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의학석사 학위논문

정맥 기형에 대한 1차 치료로서의 외과
적 절제

Surgical excision as a first-line
treatment for venous malformations

2020년 01월

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이 논문을 의학석사 학위논문으로 제출함

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Abstract

Surgical excision as a first–line treatment for venous malformations

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Venous malformations (VMs) are results from an error in vascular morphogenesis. Congenital slow–flow vascular anomalies, especially involve the head and neck. VMs are blue, soft, and compressible. Lesions are composed of thin–walled, dilated, sponge–like channels of variable size and mural thickness. (1) As usual, sclerotherapy is the first–line treatment for VMs patients. (2) Here, we investigated surgical treatment is also a good

choice for VMs patients as a first-line treatment.

This retrospective study included patients who presented with the diagnosed with VMs from 2009 to 2019 registered at Seoul National University Children's Hospital. The study consists of four parts; 1, VM grading, 2, resection rate measurement, 3, peer review (3) evaluation of pre-op. and post-op. photographs, and 4, statistical analysis of results.

Surgical results were evaluated in two ways. One was the resection rate which was defined as the proportion of postoperative amount volume to the preoperative amount. The other one was peer-review evaluation contains resection score, contour score, and scar score (Table. 1). The results were evaluated by three board-certified plastic surgeons. (3) One-way ANOVA was used as a statistical analysis of results (IBM SPSS Statistics 25).

A total of 102 patients and 126 surgical procedures were evaluated. The mean follow-up time was 2.2 years (range: 0–7.3 years). Two patients had two multiple lesions, resulting in a total of 104 lesions for VMs (Pure VMs, 102 cases, and combined type VMs, 2 cases). In 46 patients, we compared lesion size (volume and max diameter) between the preoperative MRI and postoperative sonography. The average resection rate was $87.59 \pm 26.52\%$ and 32 patients were completely cured. Peer review results are shown in Table 6. The mean resection score, contour score and scar score by peer review were 3.96 / 5.00, 3.18 / 4.00, and 2.98 / 4.00, respectively. According to the results of statistical analysis, we found that the higher the clinical classification grade, the lower the postoperative score (low scores indicate poor results). The incidence rate of complications was 15.08% (19/126), including recurrence, infection, dehiscence, anal fistula, hematoma, revision surgery, and blood transfusion. (4)

We have presented the evaluation results of 102 patients who have

undergone surgical treatment as a first–line treatment for VMs. The resection rate was 87.59% in 46 patients and the resection score was 3.95 / 5.00 of 126 procedures. It shows that surgical treatment is effective enough as a first–line treatment of VMs.

Keywords: Venous malformations/ evaluation/ surgery/ retrospective/ pediatric

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1. Introduction

Vascular anomalies are called by various names, and in 1982, Mulliken and Glowacki successfully divided tumors and malformations according to their biologic endothelial cell characteristics. (1) Vascular malformations are divided into fast-flow vascular malformations and slow-flow malformations. Then, slow-flow vascular malformations are divided into capillary, venous, lymphatic, and combined vascular malformations.

The first-line treatment for venous malformation is non-surgical treatment such as sclerotherapy. (5) Sclerotherapy has been used extensively for the treatment of VM. (2) The sclerosants most commonly used include ethanol and sotradecol. Complications of sclerotherapy include skin and mucosal injury swelling leading to airway compromise, infection, and nerve injury. Also, laser therapy provides good control of VM. (6) The Nd: Yag laser can be used via a fiber attached to an endoscope to treat intraoral and airway venous malformations.

On the other hand, surgical treatment was not chosen as first-line treatment because the total lesion is difficult to remove by surgical treatment. The risk of recurrence rate is higher because hidden channels adjacent to the visible lesion are not excised. However, some studies have reported that surgical treatment combined with sclerotherapy has a better result than single sclerotherapy or single surgical treatment. (8) Research with single surgical treatment, long-term follow-up, and the big-scale result is rare. So we tried to analyze our surgical results. For VM diagnosis, a physical examination is very important. Unlike involuted hemangioma, the VM does not regress and grows as the child grows. Thus, radiological measurements are used to define the lesion prior to surgery. For example, MRI data show the depth of invasion, distribution, location of the lesion, and grade using size and margin. (7) CT and D-USG are also helpful for diagnosis, especially D-USG is one of the diagnostic methods with good caustic ratio.

Thus, in this study, we analyzed the result of venous malformations with single surgical treatment by investigating its clinical and physical features, postoperative complications, and the outcome of surgical treatment evaluation. The aim of this study was to evaluate the clinical outcome of surgical treatment for VMs.

2. Patients and methods

This retrospective study included 102 patients who were diagnosed with VMs in our hospital from 2009 to 2019. The suspected VMs were diagnosed based on the result of patients' history, Doppler sonography findings, magnetic resonance imaging findings, and intra-operation biopsies results. According to ISSVA classification-2014, there are 4 types of VMs. In our study, only a simple VM was included. 102 patients with 104 lesions were undergone surgical treatment as a first-line treatment. This study was conducted with the written informed consent of the subjects and was approved by IRB. Patients' data from the Electronic Medical Record were collated to a chart. Age, blood types, height, weight, BMI, sex, pathological diagnosis, pre-operative MRI, post-operative sonography, surgery times, follow up date, pre-operative lesion max diameter and post-operative ultra-SONO findings were collected to a chart. We also calculated each follows up days. At outpatient clinic, we measured the change of lesion size by D-USG which was simple and cost-effective.

2.1 Anatomical location

The lesion locations divided into 14 parts according to reference. (10)

2.2 Involve layer

Involve layers divided into 11 parts according to magnetic resonance imaging findings and refer to other researches.

2.3 Venous malformation grading

The VMs into four grades based on the size and margin in preoperative MRI, Goyal grade: grade 1, well-defined ≤ 5 cm; grade 2A, well-defined >5 cm; grade 2B, ill-defined ≤ 5 cm; and grade 3, ill-defined >5 cm. (11)

2.4 Surgical details, hospitalization, complications

Surgical details collected from the electronic medical record, complications contain recurrence, infection, dehiscence, anal fistula, hematoma, scar widening. The surgical procedure was performed in one or several stages by one plastic surgeon. The goal was to remove as much of the lesion as possible to change lesion contour.

3. Evaluation

3.1 Part 1 : Comparison of results from preoperative and postoperative medical pictures.

Volume-volume measurement: Review of Electronic Medical Record and Magnetic Resonance imaging. Total 102 patients' pre-operative MRI max diameter, preoperative MRI volume, most recent post-operative ultra SONO max diameter, post-operative ultra SONO volume was measured by the following formula. And the EMR MRI, SONO findings were also collected. patients have MRI findings X, Y, Z size in EMR, they measured MRI-SONO volume by $V=X*Y*Z$;

The patient who don' t have MRI findings, X, Y, Z size in EMR, we made several stacks, calculated the approximate volume. $V^3 = \frac{1}{3}[s1+s2+\sqrt{(s1*s2)}] h1 + \frac{1}{3}[s2+s3+\sqrt{(s2*s3)}] h2$. (if 3 stacks). Patients have preoperative MRI findings and postoperative ultrasonography findings were 46 totally. (46/102)

3.2 Part 2 : Panel evaluation by comparing the preoperative postoperative photographs.

A panel evaluation (3): peer-review evaluation contains resection score, contour score, and scar score. Three or four medical color photos form operative evaluation slides. They were shown to three board-certified plastic surgeons who did not participate in the surgery in this study. Each slide was shown to experts for 10 seconds. (Pre-op. slide with 3 photographs 10s, intra-op. slide with 3 photographs 10s, post-op. slide with 3 photographs 10s.) Peer review evaluation total has done in 126 surgical procedures, 80 procedures with pre-operative, intra-operative, post-operative 3 slides, 46 procedures with pre-operative, intra-operative 2 slides. Totally evaluated 1134 pic. of patients. The evaluation contains 5 point-scale resection rate: 1. Poor, 2. Fair, 3. Average, 4. Good, 5. Excellent. 4 point-scale contour rate: 1. Poor, 2. Good, 3. Very good, 4. Excellent. 4 point-scale scar rate: 1. Poor, 2. Good, 3. Very good, 4. Excellent. (12)

Table 1. Resection, contour, scar evaluation grading details and descriptions.

Resection	Description
Grade / definition	Description
1. Poor	No change in size before and after surgery.
2. Fair	Minor improvement. The lesion is clearly identifiable.
3. Average	The size has reduced to a certain extent. But still remain some malformation.
4. Good	Major improvement. The size has decreased significantly, with a little malformation.
5. Excellent	Apparently cured, malformation essentially gone, barely discernible.
Contour	
Grade/ definition	Description
1. Poor	No change. Compared with before the operation, the contour has not changed.
2. moderate	Minor improvement. Pronounced protuberance or depressed, but the contour is better than before surgery.
3. Good	Major improvement, but slightly uneven contour compared with the surroundings.
4. Excellent	Nearly matched contour with surroundings.
Scar	
Grade/ definition	Description
1. Poor	Pathologic scar such as keloid, hypertrophic scar and severe scar widening with severe stitch mark.
2. moderate	An obvious scar, moderate hypertrophic scar and scar widening with stitch mark.
3. Good	Slightly visible scar, mild scar widening.
4. Excellent	Hardly noticeable, rarely visible.

4. Statistical analysis

Results were expressed as means \pm standard deviation among the patients' characteristics. Statistical analysis of evaluation results performed by One-way ANOVA. The difference was statistically significant at P-value < 0.05. Peer review evaluation measured by Cronbach' s alpha test.

5. Results

5.1 Patient characteristic

A total of 102 patients who were diagnosed with venous malformation at our hospital from 2009 to 2019 in this study. Among them, the lesion sites were head and neck in 48 patients (46.2%), hand and foot with 28 patients (26.9%), extremity 21 (20.2 %), and trunk and perineum 7 (7.3%). Specifically, the lip was the most common lesion site 20 (41.7%) and cheek 10 (20.8%) following in the head and neck part. The average age of surgery was 10 years old, and 46 patients were male, 56 patients were female. The average weight, height was 18.98kg and 125.5cm. The average follow-up period was 2.18 years. The clinic characteristics of all the registered venous malformation (VM) patients who underwent surgery

Table 2. Summary of Demographic and Clinical Characteristics

Characteristics	VM Patients
Age (first surgery), median	10 years, 8 years
Male	46 (102)
Female	56 (102)
BMI	18.90kg/m ²
Anatomical Location	
Scalp	4
Face	40
Neck	4
Thorax	2
Back/flank	4

Arm/elbow	4
Forearm/wrist	4
Hand	20
Pelvis/hip	1
Thigh/knee	4
Leg/ankle	9
Foot	8
Involve layer	
Skin	1
Subcutaneous	36
Muscle	1
Skin, subcutaneous	25
Subcutaneous, muscle	12
Subcutaneous, muscle, bone	1
Subcutaneous, muscle, mucosal	3
Skin, mucosal, muscle	1
Skin subcutaneous, muscle	11
Skin, subcutaneous, bone	1
Skin, mucosal, subcutaneous, muscle	12
Total lesions	104 (102+2)
Goyal grading	
Preoperative venous malformation	
MRI findings	
grade 1, well-defined ≤ 5 cm	59 (57%)
grade 2A, well-defined > 5 cm	4 (4%)
grade 2B, ill-defined ≤ 5 cm	29 (29.28%)
grade 3, ill-defined > 5 cm	12 (11%)
Treatment	
Total Surgery	126
W-plasty	36
Z-plasty	6
Radical excision	88
Debulking excision	38

5.1.1 Anatomical location

The location of the lesions was evaluated through physical examination and imaging tests in patients in whom a VM was suspected. Table 1 shows the anatomical location divided into 12 parts according to other research. The most common patients were in head and neck area about 48.

The VMs in all 102 patients with 104 lesions were classified based on the depth and involved anatomic layers, as follows: the skin, subcutaneous tissue, the muscle layer, and the mucosa layer and so on. The skin layer was invaded in 49% of patients. The most common region of invasion was the skin (including subcutaneous tissue) along with the muscle and mucosa in 101 lesions (97.1%), same as the skin muscle layer only in 41 lesions (39.4%), the skin (including subcutaneous tissue) and the muscle layer in 11 lesions (11.5%), and the skin and subcutaneous layers only in 25 patients (24%). Approximately 90 % of VMs have superficial parts that involve the skin. (13)

5.1.2 Venous malformation gradings

The unifocal type in 102 patients was 98.1% according to MRI findings, except 2 patients with multifocal type. The lesion size was smaller than 5 cm in 88 patients (84.6%) and more than 5 cm in 16 patients (15.4%). The margin was well-defined in 63 patients (60.1%) and ill-defined in 31 patients (39.4%). (Figure 1) Grade 1, well-defined ≤ 5 cm, 59 (57%) was the most common type.

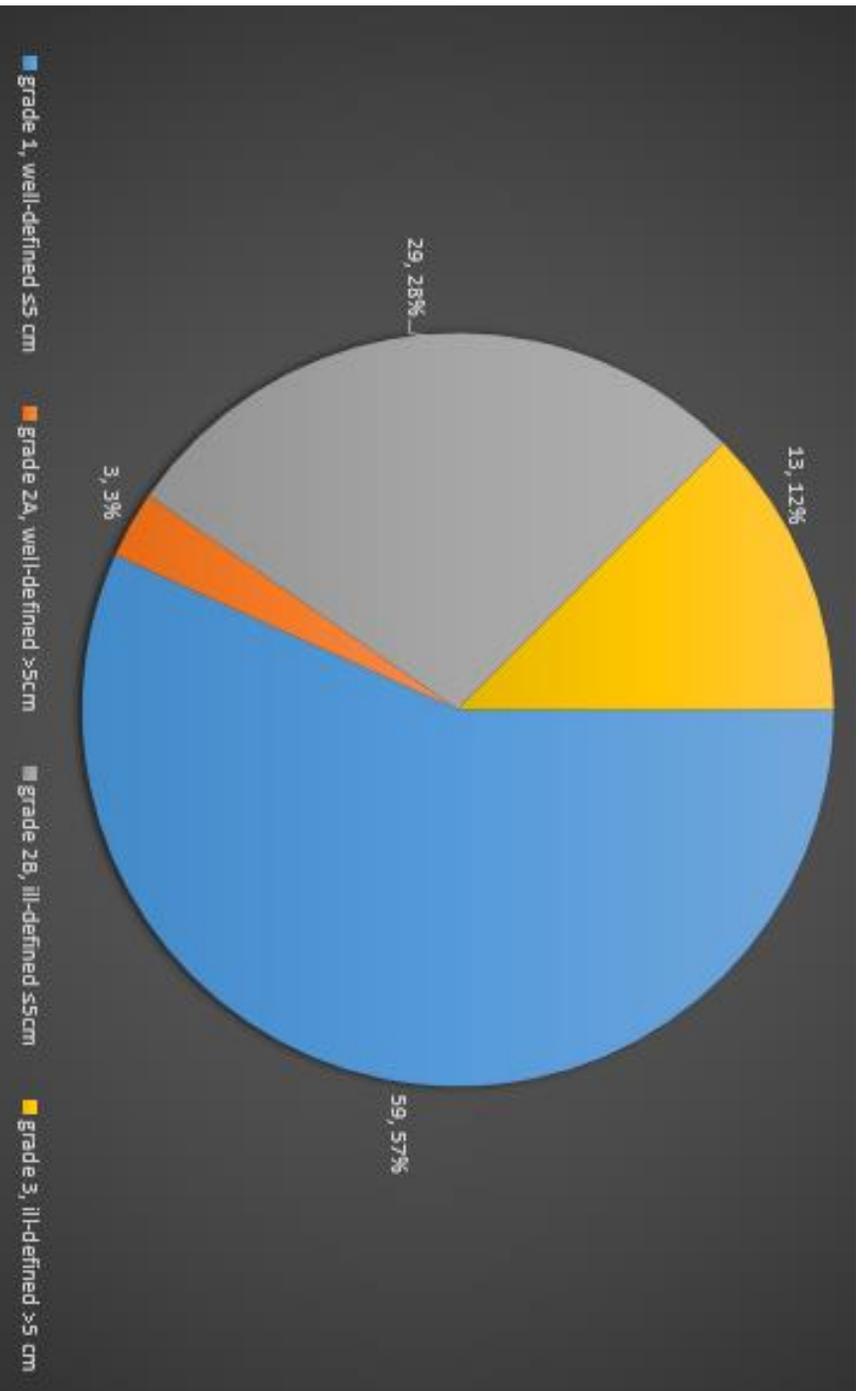


Figure 1. Venous malformation grading results.

5.1.3 Hospitalization, Surgical details, follow up days

The details of the surgeries are shown below in Table 3.

Table 3. The details of the surgeries (N =126)

Details	Number
Number of surgeries	
1 procedure	102
2 procedures	18
3 procedures	5
4 procedures	1
Total	126
Type of surgery	
Radical excision	88
Debulking	38
Hospitalization (mean duration of days)	
1st surgery	4.25
2nd surgeries	5.25
3rd surgeries	5.67
4th surgeries	5
Hospitalization Range (days)	
Day surgery	9
1-3	47
4-6	55
7-9	16
Over 10	3
Follow up days (average)	796.7days
Total	126

5.1.4 Postoperative complications

All potential complications were recorded in Table 5 as shown below.

Table 4. The complications of the surgeries (N =126)

Complications	Number	Percentage of patients
Reccurence	4	20
Delay healing (dehiscence)	8	40
Anal fistula	1	10
Hematoma	3	15
Infection	1	10
Blood transfusion	2	10
Total	19	

5.2 Evaluation results

Part 1. In 46 patients, we measured the preoperative and postoperative. lesion size with magnetic resonance imaging and sonography. Pre-op MRI post-op SONO lesion volume was measured. The average reduction rate was 87.59% (ST=26.52).

Part 2. Peer review was done in 96 patients, 126 operations preoperative and postoperative medical photos. Each slide displayed 3 to 4 medical pictures and reviewers compared pre-op, intra-op, and post-op slides of each patient. The results are shown in table 5.

Result of resection	Excellent	Good	Average	Fair	Poor	Cronbach a***
Preoperative-long term post operative *	44 (46.81%)	36 (38.30%)	9 (9.57%)	5 (5.32%)	0 (0%)	0.748
Preoperative-immediate post operative **	17 (53.13%)	12 (37.50%)	3 (9.38%)	0 (0%)	0 (0%)	0.672
Result of contour	Excellent	Good	Moderate	Poor	Cronbach a	
Preoperative-long term postoperative *	52 (55.32%)	36 (38.30%)	6 (6.38%)	0 (0%)	0.625	
Preoperative-immediate post operative **	15 (46.88%)	15 (46.88%)	2 (6.25%)	0 (0%)	0.673	
Result of scar	Excellent	Good	Moderate	Poor	Cronbach a	
Preoperative-long term postoperative *	45(47.87%)	31(32.98%)	15(13.96%)	3(3.19%)	0.851	

* Preoperative-long term postoperative: 94 operations in 126 operations preoperative intraoperative postoperative medical photo were evaluated.

** Preoperative-immediate post operative: 32 operations in 126 operations preoperative-immediate postoperative medical photo were evaluated.

*** Cronbach's alpha is a function of the number of items in a test, the average covariance between item-pairs, and the variance of the total score.

Table 5. The evaluation results of peer review.

5.3 Statistics Analysis of evaluation results

Contour and scar scores showed a significant relationship between the gradings.

		ANOVA				
		Sum of Squares	df	Mean Square	F	Sig.
resection	Between Groups	5.061	3	1.687	2.488	.064
	Within Groups	82.732	122	.678		
	Total	87.793	125			
contour	Between Groups	4.326	3	1.442	4.419	.005
	Within Groups	39.809	122	.326		
	Total	44.135	125			
scar	Between Groups	9.966	3	3.322	5.390	.002
	Within Groups	55.468	90	.616		
	Total	65.434	93			

Table 6. Statistics analysis results.

6. Cases reports

Representative cases of grade 1, 2, and 3 can be found below.

6.1 A case of VM with surgical treatment

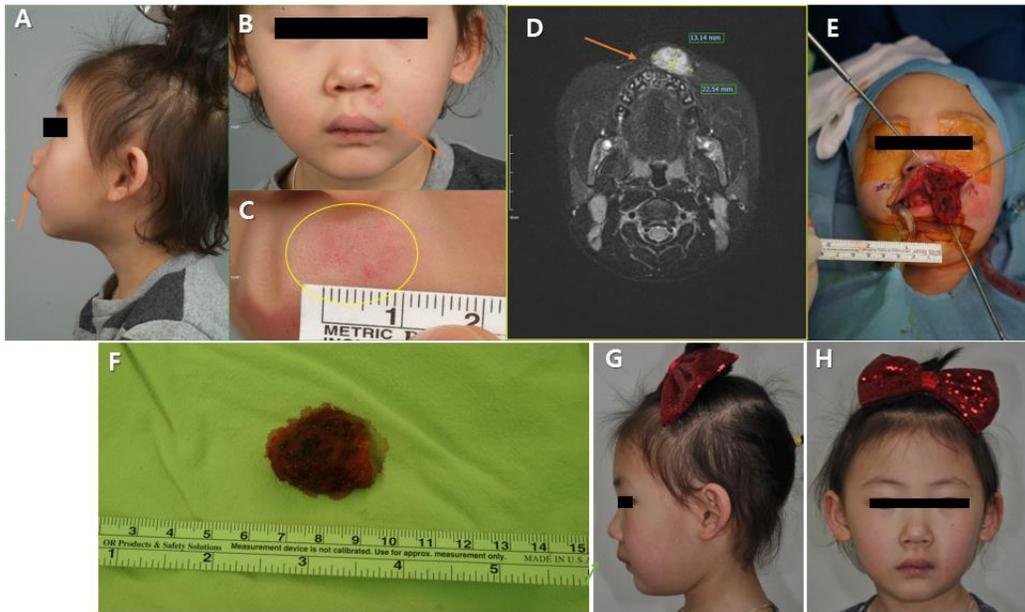


Figure 2. Grade 1 patient. (A), (B), (C) A patient with grade 1, upper lip, Pre-operative photograph. (D) Axial view, Hyper-intense on the MRI image. (E) Intra-operative photograph, surgery was performed with excision line. (F) Venous malformation isolated. (G), (H) Post-operative medical photograph, out clinic. Excision line was normal appearance.

6.2 A case of VM with surgical treatment



Figure 3. Grade 2a patient. (A) and (B) A patient with grade 2B, left upper lip, Preoperative photograph. (C) Preoperative MRI, sagittal view About 5.9 mm-sized ill-defined lesion at the upper lip subcutaneous, muscle area. Preoperative T2-weighted sequence MRI, sagittal view of lesion side. High signaled mass. Demonstrates typical enhancement. (D), (E) Intra-operative photograph, subcutaneous dissection and removal of mass. (F), (G) Post-operative medical photograph took from out clinic in POD 39, POD 438.

6.3. A case of VM with surgical treatment

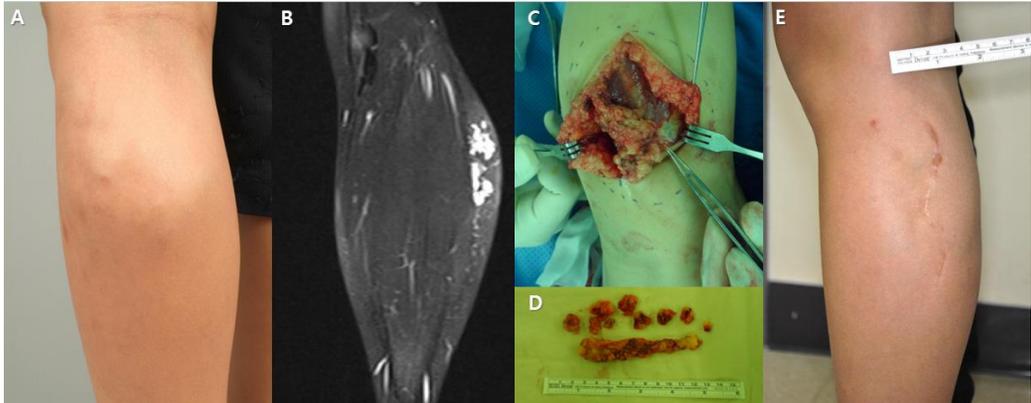


Figure 4. Grade 3 patient. A patient with grade 3, left lower leg, Preoperative photograph. (B) Preoperative MRI, coronal view About 8 cm-sized ill-defined lesion at the left lower leg subcutaneous area. Preoperative T2-weighted sequence MRI, transverse view of lesion side. High signaled mass. Demonstrates typical enhancement. (C) Intraoperative photograph, surgery was performed with w-shape excision line. (D) Excised venous malformation. (E) Post-operative medical photograph took from out clinic in POD 1558.

7. Discussion

VMs expands over a lifetime with pain and swelling being the most common symptoms, especially in adolescence caused by circulating hormones. (14) So aggressive treatment can be useful in pediatric VM patients, it can minimize the rate of lesion growth, also reduce pain.

Treatment of slow-flow VMs with sclerotherapy technically has a success rate of close to 100 % (15) But, the efficacy of treatment for VMs is difficult to evaluate and compare, as the standard assessment is different among the studies. (16)

During sclerotherapy and surgical treatment results evaluation, self-questionnaire, satisfaction, panel assay was used. Patients who undergo sclerotherapy, satisfaction based on self-assessment seems to be a useful tool, since all studies show comparable satisfaction is up to 80 % of cases, as Nakamura et al. (17) (18) (19) (20)

Also in some surgical treatment journals, satisfaction rate calculated to mean 4.26/5. (21) ; 4.1/5 (22) which means that surgical treatment results indicate a more than 80% satisfaction rate in these journals.

Questionnaires are a well-known method to measure the patients' subjective outcome and satisfaction after sclerotherapy. (23) Berenguer et al. reported an overall degree of satisfaction with the treatment of craniofacial venous malformations in children and adults in 78 % of patients (4)

Some studies evaluated the surgical results with objective quantification (MRI data). Roh et al. (24) Criteria for the evaluation of surgical treatment, divided into three evaluation parts. Remission, Improvement, No change. 75% of patients lead to remission or improvement, however, 25% of the patients did not show definite change after surgery.

Therefore, in our study, summing up the previous experience, we construct an objective evaluation model. One is the medical record change evaluation, the other one is panel assay which divided into 3 parts: Reduction, Contour, Scar rate.

Complication rates in children treated with sclerotherapy are common. (25) Sclerotherapy for slow flow malformation has complications included

blistering, swelling, erythema, ulceration, cellulitis, alopecia, hyperpigmentation, scarring, and headache. (26) (27) (28) (29) (30) (31) (32) (33) (34) Major complications are reported in 10–30 % (35)

Our surgical treatment results showed, The incidence rate of complications was 15.08% (19/126), including recurrence, infection, dehiscence, anal fistula, hematoma, revision surgery, and blood transfusion

From what has been discussed above we used the panel assay method to evaluate surgical outcomes and evaluate medical records as an objective assessment. Our cohort showed based on data with a resection 3.96 / 5.00, contour 3.18 / 4.00, and scar 2.98 / 4.00, respectively. These results indicate more than 80% of surgical results in remission or improvement. Also, medical photograph measurement results show 87. 59% reduction rate. 41 patients out of 46, the reduction rate over than 50%. This result is better than previous study. (24)

The significance of this study is as follows : Objective evaluation criteria were used to measure the outcome of surgery, surgical treatment was the first–line.

8. Limitations:

In our study, the data collected from the EMR retrospective case series with surgical treatment protocols. For this reason, the main limitation is that data couldn' t cover all 102 patients pre–op MRI post–op SONO lesion max diameter, volume. Some patients did not have SONO measurements at the outpatient clinic. Only 46 patients' follow up volume was measured by SONO.

After that, we only had SONO follow–up data, not MRI. One reason is that it is easier and more efficient to follow up patients in outpatient clinics. MRI can found lesions more accurately, but it is more difficult to detect in the outpatient clinic than SONO. Also, MRI is a financial burden for patients.

9. Conclusion:

In conclusion, we have presented the evaluation results of 102 patients who have undergone surgical treatment as a first–line treatment for VMs. Here, the medical record showed a low complication rate.

Furthermore, based on these results, the resection rate was 87.59% in 46 patients and the resection score was 3.95 / 5.00 of 126 procedures. Therefore, surgical treatment was effective enough as a first-line treatment of VMs.

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국문초록

정맥 기형에 대한 1차 치료로서의 외과적 절제

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정맥 기형 (VMs)은 혈관 형태 발생학(morphologic) 오류로 인한 질병입니다. 선천적인 혈관 이상(vascular anomalies)이며, 특히 두경부에서 많이 발생하는 질병입니다. VMs은 흔히 푸른색을 띠고, soft 하고 외부의 압력으로 압축 가능합니다. 병변은 얇은 벽, 확장된 스폰지 형, 채널 형 등으로 구성되어 있습니다. 일반적으로 경화 요법은 정맥기형 환자의 첫 번째 우선 치료방법(first-line)입니다. (2) 저희 연구에서는, 일차 치료법으로 외과적 치료를 선택하였고 그 결과를 평가하였습니다.

이 후향적 연구는 2009 년부터 2019 년까지 서울 대학교 어린이 병원에서 VM 진단을 받고 수술적 치료를 받은 환자의 데이터를 포함하였습니다. 이 연구는 주로 네 부분으로 구성됩니다. 첫번째로 VM grading은 선행연구 참고로 기록하였고 두번째로 절제율(resection rate) 측정 및 평가 세번째로는 전문가 평가 그리고 수술전후 의학기록 변화이며 마지막으로 결과의 통계적 분석입니다.

수술 결과는 두 가지 방법으로 평가되었습니다. 하나는 수술 후 병변의

변화된 절제율 평가이고, 다른 하나는 전문가 평가로 절제 점수, 윤곽 점수 및 흉터 점수를 세가지 사항을 포함합니다 (표 1). 결과는 3 명의 성형 외과 전문의 의사에 의해 평가하였습니다. (3) 일원 분산 분석을 결과 통계 분석으로 사용했습니다 (IBM SPSS Statistics 25).

전체 102 명의 환자와 126 개의 수술 절차가 평가되었습니다. 평균 추적 관찰 시간은 2.2 년 (범위: 0-7.3 년)입니다. 그 중 2 명의 환자가 2 개의 다중 병변을 가졌으며 때문에 VM은 모두 104 개의 병변 부위를 기록하였습니다. 46 명의 환자에서 수술 전 MRI와 수술 후 초음파 검사 사이의 병변 크기 (볼륨 및 최대 직경)를 비교했습니다. 평균 절제율은 87.59% 였으며 그 중 32 명의 환자가 완전히 치료되었습니다. 전문가 평가 결과는 표5에 나와 있습니다. 전문가 평가에 의한 평균 절제 점수, 윤곽 점수 및 흉터 점수는 각각 3.96/5.00, 3.18/4.00 및 2.98/4.00입니다. 통계 분석 결과에 따르면, 임상 분류 등급이 높을수록 수술 후 점수가 낮습니다 (낮은 점수는 불량한 결과를 나타냄). 합병증의 발생률은 재발, 감염, 부종, 항문 fistula, 혈종, 수정 수술 및 수혈을 포함하여 15.08 % (19/126)였습니다. (4)

우리는 VM의 1차 치료법으로 수술 치료를 받은 102 명의 환자에 대한 평가 결과를 평가했습니다. 절제비율은 46 명의 환자에서 87.59 % 였고, 절제술 점수는 126개의 절차 중 3.95/5.00이었다. 때문에 수술 치료가 다른 치료방법과 비교하였을 때에 VM의 1 차 치료로 효과적이라는 것을 보여줍니다.

주요어: 정맥기형/평가/수술적 치료/후향적 연구/소아 환자

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