


Cervical lymphatic malformations amenable to transhairline robotic surgical excision in children

A case series

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Abstract

Lymphatic malformations are rare benign malformations that predominantly occur in the head and neck region. The advent of surgical robots in head and neck surgery may provide beneficial outcomes for pediatric patients. Here, we describe our experiences with transhairline incisions for robot-assisted surgical resection of cervical lymphatic malformations in pediatric patients.

In this prospective longitudinal cohort study, we recruited consecutive patients under 18 years of age who were diagnosed with congenital cervical lymphatic malformations and scheduled for transhairline approach robotic surgery at a single medical center. We documented the docking times, console times, surgical results, complications, and postoperative follow-up outcomes.

The studied patients included 2 with mixed-type lymphatic malformations and 2 with macrocystic-type lymphatic malformations. In all 4 patients, the incision was hidden in the hairline; the incision length was <5 cm in 3 patients but was extended to 6 cm in 1 patient. Elevating the skin flap and securely positioning it with Yang retractor took <1 hour in all cases. The mean docking time was 5.5 minutes, and the mean console time was 1 hour and 46 minutes. All 4 surgeries were completed endoscopically with the robot. The average total drainage volume in the postoperative period was 21.75 mL. No patients required tracheotomy or nasogastric feeding tubes. Neither were adverse surgery-associated neurovascular sequelae observed. All 4 patients were successfully treated for their lymphatic malformations, primarily with robotic surgical excisions.

Cervical lymphatic malformations in pediatric patients could be accessed, properly visualized, and safely resected with transhairline-approach robotic surgery. Transhairline-approach robotic surgery is an innovative method for meeting clinical needs and addressing esthetic concerns.

Abbreviations: EJV = external jugular vein, GAN = greater auricular nerve, LMs = lymphatic malformations, SCM = sternocleidomastoid muscle.

Keywords: cervical, lymphatic malformation, pediatric, robotic surgery, transhairline approach

1. Introduction

Lymphatic malformations (LMs) are rare benign malformations that predominantly occur in the head and neck region and commonly affect children aged >2 years.^[1,2] Over recent decades, treatment modalities, such as surgical resection, intralesional sclerotherapy, and laser ablation, have gained popularity, either as single treatments, combination treatments, or staged treatments.^[1,3–6] Treatment choices should be individualized because

cervical LMs often have ill-defined margins, and inappropriate surgical procedures can result in debilitating complications due to the presence of intricate nervous structures in the neck, the very limited surgical working space, and the proximity of LMs to the airway and major vessels.

The emergence of robot-assisted head and neck surgeries for benign malformations offers the advantages of magnification, scaled hand movements, minimization of hand tremors, and

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“round-the-corner” surgical views, especially in the cases of transoral robotic surgeries performed via natural orifices or through concealed surgical incisions.^[7–11] In this study, we describe our experiences with transhairline incisions for robot-assisted surgical resection of cervical LMs in pediatric patients.

2. Materials and methods

2.1. Patients

For this prospective longitudinal cohort study, we recruited consecutive patients under 18 years of age diagnosed with congenital cervical LMs and scheduled for transhairline approach robotic surgery at our hospital from January 1, 2015 to December 31, 2019. We selectively included patients whose imaging findings depicted single unilateral macrocystic- or mixed-type LMs with nearly demarcated borders. We excluded patients with histories of prior surgery or irradiation of the neck, contraindications to surgery under general anesthesia, LMs located in the facial/parotid/cheek region or the oropharyngeal region, or LMs with de Serres stage IV and V.^[12]

A watchful waiting approach was implemented for patients who were <12 months of age. This decision was made both due to local cultural beliefs and to allow time for a proper adaptation of our protocols for applying Yang retractor via a transhairline incision.

2.2. Preoperative assessments

The selected patients underwent preoperative assessments with at least one imaging modality (i.e., sonography, computed tomography [CT], or magnetic resonance imaging [MRI]). The patients underwent clinicoradiologic staging as proposed by de Serres et al.^[12]

2.3. Operative preparation and instrumentation

The detailed operative procedures were based on a previously published protocol.^[11,13] In brief, the patient was placed under general anesthesia with the head turned to the side contralateral to the LM. A transhairline incision was then made. The surgery was performed with a da Vinci surgical robot of either the Si or Xi

models (Intuitive Surgical; Sunnyvale, CA). A 30° dual-channel endoscope was used. For the Si model robot, the 2 instrument arms were equipped with a 5-mm Maryland forceps on the left and a 5-mm spatula monopolar cautery or a Harmonic curved shear (Ethicon; Bridgewater, NJ) on the right. For the Xi model robot, the instrument arms were equipped with the corresponding 8-mm instruments.

2.4. Operative procedure

A skin incision was made in and along the hairline behind the auricle with a cold scalpel. The incision continued through the subcutaneous fat until reaching the sternocleidomastoid muscle (SCM), and the flap was elevated anteriorly toward the target lesions.^[13–15] The posterior belly of the digastric muscle was located by dissecting the lower border of the submandibular gland. The surgical field was exposed after flap elevation with the aid of a self-retaining Yang retractor^[16] (US patent 9,526,485; Fig. 1A and B). The spinal accessory nerve was recognized and skeletonized at the posterior border of the SCM. Special care was taken to avoid injury to the hair follicles during the incision and subcutaneous dissection.

After a tunnel toward the cervical lesion had been created and secured, the robotic surgical system was introduced, and important structures, such as the greater auricular nerve (GAN), the SCM, and the external jugular vein (EJV) were identified and protected to avoid inadvertent injuries. In the case of patient no. 4, an additional 3-cm cervical horizontal incision was created in the lower neck for dissection of a huge cystic mass that impeded simultaneous dissection along the margin and traction through the same transhairline incision. The 3-cm cervical horizontal incision aided counter-traction with the third operating arm. The lesions were carefully dissected by exerting gentle pressure on the retracted skin flap to facilitate surgical field exposure. For the larger lesions of patient no. 4, decompression was performed via fluid aspiration, the needle pricking site was sealed, and the cyst was completely removed. During robotic surgery, extra care was taken to identify and delineate other important structures, such as the great vessels, the vagus nerve, the recurrent laryngeal nerve, and the trachea. After the entire

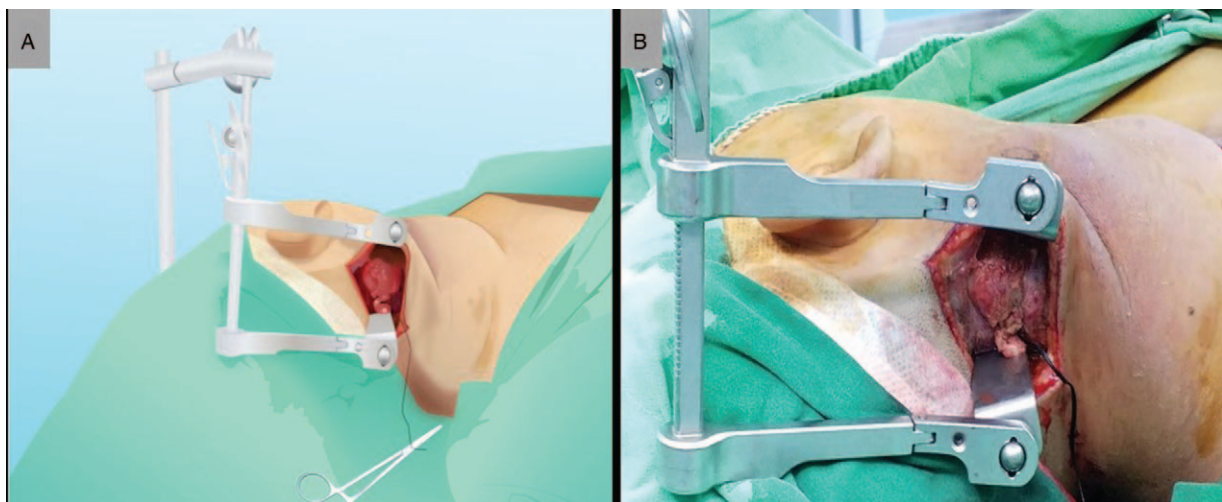


Figure 1. (A) Schematic illustration of the usage of Yang retractor and (B) photograph of Yang retractor being used during surgery in patient no. 4 after aspiration of approximately 750 mL serosanguinous fluid from the cervical LM. LMs=lymphatic malformations.

operation, a routine drainage tube was placed, and surgical wounds were closed layer by layer. If the drainage volume was <10 mL/d, then the drain could be safely removed. The patients were observed for any signs of neural injury, seroma formation, facial palsy, or hematoma before discharge.

In all cases, the surgery was performed by a single surgeon approved to perform robotic surgeries by the hospital's internal credentialing process. The surgeon had to obtain near-perfect scores in robotic simulator sessions and 2 animal laboratory sessions to obtain this approval. Because the robotic surgical system does not provide tactile sensation, the assisting surgeon at the bedside played an important role in alerting the console surgeon whenever the dissection was too close to the stretched skin.

2.5. Postoperative assessments

The patients underwent postoperative assessments with at least one imaging modality (i.e., sonography, CT, or MRI) between 1 month and 1 year after surgery.

2.6. Ethical considerations

This study was approved by the hospital's institutional review board, and informed consent was obtained from the patients, their parents/legal guardians, or both.

3. Results

Five patients underwent transhairline-incision robotic surgery for preoperative diagnosis of macrocystic LMs (i.e., cystic diameters >2 cm) and microcystic LMs (i.e., cystic diameters <2 cm) at our hospital between July 15, 2015 and July 14, 2019. Four patients met the inclusion criteria and were included in the study, and 1 patient with a postoperative diagnosis of immature teratoma was excluded. The included patients were 2 girls and 2 boys with a mean age at operation of 4.4 years (range, 19 months–11 years; Table 1). Three patients had cervical LMs on the left side, and the other had LMs on the right side. The preoperative diagnoses were macrocystic LMs for 2 patients and mixed-type LMs (i.e., macrocystic and microcystic LMs) for the other 2 patients.

The preoperative lateral-view image of patient no. 1 (Fig. 2A) showed the left cervical macrocystic LM. A 3-cm transhairline incision was planned, and docking of Yang retractor ensued after complete dissection of the subcutaneous plane. The cervical lesion was clearly visualized (Fig. 2B).

One patient (case no. 2, Fig. 3A and B) had previously undergone 2 courses of doxycycline sclerotherapy at another hospital, although she subsequently experienced progressive enlargement of the neck mass. Additionally, preoperative imaging studies of patient no. 4 showed left-sided tracheal deviation (Fig. 4C and D). Otherwise, clinical examinations

revealed no additional clinically important findings. Patients nos. 2 and 4 each received a course of intravenous antibiotics for neck infections just before the operation.

In all 4 patients, the incision was hidden in the hairline (as shown in Fig. 4B). The incision length was <5 cm in the cases of patients nos. 1, 2, and 3, although it was extended to 6 cm in the case of patient no. 4 for 2 reasons. First, the large size of the lesion (largest diameter, 14 cm) necessitated more room for surgical manipulation as seen in Fig. 4A. Second, an intraoperative hemorrhage occurred during the operation due to inadvertent rupture of the ipsilateral distal EJV. For the same reason, an additional 3-cm horizontal incision was created in the lower neck (Fig. 4B, arrowhead) to aid counter-traction. Elevating the skin flap and securely positioning it with Yang retractor took <1 hour in all cases. The mean docking time was 5.5 minutes, and the mean console time was 1 hour and 46 minutes. All 4 surgeries were completed endoscopically with the robot without conversion to open surgery. Bleeding was minimal (i.e., <10 mL) in 3 patients, although the EJV rupture in the case of patient no. 4 yielded a final blood loss of 250 mL despite prompt repair (Table 2). Cystic fluid aspiration from a large LM yielded approximately 750 mL of serosanguinous fluid in patient no. 4.

The postoperative period was uneventful. The average total drainage volume was 21.75 mL (Table 3). No patients required tracheotomy or nasogastric feeding tubes. No cases of surgery-associated hematoma or wound infection were observed. The patients were meticulously monitored for signs of postoperative nerve injury, and no cases of Horner syndrome, facial palsy, marginal mandibular branch injury, or phrenic nerve injury were noted. However, an imaging assessment 11 months after surgery showed that patient no. 2 had a residual 2.3 cm × 3.36 cm × 1.08 cm cystic mass (Fig. 3E and F), which was approximately 5.3% of the LM's original size measured around 4.0 cm × 5.9 cm × 6.0 cm (Fig. 3C and D). Furthermore, an imaging assessment 12 months after surgery showed that patient no. 4 had 2 residual small round cystic lesions with dimensions of 1.3 cm × 1.3 cm and 1.05 cm × 2.3 cm (Fig. 4E and F), respectively, approximately 2.5% of the LM's original size preoperatively (Fig. 4C and D). The patients were kept under vigorous observation, and all residual lesions subsequently resolved spontaneously.

4. Discussion

In this study, we showed, for the first time, that surgical LM resection with the da Vinci robotic system can be safely performed in pediatric patients and that it comes with cosmetic advantages, uneventful recoveries, and good prognoses. There were no cases of postoperative complications, cranial nerve injury, or prolonged drainage, and none of our patients needed a tracheotomy or nasogastric feeding at the end of the follow-up period.

Table 1
Characteristics of the study cohort.

Case no.	Age at operation	Sex	Age at presentation	BMI at operation	Preoperative LM diagnosis	Previous treatments	Postoperative LM diagnosis	Side of neck	Location	Preoperative infection?	Stage
1	1 y 7 m	F	1 y 4 m	12.9	Macrocystic	None	Macrocystic	Left	Level V	No	I
2	3 y 2 m	F	At birth	13.3	Mixed	Doxycycline (2 courses)	Mixed	Left	Level I–V	Yes	III
3	1 y 11 m	M	1 y 10 m	16.5	Macrocystic	None	Macrocystic	Left	Level IV	No	I
4	11 y	M	At birth	25.6	Mixed	None	Mixed	Right	Level I–V	Yes	III

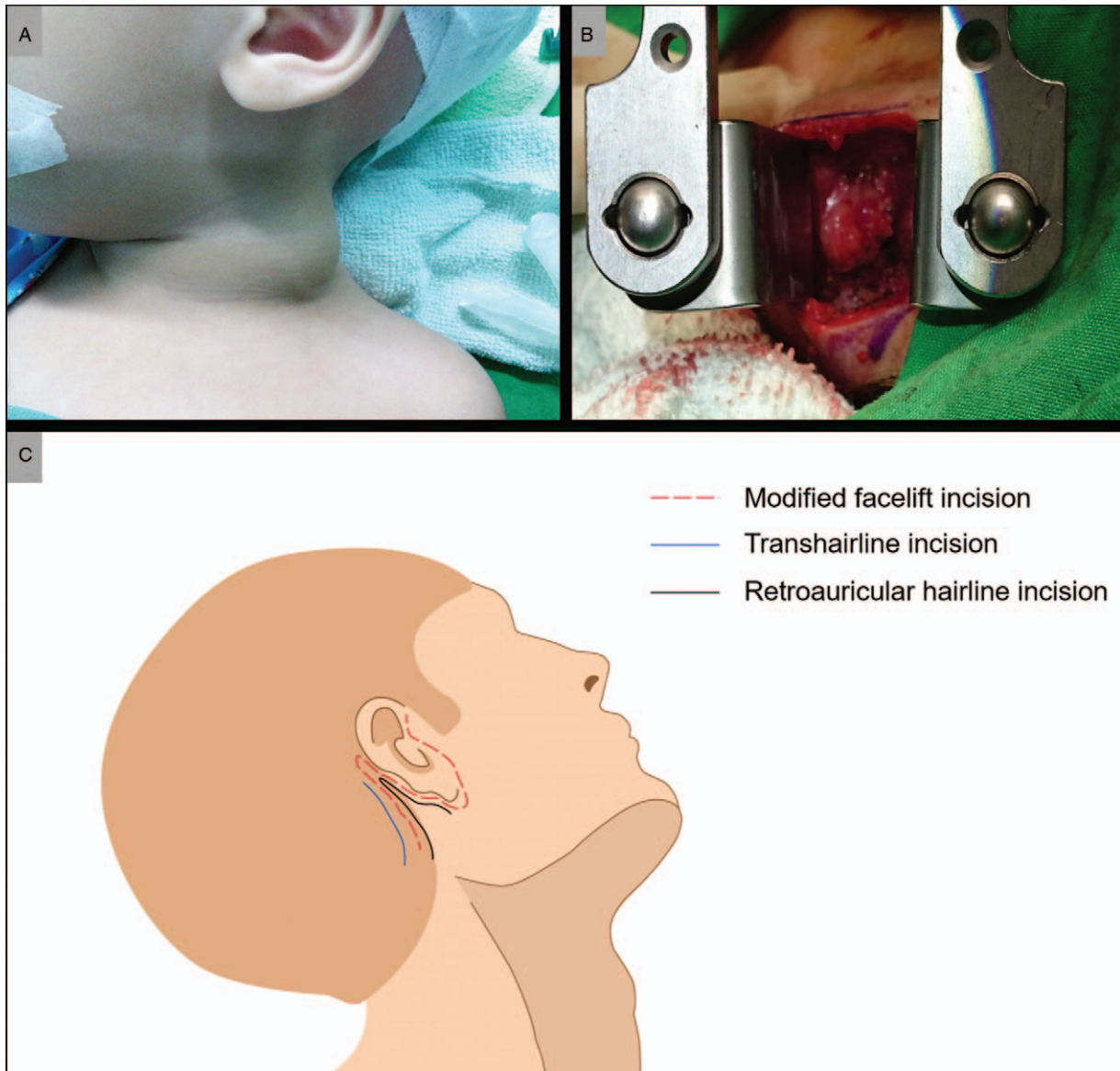


Figure 2. Patient no. 1. (A) Preoperative lateral view showing the left cervical lymphatic malformations. (B) Docking of Yang retractor with a clear surgical view of the cervical lymphatic malformations. (C) Schematic diagram of different minimally invasive approaches for cervical lesions.

Approximately half of LMs (i.e., 45–52% of LMs) occur in the lymphatic fertile region of the head and neck.^[17,18] A retrospective study found that 50% of patients with LMs had asymptomatic neck masses at birth, and 90% of LMs are diagnosed during the first 2 years of life. The treatment of LMs encompasses several modalities, with combined surgical resection and sclerotherapy being an emerging standard of treatment.^[1,3–6] Treatment for LMs is best performed by a multidisciplinary team.^[4,19] The optimal therapeutic approach depends on LM types. The classification system most relevant to treatment decisions categorizes cases according to cyst size into macrocystic LM (previously known as cystic hygroma), microcystic LM (previously known as lymphangioma), and mixed-type LM.

In general, macrocystic LMs are more responsive to either surgical resection or sclerotherapy than microcystic and mixed-type LMs, with the reported treatment response rates for

macrocystic LMs ranging from 86% to 100%.^[20–22] Several sclerosant agents have been used for LM treatment, including ethanol, OK-432, Ethibloc, sodium tetradecyl sulfate (STS), bleomycin, and doxycycline. Combinations of multiple agents (i.e., intralesional irrigation with a 3% STS foam or 98% absolute ethanol before a doxycycline injection) have been trialed, and the results were promising.^[21–23] Recently, treatment protocols that extend over multiple sessions or consecutive days have been proposed, and the existing evidence suggests that such protocols promote functional restoration and yield reasonable improvements in outcomes.^[21] In our series, patient no. 2 had received 2 episodes of sclerotherapy with doxycycline before surgery, although the LM continued to enlarge despite the sclerotherapy. Patient no. 4 was offered the option of aspiration and sclerotherapy with OK-432 for the macrocystic LM before the operation; however, his parents refused the option and chose to undergo the definitive excisional surgery directly.

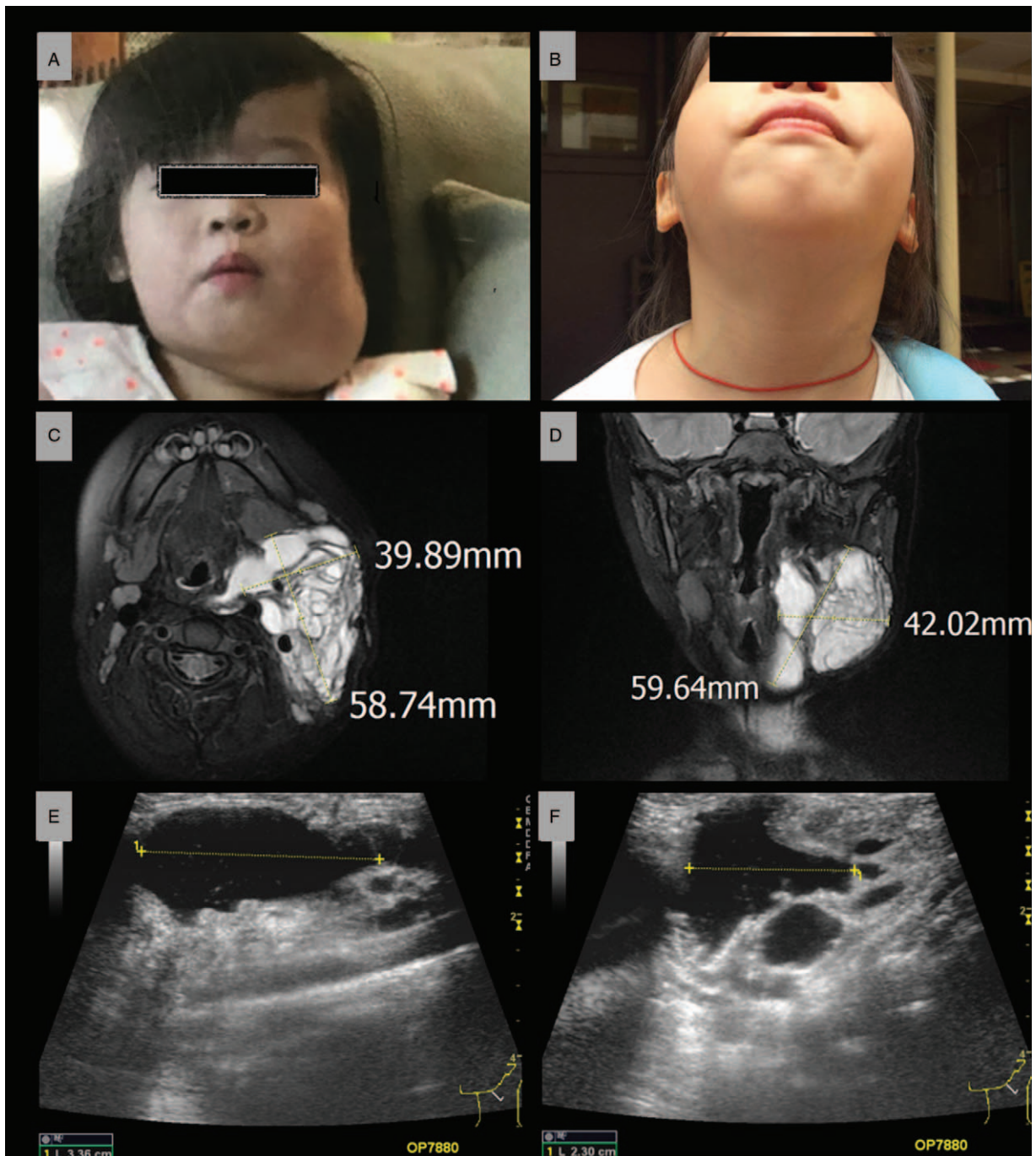


Figure 3. Patient no. 2. (A) Preoperative frontal view illustrating the location and size of the cervical lymphatic malformations. (B) Postoperative basal view of the same patient. (C) Preoperative T2-MRI axial view of the left-sided large mixed-type lymphatic malformations. (D) Preoperative T2-MRI coronal view of the left-sided large mixed-type lymphatic malformations. (E and F) Postoperative sonography images of residual lesions from 2 different angles at 11-month follow-up.

Given the relatively early presentation of LMs and the tendency for lesions to grow, parents commonly feel confused by the available therapeutic options and their respective success rates, procedure-related complication rates, prognoses, cost-effectiveness levels, and esthetic considerations. Therefore, the treatment choice for each case should be individualized based on the clinical presentation, the patient's age and clinical staging, the size and location of the lesion, the patient's comorbidities, and the patient's preferences regarding the preservation of functional and esthetic integrity.^[5,19,24] Objective malformation size reduction is

considered a secondary treatment goal.^[25] Given the inherently benign nature of LMs, the optimal extent of surgical excision must be weighed against the desire to preserve vital structures.

We included 2 patients who had mixed LMs and 2 others who had macrocystic LMs. The existing literature indicates that mixed and macrocystic LMs are more easily accessible for complete resection or even intralesional sclerotherapy than simple microcystic LMs.^[1,6,26] Traditional cervical incision approaches similar to selective neck dissection created non-negligible concerns about postoperative esthetics. Minimally-invasive

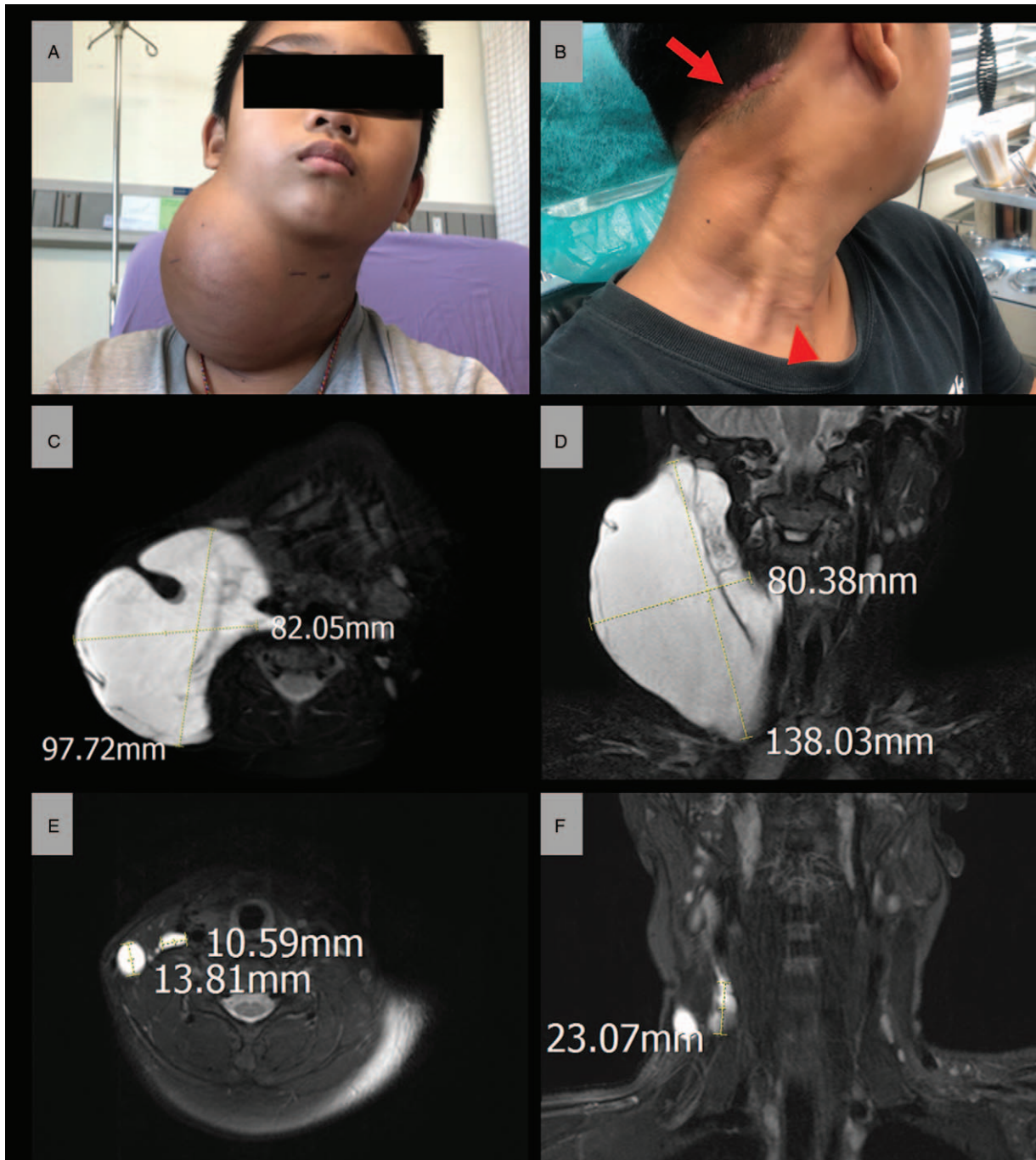


Figure 4. Patient no. 4. (A) Preoperative frontal view illustrating the location and size of the cervical lymphatic malformations. (B) Postoperative lateral view of the same patient. (C) Preoperative T2-MRI axial view of the right-sided large macrocystic lymphatic malformations. (D) Preoperative T2-MRI coronal view of the right-sided large macrocystic lymphatic malformations. Note that the trachea deviated from the midline. (E) Postoperative T2-MRI axial view of a residual lesion at 12-month follow-up. (F) Postoperative T2-MRI coronal view of a residual lesion at 12-month follow-up.

robotic neck surgery via hidden incision approaches then emerged, and the incisions could be categorized into 3 different groups: retroauricular hairline incisions (RAHIs),^[15,27,28] modified facelift incisions,^[29,30] and transhairline incisions,^[13,31–33] as shown in Fig. 2C.

Yang^[32] compared these different robotic surgery approaches in the context of submandibular gland resection and observed substantial differences in 2 variables: average target lesion size >3cm and scars in the postauricular area.

Unlike retroauricular and modified facelift incisions, transhairline incisions achieved incision lengths <5 cm; the former 2 incisions were usually longer in incision lengths. Based on this information, we chose to perform transhairline approach robotic surgeries to minimize incision lengths. Fortunately, the scars could also be adequately concealed within the hair. We found that a transhairline approach effectively reduced the sizes of both macrocystic and microcystic LMs and yielded acceptable cosmetic results.^[34]

Table 2**Operative details of transhairline-approach robotic surgery for pediatric cervical lymphatic malformations.**

Case no.	Surgical approach	Incision size <5 cm?	LM size, cm	Docking time, min	Console time	Bleeding volume <10 mL?	Conversion to open surgery?	Intraoperative complications?	Remarks
1	Transhairline	Yes	2.8 × 2.6 × 1.7	5	1 h 04 min	Yes	No	No	Hemorrhagic cyst (length, 3 cm) connected to the transverse cervical artery
2	Transhairline	Yes	5.6 × 5.4 × 5.2	4	2 h 47 min	Yes	No	No	Macrocytic and microcytic lymphangioma in levels I–V; IJV, and SCM were preserved
3	Transhairline	Yes	4.8 × 3.0 × 3.1	9	0 h 52 min	Yes	No	No	Lymphatic channel, dilated venous component, multiloculated ~4-cm LM
4	Transhairline and cervical	No, 6 cm hairline + 3 cm supraclavicular	14.0 × 7.8 × 9.0	4	2 h 21 min	No, 250 mL	No	EJV injury	Serosanguinous cystic fluid drained (750-mL volume)

Table 3**Outcomes of transhairline-approach robotic surgery for pediatric cervical lymphatic malformations.**

Case no.	Postoperative complications?	Nerve injury?	Total drainage volume, mL	Tracheotomy/Nasogastric feeding?	Postoperative residual LMs?
1	No	No	32	No	No
2	No	No	30	No	Yes, 2.3 cm × 3.36 cm × 1.08 cm (5.3% of the original size)
3	No	No	14	No	No
4	No	No	11	No	Yes, 1.3 cm × 1.3 cm and 1.05 cm × 2.3 cm (2.5% of the original size)

Our transhairline approach robotic surgery method has several drawbacks that must be acknowledged. Concerning lifting the skin flap, one major obstacle is the position of the GAN, which just overlies the SCM and is superficial to the mandibular angle. Numbness around the earlobe can be prevented with careful sub-GAN dissection and tunneling.^[28,35] Because most traction force during lifting of the skin flap is applied to the distal portion of the retractor, the tension on the GAN located at the proximal portion of the retractor is relatively minimal.^[9,36] During the operations discussed in this report, the marginal mandibular branch of the facial nerve was usually left untouched because there was no mass spanning the submandibular area. Furthermore, a small wide flap elevation can preclude angular flap tip necrosis.^[32] At our hospital, we adopted Yang retractor for skin flap elevation and thereby obviated the need for an anterior extension of the skin incision in the retroauricular sulcus. The creation of sufficient working space was generally guaranteed, and there was an adequate range of motion after docking of the robotic arms. In the case of patient no. 4, we also created a supraclavicular cervical incision to provide counter-traction from the third robotic arm in order to remove large masses from the mid-to-lower neck as proposed by Roh et al.^[14,37]

Thorough preoperative planning and sufficient discussion with the patient (or the patient's parents or legal guardians) are crucial before robotic surgery. In our case-series study, none of our patients presented with respiratory distress, abnormal breathing sounds, or heavy respiratory exertions. Therefore, preventive tracheotomies were not scheduled in advance, and the patients' postoperative recoveries were uneventful. Importantly, our patients differed from those described in other case-series reports

regarding postoperative intensive care unit (ICU) admissions. Pediatric ICU admission was routinely arranged for all of our patients to allow airway protection with endotracheal intubation and monitoring for signs of hematoma formation.

We encountered major challenges with patients nos. 2 and 4 due to their infiltrative mixed-type LMs, so their surgical console times were extended to 2 hours, which was longer than the approximately 1 hour console times for the other 2 patients. In the mixed LMs in patients nos. 2 and 4, all macrocystic lesion structures and most microcystic lesion structures were carefully resected as radically as possible while preserving the surrounding essential organs. Sonography and MRI scan obtained approximately 1 year after surgery for patients nos. 2 and 4 revealed residual lesions with volumes that were approximately 5.3% and 2.5%, respectively, of their presurgical values. Cho et al^[34] argued that a >90% reduction in tumor size after surgery should be considered a complete resection, and by that standard, our transhairline approach robotic surgeries led to complete surgical LM resections in all our patients.

In patient no. 4, an iatrogenic rupture of the distal EJV occurred, and suture ligation was performed immediately after extending the transhairline incision wound to a 6-cm length. Nevertheless, the patient's blood loss volume reached 250 mL. Decompression of the cystic fluid content was performed by aspirating approximately 750 mL of serosanguineous cystic fluid. Despite these circumstances, the patient did not experience postoperative complications, cranial nerve injury, or a large amount of postoperative drainage. Given this unfortunate event's manageability and the fact that no other such events occurred, we regard robot-assisted resection of pediatric cervical LMs as a

feasible and practical method for eradicating tumor lesions while preserving vital structures.

There are several disadvantages to performing transhairline approach robot-assisted cervical LM resections in pediatric patients. The working space created by subplatysmal tunneling may be restricted in cases involving large lesion masses. Operators may also encounter challenges, such as limited viewing space, robotic arm collision, and difficulty in assisting bedside surgeons. However, presurgical aspiration of cystic fluid or workspace accommodations for the robotic system could widen the working space and permit shorter incisions. A smaller decompressed cystic mass could also reduce the pressure applied to the respiratory and feeding passages.^[10,38] The use of different angled endoscopes could prove useful under various viewing conditions.^[9,39,40] The docking procedure, which places all arms in appropriate positions and directions, is of utmost importance.^[32]

Our transhairline-approach robotic surgery may not be suitable for lesions with severe adhesion to the surroundings, which may occur after repetitive intralesional sclerotherapy, infections, or laser coagulation. Nevertheless, in our cases, such as patient no. 2, who had undergone previous sclerotherapy, and patients nos. 2 and 4, who had signs of infection and received an antibiotics treatment course, transhairline-approach robotic surgery could still be performed successfully. Additionally, transhairline incisions can also be converted to other incision types, such as RAHIs or retroauricular, transcervical, or combined with supraclavicular incisions, as required due to the anatomical locations of lesions.^[33] Therefore, preoperative imaging studies are crucial for planning suitable docking access and surgical routes and predicting critical points concerning vital structures. Salvage plans should be prepared beforehand and improvised if the surgical team members observe that they cannot proceed with the robotic surgery.^[7,41] Surgeons should also consider managing intraoperative complications, such as cranial nerve injury, hematoma, chyle leakage, or surgical wound infection, and be mindful of the possible need for emergency tracheotomy or organ reconstruction if the surgical team members envision critical conditions (such as vascular injury or nerve injury) during operations. Then, a detailed discussion of surgical indications and risks should be provided to patients and their families.

Another disadvantage of robotic surgery is that it is a relatively experience-based procedure, and the use of robotic surgery in pediatric head and neck surgery is still in its infancy.^[42] Organized training lessons and credentialing of graduates should be required before surgeons can perform robotic surgeries on patients. Periodic maintenance training programs are recommended because dextrous surgical skills are not developed in only days.^[43]

5. Conclusions

Our study shows that transhairline approach robot-assisted surgery is a feasible method for resecting pediatric congenital cervical LMs. Compared with alternative methods, our method has the merits of a smaller incision wound, acceptable esthetic results, and adequate lesion removal even in cases involving giant lesions or previous sclerotherapy. The innovative adoption of Yang retractor facilitates elevating the skin flap, maintaining the distended subplatysmal tunnel, and providing adequate working space for the transhairline approach introduction of the robotic

da Vinci system surgical instruments. The favored surgical paradigm in the head and neck field has gradually shifted from conventional transcervical incisions to minimally invasive transhairline approach incisions. We devised a novel technique for surgical LM resection in pediatric patients, and the results are promising. Further investigations are necessary to address the technique's potential drawbacks and characterize its safety profile.

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