Intraoperative Assessment of Facial Nerve Trunk Width in Early Childhood With Cervicofacial Lymphatic Malformation

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Background: Facial nerve damage during head and neck surgery has long been an important issue. However, few publications on the gross anatomy of the facial nerve are available in the young population. The aim of this study was to provide in vivo measurements of the facial nerve trunk during lymphatic malformation (LM) resection and to determine the association between the trunk width and patient- and disease-related variables.

Methods: We conducted a retrospective analysis of 11 consecutive pediatric patients (11 facial nerve trunks) who underwent cervicofacial LM resection. The facial nerve of the affected side was dissected, and its trunk width at bifurcation was measured using calipers under a microscope during the operation.

Results: Eleven patients younger than 6 years were enrolled. The median width of the facial nerve in patients younger than 1 year was 1.15 mm; it was 2.5 mm in those older than 1 year. Trunk width was significantly greater in patients older than 1 year than those younger than 1 year, whereas no statistical significance was found when comparing other age groups. Patient weight was positively correlated with trunk width, whereas LM grade and diameter showed no significant correlation.

Conclusions: The significantly greater width of the facial nerve trunk in LM patients older than 1 year than those younger than 1 year suggests that the age of 1 may be a threshold for facial nerve hypertrophy and growth acceleration. This study provides informative in vivo data to help understand facial nerve characteristics in young patients.

Key Words: pediatric facial nerve, in vivo nerve width, nerve hypertrophy, cervicofacial lymphatic malformation

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Facial nerve dissection is associated with a high rate of complications.[1,2] Facial paresis or paralysis has long been a serious lifelong complication for patients who undergo cervicofacial dissection procedures, such as resection of lymphatic malformation (LM).[3] Especially in children, such a permanent complication is devastating. Understanding the nature of the facial nerve is important in avoiding facial nerve injury, so there have been a large number of efforts to determine the characteristic anatomy and function of the facial nerve.[1] However, few publications examining the gross anatomy of the facial nerve in vivo are available, and data are still lacking, especially in the young population. Publications so far have been limited to indirect studies using computed tomographic[4] imaging, magnetic resonance imaging,[5] ultrasonography,[6] or cadavers.[7–9] There is still doubt as to how such indirect findings are correlated with actual intraoperative findings.

In our clinical experience of facial nerve dissection during head and neck surgery in pediatric patients, facial nerve width was observed to be much larger than expected based on previous findings in adult patients. We therefore performed direct measurements of the facial nerve trunk during LM resection in young patients to subject our clinical observations to statistical analysis. The final aim of this intraoperative observational study was to provide in vivo data on the pediatric facial nerve and to determine the association between nerve width and patient- and disease-related variables.

METHODS

The medical records of all consecutive patients with cervicofacial LM who presented to the pediatric plastic surgery clinic, Samsung Medical Center, Seoul, Republic of Korea, from 2011 to 2016, were retrospectively reviewed. Inclusion criteria were a diagnosis of cervicofacial LM, younger than 6 years, and total or subtotal resection of LM with facial nerve dissection. Exclusion criteria included cervicofacial LM patients who did not require facial nerve dissection.

Data on patient demographics, preoperative LM staging, preoperative LM diameter, and intraoperative measurements of facial nerve trunk width were collected. LM staging was scored according to the criteria defined by de Serres et al.[10] Stage I patients have unilateral infrahyoid disease; stage II, unilateral suprahyoid disease; stage III, unilateral infrahyoid and suprahyoid disease; stage IV, bilateral suprahyoid disease; and stage V, bilateral infrahyoid and suprahyoid disease. The largest transverse diameter of LM was preoperatively measured in every patient using magnetic resonance imaging or computed tomography. Facial nerve trunk width in this study was defined as the width of the main nerve bundle at the first bifurcation, immediately before splitting off into the temporofacial and cervicofacial branches (Fig. 1). Near-total resections including areas surrounding major neck vessels and extensive LM lesions were performed by a pediatric general surgeon (J.M.S.). Meticulous dissection with full exposure of the facial nerve trunk including the marginal mandibular branch was done by a plastic surgeon (S.Y.L.) under microscopic inspection. For each patient, the facial nerve trunk was observed and trunk width at bifurcation was measured using calipers under a microscope (Fig. 2). The values were recorded in millimeters.

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This work is in accordance with the World Medical Association Declaration of Helsinki as a statement of ethical principles for medical research involving human subjects, including research on identifiable human material and data.

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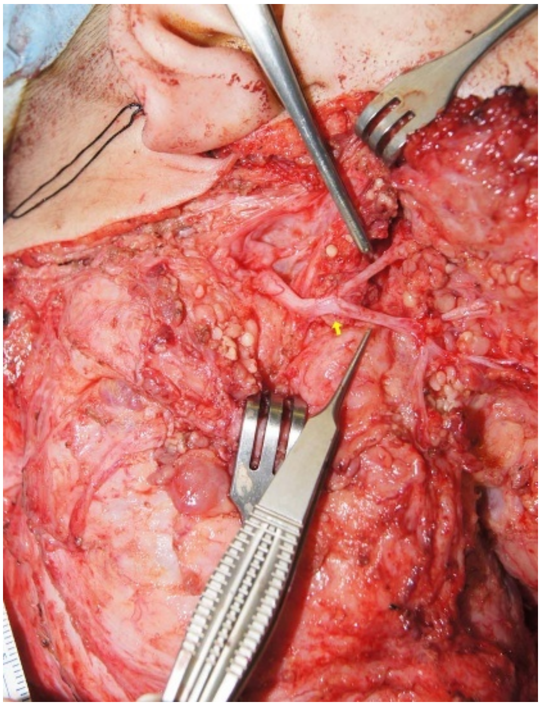

FIGURE 1. A yellow arrow indicates the main bundle of facial nerve at first bifurcation which is giving off temporofacial and cervicofacial branches (the patient's age at dissection was 2 years and 8 months old).

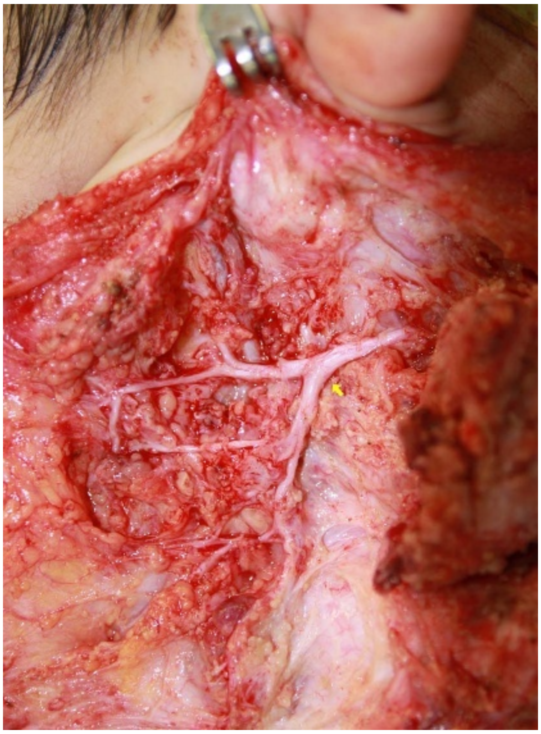

FIGURE 2. A yellow arrow indicates the facial nerve trunk which was 3.8 mm in width under surgical microscope (the patient age at dissection was 5 years and 11 months old).

We divided our patients into 2 groups by age, older and younger than 1 year, and compared the width of the facial nerve trunk between the 2 groups using the Wilcoxon rank-sum test. In the same manner, we compared the width in other age groups (2, 3, 4, and 5 years as cutoff values). To determine the relationship between facial nerve trunk width and patient weight, diameter of LM, and grade of LM, we used Spearman correlation coefficients. All statistical analyses were performed with SAS version 9.4 (SAS Institute, Cary, NC).

RESULTS

Of the total number of cervicofacial LM patients seen during the study period, 11 patients who met our criteria, accounting for eleven facial nerve dissections, were enrolled in this study. Patient demographics, preoperative LM staging, preoperative LM diameter, and intraoperative measurement of facial nerve width are shown in Table 1. All enrolled patients were younger than 6 years. Four patients were younger than 1 year and 7 patients were older than 1 year at the time of dissection. The median width of the facial nerve trunk in all patients was 2.1 mm. When divided by age, the median value was 1.15 mm for patients younger than 1 year and 2.5 mm for patients older than 1 year. The median width was 1.55 mm in patients younger than 2 years and 2.55 mm in patients older than 2 years; 1.9 mm in patients younger than 3 years and 2.5 mm in patients older than 3 years; 1.95 mm in patients younger than 4 years and 2.8 mm in patients older than 4 years; and 2 mm in patients younger than 5 years and 3.3 mm in patients older than 5 years. Facial nerve trunk width was significantly greater in patients older than 1 year than in those younger than 1 year (mean score, 8 vs 2.5; $P = 0.0107$ by Wilcoxon rank-sum test). No statistical difference in facial nerve trunk width was found when other ages were used for the cutoff (mean score, 4.2 vs 8.2; $P = 0.0552$; mean score, 4.7 vs 8.3; $P = 0.1082$; mean score, 4.9 vs 9; $P = 0.0827$; mean score, 5.1 vs 10; $P = 0.0771$ for 2, 3, 4, and 5 years old, respectively, by Wilcoxon rank-sum test) (Table 2). Patient age was positively correlated with facial nerve trunk width (Spearman $r = 0.8793$, $P < 0.01$) (Fig. 3) and patient weight, ranging from 3.7 to 21.9 kg, showed a positive correlation with facial nerve trunk width as well (Spearman $r = 0.8727$, $P < 0.01$) (Fig. 4). LM grade was distributed as follows: grade I,

TABLE 1. Patient and Disease-Related Characteristics

Variables	Value
No. patients	11
No. FN trunk	11
Age, mean ± SD, mo	30.07 ± 26.36 (range, 0.76–71)
Weight, mean ± SD, kg	12 ± 6.26 (range, 3.7–21.9)
Sex, n (%)	
Male	7 (64%)
Female	4 (36%)
LM grade, n (%)	
Grade I	1 (9%)
Grade II	1 (9%)
Grade III	9 (82%)
Grade IV	0
Grade V	0
LM diameter, mean ± SD, mm	74.44 ± 29.62 (range, 35 to 137.98)
LM laterality, n (%)	
Right	7 (64%)
Left	4 (36%)
Bilateral	0

FN, facial nerve.

TABLE 2. In Vivo Width of Facial Nerve Trunk in Pediatric Patients With Cervicofacial Lymphatic Malformation

		FN Width, mm				
Group	**N (%)**	**Minimum**	**Maximum**	**Median**	**Mean score ± SD**	***P***
Age < 1 y	4 (36%)	1	1.9	1.15	2.5 ± 5.3	0.0107*
Age > 1 y	7 (64%)	2	3.8	2.5	8 ± 5.3	
Age < 2 y	6 (55%)	1	3	1.55	4.2 ± 5.5	0.0552
Age > 2 y	5 (45%)	2.1	3.8	2.5	8.2 ± 5.5	
Age < 3 y	7 (64%)	1	3	1.9	4.7 ± 5.3	0.1082
Age > 3 y	4 (36%)	2.1	3.8	2.5	8.3 ± 5.3	
Age < 4 y	8 (73%)	1	3	1.95	4.8 ± 4.9	0.0827
Age > 4 y	3 (27%)	2.2	3.8	2.8	9 ± 4.9	
Age < 5 y	9 (82%)	1	3	2	5.1 ± 4.2	0.0771
Age > 5 y	2 (18%)	2.8	3.8	3.3	10 ± 4.2	

*P value < 0.05, Wilcoxon-rank sum test.

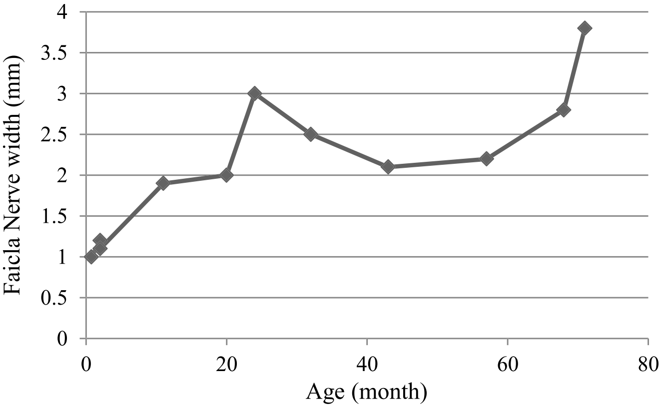

FIGURE 3. Facial nerve width on the *y*-axis with patient age by month on the *x*-axis in pediatric lymphatic malformation patients.

1/11 (9%); grade II, 1/11 (9%); grade III, 9/11 (82%); and no grade IV or V. On preoperative magnetic resonance or computed tomography imaging, the largest transverse diameter of LM for each patient was found to range from 35 to 137.98 mm. LM grade and LM diameter were not significantly correlated with facial nerve trunk width (Spearman $r = -0.0809$, $P = 0.8131$ and $r = -0.47381$, $P = 0.141$, respectively) (Table 3).

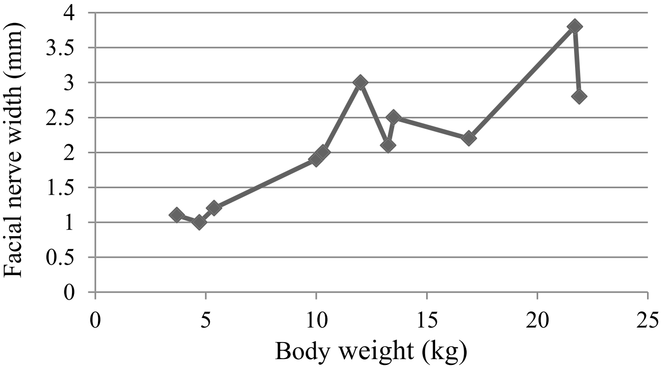

FIGURE 4. Facial nerve width on the *y*-axis with body weight on the *x*-axis in pediatric lymphatic malformation patients.

DISCUSSION

Interestingly, our observations indicated that a patient aged 1 year was the threshold for facial nerve hypertrophy. When we divided our LM patients into 2 groups, younger and older than 1 year, facial nerve trunk width became larger at about the age of 1 year and showed no statistical significance thereafter. We suggest that the age of 1 year, which is the end of infancy, is a threshold for facial nerve enlargement, whereas the nerve is known to develop until approximately 4 years after birth in previous study.[11]

The presence of a significant difference in facial nerve width at a certain age threshold suggests that LM may affect surrounding tissue differently after that age. Another hypothesis is that facial nerve growth and development are programmed from birth to accelerate during the first year of life, regardless of the presence of LM. Because our study was confined to patients with cervicofacial LM, we could not compare the subjects with healthy pediatric controls or other pediatric patients without LM but with other disease entities. Further intraoperative studies could be conducted to determine whether or not a similar pattern of nerve hypertrophy and growth is observed in those groups.

Our pediatric LM patients older than 1 year, who had a median facial nerve width of 2.5 mm, had larger facial nerve trunks than those previously observed in sonographic studies in healthy adults. We propose 2 possible explanations. (1) LM adjacent to the facial nerve may act as a potent inducer of nerve hypertrophy. LM has been reported to be associated with genetic mutations causing adjacent tissue hypertrophy,[12] especially with a *pik3ca* gene mutation. PIK3CA-related overgrowth[13] induces anomalies of the nervous system and segmental body overgrowth, such as lymphatic, vascular, skeletal, or combined hypertrophy. An anecdotal report[14] suggests that nerves in vascular

TABLE 3. Correlations of Patient Weight, LM Grade, and Diameter with In Vivo Facial Nerve Trunk Width in Pediatric Patients With Lymphatic Malformations

	Weight, kg		**LM Grade**		**LM Diameter**	
FN width, mm	*r*	*P*	*r*	*P*	*r*	*P*
	0.87	0.0005*	−0.081	0.81	−0.47	0.14

*P-value < 0.01.

malformations have close relationships during growth, because nerves and blood vessels use common molecular pathways to differentiate and proliferate. As LM is defined as a cluster of abnormal vessels filled with clear lymphatic fluid, abnormal LM growth may lead to adjacent nerve hypertrophy via cross-talk between the vessels and nervous system. Our intraoperative findings are consistent with the previous publications showing that LM may cause nerve hypertrophy. (2) Sonographic data may not be a good substitute for intraoperative data, so we cannot conclude that nerve width was definitely greater in our pediatric patients than in adults. There have been no intraoperative studies done in a healthy control group for comparison, because healthy children do not undergo facial nerve dissection.

Facial nerve trunk width was observed to increase in size with an increase in patient weight. In contrast to chronological age, patient weight would be a difficult criterion to use for establishing a threshold for hypertrophy because of its highly variable nature. Patient weight may change significantly, especially in young children, depending on socioeconomic or nutritional status.[15,16] Though our LM patients showed a positive correlation between weight and facial nerve width, the data are not sufficient to determine how body growth affects nerve growth.

The stage and diameter of LM showed no significant correlation with facial nerve trunk width. LM grade and diameter may be subject to measurement error, which would make it difficult to define a clear correlation with facial nerve growth in this study. The de Serres classification used for this study is a limited staging system which subjectively categorizes LM according to laterality and location. For more comprehensive and objective determination of LM severity, Cologne Disease Score values may be added in future investigations. Preoperative LM diameter was measured by several different physicians from the department of radiology during the long study period, which may have created some interobserver differences in measurement.

Because this study was conducted during a nerve-preserving procedure, we measured nerve width with calipers as an alternative to nerve diameter, whereas it is more reliable to measure the diameter of a nerve under histological examination of a resected nerve specimen.[17] For maximal accuracy, our measurements were performed under a surgical microscope by 2 senior surgeons.

There are many segments of the facial nerve. Its main trunk at bifurcation was measured to minimize interobserver error. The facial nerve has the most complex and variable structure of all the cranial nerves[18] and is often mingled and distorted by enlarged LM. Intraoperatively, the nerve trunk seemed to be most consistent at the point immediately before its initial bifurcation into the temporofacial and cervicofacial branches.

This study provides intraoperative data on the pediatric facial nerve from 11 consecutive cases. Although the presence of LM may be a confounding variable, this study still suggests that growth acceleration of the facial nerve may begin in a very early stage of life and may be affected by hypertrophic conditions, such as LM. Further imaging studies in healthy pediatric subjects could be conducted to investigate the growth of the facial nerve. The local effect of LM on the facial nerve would be an attractive area for further study to lend our findings more accuracy.

CONCLUSIONS

In our intraoperative measurements, the width of the facial nerve trunk was significantly greater in LM patients older than 1 year than in those younger than 1 year, suggesting that the age of 1 year may be a threshold for facial nerve hypertrophy and subsequent growth acceleration. The size and severity of LM showed no significant correlation with facial nerve growth. Although further investigation is needed, this study provides informative in vivo data to help understand facial nerve characteristics in young patients.

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