	not done	2.800	0.829-9.458	0.107
Postoperative C/D ratio (<50%, ≥50%)	<50%	3.208	0.382-26.910	0.441

Fischer's exact test, * p <0.05

OR: odds ratio, 95% CI: confidence interval, C/D ratio: cord-sac ratio

Discussion

Our rate of complete untethering was 79/81 (98%). Although our indication of surgery is rather conservative, we attempted complete untethering and maximum safe decompression as much as possible. In 2 cases of chaotic type, however, the

intra-lipoma location of ventral roots precluded aggressive complete untethering in spite of various surgical methods. In some cases of complex anatomy, neurological status was deteriorated even untethering was complete. Our experience of IOM (data not shown) suggested that the neurological damage occurred mainly at the time of untethering rather than the resection of lipoma.

The rate of radical resection of lipoma was 67/81 (83%). Again, chaotic type was the main hurdle for the radical resection of lipoma in 2 cases. In early years when the other 12 cases of incomplete resection of lipoma were operated on, our team was anxious about the cord injury during the procedure of radical resection of lipoma. We stopped radical resection if frequent and sustained abnormal EMG responses occurred or significant changes in MEP (a decrease in amplitude >50% or prolongation in latency >10%) were observed even transiently. We thought that the removal of additional small amount of fat tissue with a risk of cord injury is not acceptable. However, as we found that those cases showed no postoperative deficits, the range of fat removal became gradually more radical.

The neurological outcome in this series is marked by the fact that the proportion of poor outcome groups with neurological deterioration (10%) is higher than that reported by previous leading groups, 4.2%-7.9% (3, 7). This is probably because of the exclusion of caudal or filar lipoma which requires relatively simple and safe procedures in this study. If caudal lipoma is included, the rate of neurological complications is reduced to 7%. In addition, the operation policy of our institute,

aggressive untethering is attempted once the operation is determined although the indication for surgery is rather conservative, the initial learning curve of radical resection of lipoma and newly introduced and less settled IOM might also influence the result. The rate of exacerbation was similar in all patients, with or without preoperative abnormal findings. In patients whose preoperative work-up results were normal, not a few patients worsened after the operation. This means that the risk of untethering itself is not low. Surgeons should be careful with this in mind when deciding the surgery.

In cases of chaotic type lipoma or true LMMC, untethering is not easy due to complicated and distorted anatomical structures, and there is a possibility of deterioration during this process (1, 3). Our results show that patients in this group have much worse neurological outcomes.

Whether radical lipoma excision was possible is not likely to affect neurological damage directly. However, this is an indirect reflection of how complex the lesions were. Availability of radical excision means that the lesions were clear to identify the anatomical structure so that untethering can be achieved properly as well as removal of lipoma and fibrous tissue. The statistical significance of the postoperative C/D ratio in multivariate analysis and the availability of complete pial reconstruction as correlated with group of lipoma types are also probably due to some influences of complex anatomy to these factors.

The difference in anatomical complexity according to the group of lipoma types also affected the operation timing. Although the age did not affect the neurological outcome on univariate analysis, the group of high-risk lipoma types required surgical treatment at a relatively early infancy. This suggests that thorough and frequent clinical attention is needed in these high-risk group patients from a very young age.

One of distinct features in our results is that preoperative syrinx was observed more frequent in the low risk lipoma group. However, there is no correlation between preoperative syrinx and postoperative neurological outcome. This seems to be different from the known evidence and our clinical experiences that syrinx is associated with neurological deterioration (6, 13). Perhaps, this may be related to age at surgery. In the high-risk lipoma group, proportion of the patients who got the operation at younger age (< 3 mo) group was much larger than in the low-risk group, which might be the result of our surgical indication. This period might be too early to form the syrinx in the patients. The results of the correlation between age group and preoperative syrinx support in our data support this indirectly (Table 6). Our results do not seem to indicate that preoperative syrinx is less common in complex lipomas unless there is no comparison at similar age.

Table 6. Distribution of patients according to the age group and preoperative syrinx

	No syrinx	Syrinx	Total
<3mo	23	9	32
>=3mo	22	27	49
Total	45	36	81

Chi square test, Odds ratio = 3.136, 95% confidence interval: 1.208-8.145, *p*=0.022

Initially we thought that the neurological outcome may be worse in early years of this study because of our lack of experience in radical resection of lipoma and IOM. However, our yearly trend of postoperative neurological complication rates showed that it was not influenced by the year of operation but depended on the proportion of the group of high risk lipoma types. The learning curve of the new methods, radical resection of lipoma and IOM, had less impacts compared to the lipoma type during this period.

One of the concerns is how the specific changes of the IOM have affected the postoperative neurological outcome (14-16). However, since the systematic IOM was mainly carried out in the latter part of the included period, there was a limit in analyzing these results. This can be further investigated through subsequent studies.

Conclusion

Complete untethering and radical excision of lipoma was achieved in the most lumbosacral lipoma cases. Except for simple filar and caudal lipomas, the postoperative neurological complication rate which includes voiding and defecation problems was higher than the previous reports. There was no difference of this rate in the group that had no abnormality in the preoperative examination, which implies careful consideration is needed when deciding the surgery. In particular, complex lumbosacral lipomas such as chaotic type and true lipomyelomeningocele, there was a significant correlation with postoperative neurological deteriorations.

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초 록

서론:

요천추 지방종의 수술의 적응증, 수술시기 및 방법에 대해서는 논쟁의 여지가 있다. 무증상의 요천추 지방종의 자연 경과를 살펴본 연구들에 따르면 10년 동안 악화되는 비율은 35%~45%되는 것으로 알려져 있다. 한편, 무증상 요천추 지방종의 수술과 관련된 신경학적, 비뇨기과적 합병증은 약 5% 정도이다. 요천추 지방종의 근치적 절제와 함께 신경기원판을 완벽하게 재건해주는 수술방법은 수술 후 장기성적이 우수하고 합병증의 비율이 낮다고 알려져 있어 지지를 받고 있다. 우리는 적극적인 척수결박해소술과 지방종의 근치적 절제를 시행한 초기 5년의 성적을 분석하여 보았다.

방법 및 결과:

2009년부터 2014년까지, 81 건의 배측(背側)형(dorsal type), 이행형(transitional type), 혼재형(chaotic type)의 척수원뿔지방종 및 진성 척수지방수막류(true lipomyelomeningocele, true LMMC) 첫수술 증례들이 있었다. 미부형(尾部)형(caudal type)과 종사형(filar type)은 제외되었다. 완전한 척수결박의 해소는 79례(98%)에서 가능하였다. 지방종의 근치적 절제는 모든 증례에서 시도되었고 67례(83%)에서 근치적절제가 이루어졌다. 수술후 신경학적합병증은 8례(10%)에서 나타났다. 8명 중, 4명의 환자는 수술전 검사에서 이상소견이 관찰되지 않았다. 지방종형의 그룹(배측형+이행형 대 혼재형+진성척수지방수막류)과 지방종의 근치적 절제 가능여부가 단변량분석에서 수술후 결과에 영향을 주는 요인으로 나타났다(각각 $p<0.001$, $p=0.027$). 지방종형의 그룹, 지방종의 근치적 절제 가능여부 그리고 수술

후 척수/경막비가 다변량분석에서 유의한 결과를 보여주었다(p=0.025, p=0.049, p=0.031).

결론:

후발주자로서 적극적인 척수결박해소술과 지방종의 근치적절제를 시행함에 있어, 요천추 지방종에 대한 수술적 치료계획 수립시에 수술과 관련된 합병증을 과소평가해서는 안되며 신중한 결정을 내리는 것이 필요하다. 수술결과에는 해부학적 복잡도의 반영인 지방종형의 그룹이 중요한 요소이다.

주요어: 요천추 지방종, 신경학적 결과, 근치적 절제, 합병증, 척수결박해소술

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