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Case Report

Lessons learned to aid the successful outcome of pediatric recurrent laryngeal nerve reinnervation

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ABSTRACT

While Ansa to recurrent laryngeal nerve reinnervation is gaining popularity in the treatment of unilateral vocal fold immobility, little has been reported on commonly encountered surgical challenges with this procedure. Here, we present a cohort of 21 pediatric patients who underwent this procedure with a full description of techniques used to overcome common challenges with this procedure. We report vocal and swallowing outcomes for these patients, with an overall success rate of 19/21 (90.5%) patients, with success defined as improvement of a half-consistency or resolution of vocal issues.

1. Introduction

Ansa to recurrent laryngeal nerve reinnervation (ANSA-RLN) has emerged over the past decade as an effective option in the treatment of unilateral vocal fold immobility (UVFI) in the pediatric population and is gaining popularity [1–4]. This procedure is an additional option to be considered with injection medialization laryngoplasty (IML) and thyroplasty [5]. A recent survey of the ASPO pediatric laryngology working group revealed that in the setting of a longstanding iatrogenic UVFI, 22% and 37% would offer ANSA-RLN and concomitant IML as a first line therapy in young children and teenagers, respectively [6]. However, this increasingly popular procedure presents particular surgical challenges that demand careful consideration.

ANSA-RLN reinnervation was first described in adults by Frazier in 1924 [7], with a report by Crumley in 1991 of the first reinnervation in an 8 year old child [8]. ANSA-RLN provides an advantage over other surgical interventions for UVFI in its ability to re-establish muscle-tone and bulk, thereby preserving vocal fold tension [4]. The procedure has been further briefly described in the context of cohort studies [5,8], however the literature has failed to describe many important surgical considerations of this nuanced procedure. Previous accounts have outlined evaluation of these patients along with preoperative, perioperative, surgical, and postoperative considerations, however they do not fully discuss many of the challenges this procedure can present [9]. Challenges include reliable identification of the ansa cervicalis, adequate mobilization of the thyroid gland with preservation of vasculature, reliable identification of the recurrent laryngeal nerve (RLN), and choice of anastomosis technique depending on nerve caliber.

We describe surgical techniques for ANSA-RLN that have proven to be successful over the course of twenty-one cases by the senior author. We have found reliable landmarks by which identification of the ansa cervicalis is the most fruitful, thyroid dissection techniques to provide safe mobilization with preservation of necessary vasculature along with rapid and consistent identification of the RLN, and methods of neurorrhaphy that provide an adequate conduit for ANSA-RLN. We present 21 cases to support that these techniques have proven to provide successful outcomes for our patients.

2. Methods

Study patients were recruited from all patients referred to our tertiary care center for feeding difficulty or hoarseness as a result of vocal cord paralysis who underwent ANSA-RLN between June 2006 and September 2018. All patients first underwent IML both prior to reinnervation as well as at the time of ANSA-RLN. All patients had lesions on the left side. Sixteen of the twenty-one patients had UVFI following thoracic surgery, 1 had UVFI following esophageal surgery, 2 had UVFI following neurologic surgery, 1 had UVFI following laryngotracheal reconstruction, and 1 had UVFI of idiopathic etiology (Table 1). Twelve patients presented with both aspiration and vocal issues, 5 were seen

Abbreviations: ANSA-RLN, ansa to recurrent laryngeal nerve; RLN, recurrent laryngeal nerve; UVFI, unilateral vocal fold immobility; TVF, true vocal fold

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Table 1
Demographics, Medical History, and Presenting symptomatology.

Patient Information	Total (n = 21)
Sex	
Male (%)	11 (52%)
Female (%)	10 (48%)
Age	
Median	5.0
IQR	3.0–14.0
Etiology	
Thoracic surgery	16 (76%)
Neurosurgery	2 (9.5%)
Esophageal surgery	1 (4.5%)
Airway reconstruction	1 (4.5%)
Idiopathic	1 (4.5%)
Presenting symptom	
Vocal issues (%)	4 (19%)
Aspiration (%)	5 (24%)
Both (%)	12 (57%)

IQR = Interquartile Range.

for aspiration alone, and 4 were seen for vocal issues alone. Patients were included in the study if they were seen in follow-up at least 6 months following ANSA-RLN. Resolution of vocal issues was noted & agreed upon by subjective assessment by the examiner, the caregiver, and the patient. Resolution of aspiration was defined by recommendation to liberalize diet by at least a half-consistency (e.g. half-honey to nectar thick liquid).

Continuous variables are reported using median and interquartile ranges (IQR). Categorical data are presented as percentages.

2.1. Surgical technique

A lateral neck incision just superior to the level of the cricoid (Fig. 1) was planned for adequate exposure. The incision was carried down through skin and subsutaneous tissue in a standard fashion. Both blunt and electrocautery dissection was carried down along the anterior border of SCM, with both wide superior and inferior exposure. It is important to continue dissection down to the level of the omohyoid inferiorly, as this will lead to the proper identification of internal jugular vein.

In cases of normal anatomy, with development of dissection along the undersurface of the omohyoid and along the internal jugular vein, the loop of the ansa cervicalis was identified either medial or lateral to the vein (Fig. 2a). Early cases did involve identification of the hypoglossal nerve and branching of the decendens hypoglossi – however, we found over time that with inferior identification near omohyoid, this superior identification with the hypoglossal nerve was unnecessary. Division of the omohyoid was sometimes necessary in order to provide

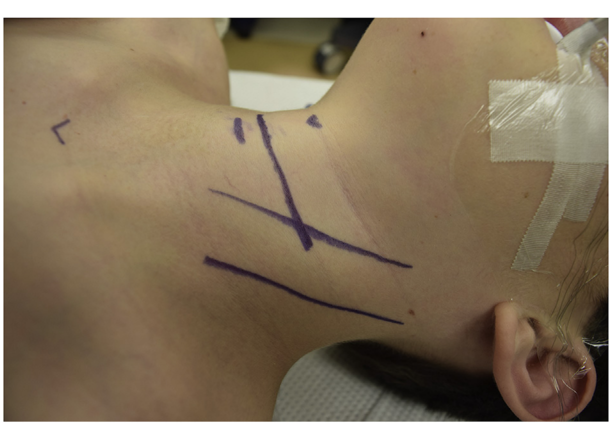

Fig. 1. Incision planning.

adequate exposure and length for tension-free anastomosis (Fig. 2b). However, anatomic variants (such as in one case of double aortic arch) sometimes led to identification of inferior branching as well.

Once the ansa cervicalis was identified, attention was then turned towards identification of the RLN. Straps were split in the midline and dissection was continued down to the cricoid cartilage. The thyroid isthmus was divided with electrocautery so as to allow for a medial to lateral dissection of the superior pole of the thyroid (Fig. 3). This was necessary in order to perform medial dissection along the trachea to Berry's ligament, which allowed for the first step in mobilization of the thyroid gland. Mobilization was aided by development of the avascular plane between the cricothyroid muscle and the medial surface of the superior lobe of the thyroid gland, allowing the superior lobe to be grasped and retracted inferiorly. Ligation of superior lobe vessels allowed for ease of exposure of the RLN, which is usually located deep to the thyroid gland prior to entry posterior to the cricothyroid joint. This was most easily encountered with careful dissection right along thyroid tissue. Adequate mobilization of the thyroid tissue medially allowed for more clear dissection in this area. Additionally, the superior parathyroid gland was best preserved with close dissection along thyroid capsule.

Following identification of both RLN and ansa cervicalis, ansa was divided low (just deep to omohyoid). Skeletonization with micro-dissection instruments provided adequate length for a tension-free anastomosis. If branching was encountered, these branches were sacrificed in order to achieve adequate length on the nerve. The RLN was similarly divided with about 1–2cm of length to cricothyroid joint. A tunnel deep to straps through the middle layer of the deep cervical fascia was created, providing a conduit for the ansa to meet the RLN (Fig. 4a). After freshening the cut ends of both nerves, end-to-end neurorrhaphy with two-point fixation was performed with 9–0 nylon using microinstruments. If the ansa cervicalis caliber was 2x larger than that of the RLN (select cases), end-to-end neurorrhapy was performed with the redundant ansa fibers not included in the anastomosis. Neurorrhaphy was always performed using 2.5x loupe magnification.

3. Results

Twenty-one patients, 11 male and 10 female, successfully underwent ANSA-RLN with the described technique. The median age (IQR) was 5.0 (3–14) years (Table 1). The procedure was performed with operative times ranging from approximately 2-3 hours depending on the ease with which the Ansa and RLN are identified (due to variation in robustness of nerve & age of child). No patients were lost to follow-up. The median length of follow-up (IQR) was 7.9 (6.13–16.3) months. Of the 4 patients who presented with vocal issues alone, all 4 experienced resolution of hoarseness after 6 months status-post ANSA-RLN. Similarly, the 5 patients presenting solely with aspiration issues demonstrated improvement by at least a half-consistency on VFSS with clinical resolution of aspiration issues after 6 months status-post ANSA-RLN. Finally, of the 12 patients who presented with both aspiration and vocal issues, 10 patients demonstrated resolution of both (83.3%), with one patient noting continued hoarseness with resolution of aspiration and another patient noting improved hoarseness with occasional episodes of choking with feeds.

4. Discussion

In this study, we present our surgical experience following 21 ANSA-RLN procedures. Our primary goal, above all else, was how to best achieve a tension-free anastomosis in order to promote speedy reinnervation and neural growth. A number of techniques are paired in order to achieve this goal, which have allowed for ease of exposure of these nerves and superior outcomes for our patients.

Ease of identification of the ansa cervicalis is paramount for success of this procedure. Anatomic study of the ansa cervicalis has yielded

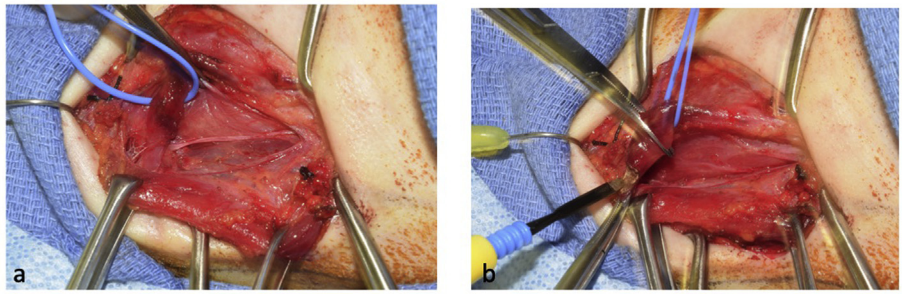

Fig. 2. Exposure of (a) ansa with (b) division of omohyoid.

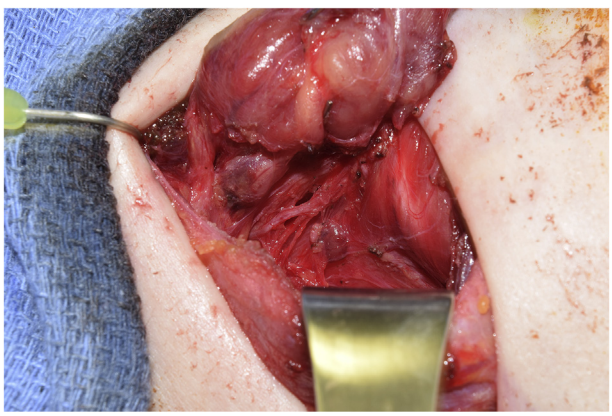

Fig. 3. Reflection of superior lobe of thyroid for RLN identification.

information of variation. Yamada described two categories of the inferior root of the ansa in relation to the carotid sheath as the medial and lateral types [10]. The lateral type describes the inferior root running anterior to the vein to form the ansa cervicalis on the anterior surface of the carotid sheath. Conversely, the medial type refers to the inferior root running posterior to the internal jugular vein and carotid sheath [11]. Yamada suggests the medial type is almost twofold more prevalent than the lateral type in the Japanese population [11]. This must, however, be considered in the context of population-based variation, as American and European textbooks tend to describe the anatomy as the lateral type [12].

Techniques to identify the RLN have been described in the literature on thyroid surgery [13]. Of course, ANSA-RLN is performed in the context of preservation of thyroid and parathyroid tissue. As none of our patients had post-operative wound healing, infectious, or endocrine issues, we report that adequate preservation of thyroid and parathyroid tissue was likely achieved. Furthermore, our experience is consistent with other literature where direct neurorrhaphy is most successful when compared to nerve-muscle pedicle and direct implantation techniques [14].

It has been suggested in cadaveric that microscope use produces superior quality digital nerve repair with a higher level of magnification associated with better clinical outcomes [15]. However, a study on digital sensory nerve repair demonstrated no significant difference in sensory recovery between repair with an operating microscope versus 2.5–3.5x loupe magnification [16]. Our experience supports the latter study, with the overwhelming majority of our patients experiencing success following two-point neurorrhaphy performed with 2.5x loupe magnification.

As ANSA-RLN procedures become more prevalent in the treatment of UVFI, the refinement and evolution of surgical technique is necessary in order to achieve optimal outcomes for patients. Here, we discuss the approach that we have found to be successful in performing this procedure. Without a 100% success rate, however, there is of course room for improvement. Apart from technique, future areas of improvement include better evaluation of neural circuit status to best predict outcomes of reinnervation through intraoperative neural testing and stimulation. Continued dialogue and larger studies will be helpful as the field moves forward and techniques continue to evolve and change.

Contributors' statement

Dr. Caloway was involved in execution of the study, acquisition of data, analysis and interpretation of data, drafting the article, and final approval of the final manuscript as submitted. Drs. Bouhabel and Hartnick were involved in planning and execution of the study, as well as critical review and revision of the manuscript and approval of the final manuscript as submitted.

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Declaration of competing interest

The authors have no conflicts of interest to disclose.

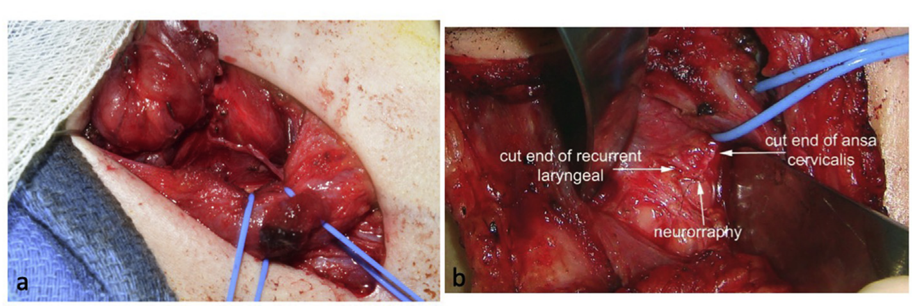

Fig. 4. Tension free anastamosis (a) following tunneling under strap muscles and (b) with neurorrhaphy.

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