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Thirty-day perioperative outcomes in resection of cervical lymphatic malformations

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ABSTRACT

Introduction: Limited information exists regarding short-term morbidity in the resection of lymphatic malformations. In order to make informed collaborative medical decisions, clinicians and families would benefit from information on 30-day outcomes and the expected course associated with surgical excision of lymphatic malformations.

Methods: A retrospective chart review was conducted to develop a case series of patients who underwent resection of lymphatic malformation at a pediatric tertiary care center between June 1, 2007 and September 30, 2016. Demographic data, disease characteristics, operative details, post-operative care, and adverse events in the 30-day post-operative period were analyzed. Primary outcomes included facial nerve dysfunction, seroma formation, re-admission, and overall rate of any complications. Secondary outcomes included operative time, duration of stay, and duration of drain placement.

Results: Forty-nine excisions were performed in 46 patients (21 male, 25 female). Median age was 5 years. All but 7 cases were performed as the initial primary intervention. Median operative time was 96 min (range 22–224). Higher stage lesions (3-5) were associated with a longer operative time ($p = .03$). Median length of stay was 2 days (range 0–35). Higher stage lesions were associated with an increased length of stay ($p = .0004$). Median duration of drain placement was 2 days (range 0–14), and was longer in higher stage lesions ($p = .0002$). Higher stage lesions ($p = .002$) and cases ultimately found to have residual disease ($p = .019$) were associated with an increased overall rate of complications; there was no association between cyst type and rate of complications. Seroma formation (31%) and transient facial nerve weakness (26%) were the two most common complications observed. There was no association between stage or cyst type and likelihood of seroma formation. Seromas resolved after a median duration of 3 months and transient facial nerve weakness resolved after a median duration of 2 months. 3 patients required return to the OR and 1 patient was readmitted.

Conclusion: The overall rate of adverse events after surgical excision of cervical lymphatic malformations is relatively low. Increased rates of complications can be expected with higher stage. Similarly, for these higher stage lesions, a longer operative time, hospital stay, and duration of drain placement can be expected.

1. Introduction

Lymphatic malformations are one of the most common pediatric head and neck vascular malformations [1]. They are typically present at birth but may only become apparent later in childhood following infection or trauma. Those occurring in the neck may be isolated to one anatomic space but more commonly have finger-like extensions that cross tissue planes making extirpation of these lesions challenging, especially if they demonstrate suprahyoid extension. Intervention is typically pursued for lesions that impact the airway, speech, or feeding, or in certain cases to improve cosmesis (Fig. 1). The mainstays of therapy are sclerotherapy and surgical excision.

In order to make well-informed medical decisions, clinicians and families would benefit from information on the expected perioperative course and rates of complications associated with surgical resection. Evaluation of outcomes following surgical resection of lymphatic malformations in the parotid region has been previously performed, but the existing literature contains little data to describe 30-day perioperative outcomes for resection of cervical lymphatic malformations in the pediatric population. While prior case series have investigated the

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Fig. 1. Clinical Presentation.
This child presented with a large right-sided lymphatic malformation. The lesion had compressed his airway, necessitating placement of a tracheostomy at an early age. The lesion also has a significant cosmetic impact.

management and outcomes associated with these lymphatic malformations, the majority of patients in these studies have undergone observation or sclerotherapy [2,3]. We attempted to query the pediatric American College of Surgeons National Surgical Quality Improvement Program – Pediatrics (ACS NSQIP-P) database, but unfortunately the associated CPT codes are not tracked. The present study is by far the largest series of surgical cases identified in the literature and aims to provide information to families on 30-day outcomes to guide medical decision-making.

2. Methods

The data for this study were collected via retrospective chart review. This historical case series included patients under age 18 years who underwent surgery from June 1, 2007 to September 30, 2016. It was approved by the Institutional Review Board of Ann & Robert H. Lurie Children's Hospital of Chicago. The study focused specifically on cervical malformations; cases that involved superficial parotidectomy were excluded as this has been addressed in another report [4]. The database query (Bio Integration Suite and Clarity) was performed by using relevant Current Procedure Terminology codes (38550, 38555), internal institutional procedure and billing codes, as well as procedure-specific keywords. Most surgical cases were performed by one of two head and neck specialists (J.M. and J.C.R.). Charts were reviewed to confirm the diagnosis and associated procedure.

Baseline and outcomes variables were then collected, including demographics, disease characteristics (e.g. stage, histology), operative details (e.g. extent of surgery, length of surgery), post-operative care (e.g. length of stay, length of drain placement), and adverse events that occurred within the 30-day postoperative period. Lesions were classified as microcystic (individual cysts < 2 cm in diameter), macrocystic (cysts > 2 cm in diameter), or mixed [2]. Lesions were staged according to the classification system devised by de Serres et al. [5], which remains in common use today (Table 1). Adverse events that were tracked included cranial nerve weakness, specifically the facial, recurrent laryngeal, and hypoglossal nerves, seroma, hematoma, wound infection, re-admission, need for additional interventions, return to OR, and any other documented complication. Facial nerve function and seroma were followed beyond the 30-day period to determine total duration, whether transient or permanent, and if additional intervention was required.

Table 1
Staging: de Serres Classification of Lymphatic Malformations.

Stage 1	Unilateral infrahyoid
Stage 2	Unilateral suprahyoid
Stage 3	Unilateral infrahyoid and suprahyoid
Stage 4	Bilateral suprahyoid
Stage 5	Bilateral infrahyoid and suprahyoid

The de Serres staging system for lymphatic malformations was described in 1995. It remains the standard means to classify the extent of the lesion. The system was designed to predict prognosis as well as outcomes and complications associated with surgical intervention.

2.1. Surgery

Surgical procedures were tailored to each individual patient's extent of disease. Each case was characterized by the extent of surgery in regards to laterality as well as involvement of specific regions, including floor of mouth, submandibular gland, parapharyngeal space, and superficial skin resection.

2.2. Statistical analysis

Data were checked for the normal distribution and outliers were removed. Records with incomplete information were excluded only from the associated outcome analysis. Continuous variables (e.g. operative time) were analyzed by using the Mann-Whitney U test. Categorical variables were analyzed by using Fisher's exact test. For statistical analysis, the stage of lesion was dichotomized into “lower stage lesions” (stages 1 and 2) and “higher stage lesions” (stages 3, 4, and 5). In regards to cyst type, microcystic and mixed lesions were grouped together and compared against pure macrocystic lesions.

3. Results

Seventy-six records were examined; of these, 27 cases were excluded due to associated parotidectomy, other procedures misclassified as excision of a lymphatic malformation, or lack of associated records to analyze outcomes (Fig. 2). Forty-nine excisions in 46 patients were included (Table 2); repeat excisions were analyzed as separate cases in 3 of the female patients. Seven cases were performed after prior excision alone, 2 after sclerotherapy alone, 1 after prior sclerotherapy and aspiration, and 1 after prior excision, sclerotherapy, and aspiration. Two patients presented with cervical lymphadenopathy. Median age at time of surgery was 5 years (interquartile range 2–11 years). There was no association between cyst type and stage of lesion ($p = .53$) or between cyst type and overall incidence of complications of any type ($p = .75$). There was, however, an association of higher stage lesions with overall incidence of complications of any type ($p = .002$). Twenty cases (40.8%) demonstrated evidence of residual cystic malformation postoperatively, indicated either by intraoperative assessment or postoperative clinical evaluation. There was no association between presence of residual disease and lesion stage or cyst type. Cases in which residual disease was noted, however, were associated with an increased rate of complications ($p = .02$).

Of the 49 cases analyzed, 46 were unilateral resections while 3 were bilateral resections, and involved a breadth of anatomic areas (Table 3). An increased overall rate of complications approached significance with parapharyngeal exploration ($p = .08$) but not with any other areas of exploration. The median operative time was 96 min (interquartile range

Fig. 2. Case Series Acquisition.
Flow chart of original case query and exclusions to produce the final set for analysis. The original query was based on CPT codes, internal institutional procedure and billing codes, as well as procedure-specific keywords. Patients having undergone parotidectomy were excluded as this population was addressed in another study. Misclassified procedures included entirely unrelated interventions, e.g. abscess drainage, which were inappropriately retrieved in the query.

Table 2
Demographics.

Sex	
Male	21 (46%)
Female	25 (54%)
Stage	
Stage 1	20 (40.8%)
Stage 2	12 (24.5%)
Stage 3	14 (28.6%)
Stage 4	0 (0.0%)
Stage 5	3 (6.1%)
Cyst Type	
Macrocystic	15 (30.6%)
Microcystic	11 (22.4%)
Mixed	23 (46.9%)
Residual Cyst	
Yes	20 (40.8%)
No	29 (59.2%)

Background demographics on the final patient population included in the study. This information was used to evaluate predictive factors relating to surgical outcomes and complications.

Table 3
Surgical details.

Laterality	
Unilateral	46 (93.9%)
Bilateral	3 (6.1%)
Extent	
Submandibular gland excision	8 (16.3%)
Floor of mouth resection	17 (34.7%)
Parapharyngeal space	28 (57.1%)
Skin excision with flap reconstruction	1 (2.0%)

Surgical details for patients in the series. Given that most patient had a stage 3 or lower lesion, it is expected that those associated surgeries would be unilateral. However, there was variety in the extent of surgery and anatomic areas in need of dissection. These data points were evaluated as predictors of complications.

74–138 min). Operative time did not vary significantly by sex, age, cyst type, previous resection, aspiration, or sclerotherapy, presence of cervical lymphadenopathy or necrosis, extent of surgery, or whether residual mass was left behind. However, higher stage lesions were associated with a longer operative time compared with lower stage lesions ($p = .03$). Median length of stay was 2 days (interquartile range 1–3 days, range 0–35 days). Higher stage lesions were associated with a greater length of stay ($p = .0004$). The association of macrocystic lesions with a shorter length of stay approached significance ($p = .06$). The other variables included above for operative time analysis were also analyzed with respect to length of stay, and were found to be non-significant. The median duration of drain placement was 2 days (interquartile range 1–2 days, range 0–14 days). Higher stage lesions were associated with an increased duration of drain placement ($p = .0002$).

Of the 49 cases, seroma formed in 15 (30.6%), with a median duration of 83 days (range 17–330 days). There was no association between stage, cyst type, or length of drain and formation of seroma. There was no association between time to resolution of seroma and cyst stage or cyst type. However, there was an association between likelihood of seroma formation and increased operative time ($p = .03$). Two patients required additional interventions for seroma – one was drained by interventional radiology, while the other returned to the OR on postoperative day 3 for a non-functioning drain. There were two cases of postoperative hematoma. One resolved on its own after 10 days, while the other required re-admission and needle drainage of postoperative day 11 and a return to the OR on postoperative day 13 to control bleeding with placement of a new drain. There were 2 cases of wound infection and one case of wound dehiscence.

Transient facial nerve weakness occurred in 13 cases (26.5%). Median time to resolution was 2 months (range 1–14 months), while permanent facial nerve weakness occurred in one case, a patient with a recurrent stage 5 lymphatic malformation. There was an increased rate of transient facial nerve weakness in microcystic malformations ($p = .04$). There was also an increased rate in cases that involved floor of mouth excision ($p = .04$) and parapharyngeal exploration ($p = .003$). Permanent hypoglossal nerve weakness occurred in the same patient with the permanent facial nerve weakness. Transient accessory nerve weakness occurred in one other case. Transient recurrent laryngeal nerve weakness occurred in 2 cases (4.1%), and took an average of 1.75 months to resolve. One of these two patients developed dysphagia during this time.

Overall, three patients required a return to the OR in the first 30 days, two of which were during the initial admission and one of which was during a re-admission. One patient returned to the OR for a non-functioning drain on postoperative day 3, as above. One returned on postoperative day 4 for repeat sclerotherapy. As already mentioned, one patient was re-admitted on postoperative day 11 for needle drainage of hematoma, and taken to the OR on postoperative day 13 for control of bleeding.

One patient with subclavicular disease experienced intraoperative bleeding that required early termination of the resection. The patient was kept intubated postoperatively for airway management. The patient remained in the hospital for 12 days and required 14 total days of antibiotics to control a surgical site infection.

4. Discussion

As one of the most common pediatric head and neck vascular lesions, lymphatic malformations present a therapeutic challenge for the pediatric head and neck specialist. While small lesions can be observed as they may regress over time, many lesions require intervention, particularly when they affect the airway, speech, feeding, infection, or appearance. While these can sometimes be treated with conservative management including antibiotics and steroids, often more interventional therapy is necessary, and patients must undergo sclerotherapy or surgery. In selecting patients for surgery and establishing expectations for outcomes, clinicians must often rely on limited personal experience.

The present study aims to analyze 30-day outcomes following

excision of cervical lymphatic malformations in the pediatric population at a tertiary academic medical center. The information is helpful not only to better inform families of expectations following surgery, but also to provide valuable data to insurance companies and other healthcare professionals on patient outcomes. In our series, serious complications were fortunately rare following surgery. Significant adverse events included postoperative intubation for intraoperative bleeding and return to the operating room for hematoma and control of bleeding.

The most common postoperative complication was seroma, which occurred in just under one-third of patients. There appeared to be an association between longer operative times and likelihood of seroma formation, potentially due to the greater extent of the lesions in these cases. Seroma formation is more likely in extensive cases as either gross or microscopic portions are often left behind. Seroma formation is not uncommon and can be expected to resolve in 3 months on average. The need for reoperation is rare and is typically only necessary for very large seromas.

Facial nerve weakness was the next most common complication, and occurred in just over a quarter of cases. Of 14 cases of post-op facial nerve weakness, almost all (13; 93%) were transient and resolved by an average of 2 months, while facial nerve weakness in one patient with Stage 5 disease requiring floor of mouth resection appeared to be permanent. For patients who require floor of mouth excision or parapharyngeal exploration, the family should be counseled regarding the increased potential risk of transient weakness. However, as with the previously cited study involving lesions of the parotid, full recovery of the nerve can be expected in most cases [4].

Previous studies have suggested that there is an increased risk of complications following resection of higher stage lesions [6]. This was confirmed in the present series, with lesions stage 3 or greater having a higher rate of overall complications than lesions stage 2 or less. Additionally, according to the literature, microcystic and mixed lesions are more difficult to treat with either surgery or sclerotherapy compared with pure macrocystic lesions, and are associated with more complications [7–9]. In the present study, an association between microcystic and mixed lesions with transient facial nerve weakness was demonstrated, and the association with a longer hospital stay approached significance. These lesions were not, however, found to be associated with higher rates of many of the other complications tracked, including incidence of seroma. Although it may be expected that microcystic or mixed lesions would have a higher probability of residual cyst compared with macrocystic lesions, we did not find this in the present study.

Surgery for lymphatic malformations has the fewest complications when performed in the posteroinferior neck, parotid, and submandibular regions [1,10], but risks are present with any procedure. Lymphatic malformations involve variable territories and potentially involve several important structures. Cranial nerves, and in particular the facial nerve, can be intimately associated with the malformation. In these cases, subtotal excision, rather than total resection, may be preferred to preserve vital structures. In the current study, two-fifths of patients had some evidence of residual disease, and for many of these patients there may have been an attempt to preserve critical structures. When there is a choice between high risk for injury to a critical structure and complete disease removal, the decision is often made to leave disease behind, and families should be counseled as such.

There are limitations in drawing conclusions from this study. The study context is a high volume tertiary academic medical center. Records beyond 30 days were not always complete, and so definitively identifying the duration of seroma or facial nerve weakness was challenging. The number of cases available for analysis limits the conclusions that can be drawn. Lastly, given our study is not a randomized controlled trial, it is designed only to demonstrate association, not causation.

5. Conclusions

To date, case series are few which have investigated the short-term outcomes of surgical intervention for pediatric lymphatic malformations of the neck. The present study is the largest identified to describe outcomes following surgical resection of cervical lymphatic malformations in the pediatric population. The overall rate of adverse events was low, the most common being seroma and transient facial nerve weakness. Major adverse events were particularly rare. The above data provide useful information to clinicians in medical decision-making, as well as set expectations to families regarding perioperative outcomes and risks. Future studies should be designed to gather additional prospective data, investigate outcomes in other healthcare delivery settings, and further elucidate the role of perioperative sclerotherapy in the management of these patients.

Conflict of interest notification

We have no conflicts of interest to disclose.

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