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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