Postoperative Observation of Children after Endoscopic Type 1 Posterior Laryngeal Cleft Repair

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Abstract

Objectives. To report the perioperative management and surgical outcomes in a large series of pediatric patients with endoscopically repaired type 1 posterior laryngeal cleft (PLC).

Study Design. Case series with chart review.

Setting. Urban, tertiary care, free-standing pediatric hospital.

Subjects and Methods. Patients who underwent endoscopic carbon dioxide laser–assisted repair of type 1 posterior laryngeal clefts between January 2006 and December 2012. Medical records were reviewed.

Results. Fifty-four patients (34 male) underwent repair of type 1 PLC. Median age was 25.5 months (range, 2-120 months). Indications for repair included aspiration (n = 39; 72%), chronic bronchitis (n = 13; 24%), and stridor with feeds (n = 2; 4%). No children remained intubated postoperatively. Thirty-three patients (61%) stayed in overnight observation (“Obs PLC”) and 21 patients (39%) stayed in the pediatric intensive care unit (“PICU PLC”) postoperatively. Between Obs PLC and PICU PLC groups, there was no significant difference in age (mean 2 vs 30 months, respectively; P = .28). Comorbidities were similar between the groups. Symptoms improved in 41 of the 54 patients (76%). No postoperative complications were noted. Two patients required revision PLC repair. The cost of admitting a patient to a lower acuity location was estimated to be 60% less per day than cost of a PICU admission.

Conclusions. The endoscopic surgical repair of a type 1 PLC is successful and has a low morbidity and complication rate. Patients may be safely managed in an observation unit and without postoperative intubation. This approach achieved a marked cost reduction in postoperative care.

Keywords
dysphagia, aspiration, posterior laryngeal cleft

A posterior laryngeal cleft (PLC) is a rare congenital anomaly resulting from failure of the fusion of the tracheoesophageal septum or the posterior cricoid lamina. This creates an abnormal communication between the larynx and the hypopharynx, which can range in severity depending on its inferior extension.1,2 Although a PLC was first described by Richter in 1792, it was not until 1955 that the first successful repair was performed by Pettersson.3,4 In 1989, Benjamin and Inglis5 developed the classification system of PLCs that is most commonly used today. Tucker and Maddalozzo6 described the submucous posterior laryngeal cleft, which is a defect in the posterior cricoid cartilage with intact overlying mucosa and interarytenoid muscle.

Patients with a type 1 PLC can present with aspiration, recurrent pneumonia, chronic lung disease, dysphagia, stridor with feeding, choking, chronic cough, and/or failure to thrive. However, Myer et al7 reported that 21% of type 1 PLCs are diagnosed incidentally without any symptoms. Formal diagnosis requires an airway evaluation under anesthesia with palpation of the posterior larynx with a right angle probe to gauge the depth of the interarytenoid notch.8 However, guidelines and indications for surgical intervention once a type 1 cleft is diagnosed are unclear.

Both modified barium swallows (video fluoroscopic swallow studies [VFSS] at our institution) and fiberoptic endoscopic evaluation of swallowing (FEES) are used in the evaluation, management, and follow-up of children with PLCs.4,9-11 Several studies have reported successful

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nonsurgical and surgical management of type 1 PLCs. Endoscopic surgical techniques of PLC repair have become the treatment of choice for type 1 PLCs that do not respond to conservative or medical management. Surgical techniques have been widely reported, but postoperative management of these children remains controversial and little evidence is available to guide practice.

To our knowledge, our study represents the largest series of surgically managed type 1 PLCs. The goal of our study was to evaluate pediatric patients with endoscopically repaired type 1 PLCs and to detail their postoperative management. At our institution, children have been admitted to the pediatric intensive care unit (PICU) after an endoscopic repair of a type 1 PLC. More recently, based on surgeon preference, some of these children have been admitted to the surgical observation unit (an outpatient stepdown unit). This study evaluates whether there is utility to justify the added expense of PICU admission in these children. We also describe our technique for carbon dioxide (CO₂) laser-assisted surgical repair of type 1 PLCs and evaluate the success rate and complication rate of this technique.

**Methods**

**Data Acquisition**

The Ann and Robert H. Lurie Children’s Hospital of Chicago Institutional Review Board approved this study. We selected patients by searching a surgical database using Current Procedural Terminology (CPT) code 31599, unlisted procedure of the larynx, from January 2006 to December 2012. Operative reports were reviewed, and patients who had undergone endoscopic repair of laryngeal clefts were identified. Patients with type 2, type 3, and type 4 PLCs and incomplete records were excluded. Demographic data including sex, age at repair, indication and presenting symptoms, and comorbidities were collected. Preoperative evaluations of swallowing were reviewed and compared with postoperative evaluations. Cases were categorized into 2 groups—patients who were managed in the postoperative observation unit (“Obs PLC”), which consists of 1 nurse for 4 postoperative patients of varying types and continuous pulse oximetry monitoring, and patients who were managed in the PICU (“PICU PLC”), which consists of 1 nurse for 2 patients and standard ICU monitoring. The nurses in both locations are certified in pediatric advanced life support and have completed yearly training courses on the recognition of respiratory distress in children and the level-appropriate management of these children. Clinical improvement was defined as tolerating a thinner consistency of liquids then was tolerated prior to surgery or a complete resolution of aspiration. The cost of a PICU admission was compared with the cost of an observation/floor status admission.

**Statistics**

Statistical analysis was performed with an analysis of variance and a parametric t test. Variables between the 2 groups were compared with the Student t test, and P values were determined by the chi-square test analysis. Epi-Info software was used to calculate the P values between numbers and percentages.

**Surgical Technique**

Type 1 PLCs were surgically repaired by an endoscopic technique previously described. Preoperative steroids are administered intravenously (dexamethasone sodium phosphate 0.5-1.0 mg/kg). Patients are placed in a supine position on the operating table. General anesthesia is initiated with the patient spontaneously ventilating. Topical lidocaine hydrochloride (2% or 4%) is atomized to the supraglottic, glottic, and subglottic airway. Laryngoscopy is performed using a Lindholm laryngoscope or a Benjamin anterior commissure laryngoscope. Bronchoscopy is performed with a Hopkins rod 0° telescope. An insufflation anesthetic technique is used, with an endotracheal tube (6.0) attached to the side port of the laryngoscope or through a flexible suction catheter (12F) placed through the nasal cavity and positioned in the hypopharynx. The patients are not intubated. The operating microscope and CO₂ laser (at a setting of 4 W, superpulse, with a 2-mm spot size) are used. Vocal cord spreaders are placed in an inverted position with the shaft running along the superior portion of the laryngoscope and the handles secured to the Louie suspension arm. A diamond-shaped incision is made with the laser in the interarytenoid area with the apex of the diamond at the depth of the cleft. Any char is wiped intermittently with a one-half-inch cottonoid dampened with oxymetazoline (0.05%) on an alligator forceps. The cleft is closed with a single purse-string suture. A 6-0 or 7-0 polydioxanone (PDS) suture on a taper needle.

**Results**

Over the 6-year period reviewed (2006-2012), 54 children underwent repair of a type 1 PLC. Age at repair ranged from...
from 2 to 120 months (mean, 25.3 months). Twenty patients were female (37%) and 34 patients were male (63%). There were multiple indications for repair. The primary diagnoses in our population were aspiration (n = 39; 72%), chronic bronchitis (n = 13; 24%), and stridor with feeds (n = 2; 4%). It was unclear how many patients had a combination of these problems. However, based on the swallowing evaluations, all the patients had some form of dysphagia with aspiration regardless of the primary indication listed for surgery.

Preoperatively, swallowing evaluation was performed with a FEES examination in 2 patients (4%) and with a VFSS in 48 patients (89%). Postoperatively, 12 patients (22%) were evaluated with a FEES examination, and 36 patients (67%) underwent VFSS; VFSS evaluations ranged from 0.15 to 59 weeks postoperatively (mean, 8.03 weeks) and FEES evaluations ranged from 2.4 to 20 weeks postoperatively (mean, 8.55 weeks). Total mean follow-up duration was 18.7 months (range, 0.26-104.6 months).

All patients underwent repair as described earlier in this article. Operative times ranged from 21 to 180 minutes (mean, 64.01 ± 30.6 minutes). No patients required intubation postoperatively. Thirty-three patients (61%) were placed in overnight observation (Obs PLC group) following repair, and 21 patients (39%) were observed in the PICU (PICU PLC group) postoperatively. There was no significant difference in age between Obs PLC and PICU PLC groups (mean 22 vs 30 months, respectively; P = .28) (Table 1). The median length of stay (LOS) for Obs PLC was 1.4 days (SD, 1.8 days), while the median LOS for PICU PLC was 3.14 days (SD, 6.3 days); P = .24. There were no intraoperative complications or postoperative complications in either the Obs PLC or the PICU PLC group. None of the children required airway intervention (such as continuous positive airway pressure, bilevel positive airway pressure, or intubation) postoperatively in either group. Six of the 33 Obs PLC patients had a supraglottoplasty performed at the same time as their PLC repair, via a cold steel technique that has been previously described. There was no significant difference in surgical indications between the 2 groups. Dysphagia improved in all children who underwent concomitant supraglottoplasty.

Overall improvement in dysphagia as determined by comparing the preoperative and postoperative swallowing assessments was found in 41 of the 54 children who underwent repair (76%). The number of children who were improved postoperatively was not significantly different between Obs PLC (26/41) and PICU PLC (15/41) groups (P = .53). Additional subgroup analysis revealed improvement in 30 of the 39 children who experienced aspiration, 10 of the 13 children with chronic bronchitis, and 1 of the 2 children who had stridor with feeds. Of the 9 children with no improvement in aspiration after surgery, 6 had more than 2 comorbidities each, including developmental delay, subglottic stenosis, chronic lung disease, tracheomalacia, seizure disorder, and/or a syndromic diagnosis. Of all the patients who had surgery, 6 were noted to have neurological comorbidities; 4 did not improve after PLC repair, and 2 improved postoperatively. Of the 41 children with dysphagia that improved, only 5 required continued thickening at a thinner consistency than was required preoperatively (usually thin nectar) at the first postoperative swallowing assessment. The rest no longer required thickened liquids.

There was no significant difference in mean age (22 vs 30 months; P = .28) or length of stay (1.4 days Obs PLC vs 4.4 days PICU PLC; P = .19) in patients who were not improved from the repair of their PLC. Additionally, postoperative location (postoperative observation unit vs PICU) was not indicative or predictive of a lack of postoperative improvement; 7 of 33 Obs PLC patients (21%) and 6 of 21 PICU PLC patients (28.6%) were not improved.

The 2 groups of patients had similar comorbidity profiles. Of the Obs PLC patients, 17 had concomitant medical problems including gastroesophageal reflux, esoinophilic esophagitis, esophageal stenosis, and prematurity. Of the PICU PLC patients, 19 had other comorbidities including asthma, gastroesophageal reflux, developmental delay, Opitz Frias syndrome, patent ductus arteriosus following closure, and prematurity. One PICU patient had several major medical problems including pyloric stenosis, vocal fold paralysis, phrenic nerve paralysis, and tracheoesophageal fistula following repair. The Obs PLC group included 6 formerly premature patients, whereas the PICU PLC group included 5 such patients. The PICU PLC patients had a larger variety of medical problems (but not necessarily more severe), although the number of patients with comorbidities was nearly the same (n = 19) compared with the Obs PLC group (n = 17).

The cost of an average 24-hour stay in the surgical observation unit at our institution is estimated at $2433, compared with $5941 for the same duration in the PICU.

Discussion

This study is the largest series describing children with type 1 PLC who were managed surgically. The number of patients with surgically managed type 1 PLCs who were described in previous studies has ranged from 1 to 28.1,4,5,8-10,12-17,19-21 Type 1 PLCs are now diagnosed more commonly, with earlier studies suggesting an incidence of 0.1% to 3%1,22-25 and more recent studies reporting an incidence of 6% to 8%.4,8,9,12,15,16 This increase in incidence has been attributed to a high index of suspicion for the presence of an airway cleft at the time of airway endoscopy, improved endoscopic detection techniques, and increased referrals to tertiary care pediatric medical centers.1,15

Several studies have reviewed management for type 1 PLCs. In 1998, Parsons et al3 reported on 41 patients with type 1 PLCs who had been observed over a 9-year period (incidence of 7.1%); all were managed conservatively with observation, thickening of feeds, and reflux precautions. In 2006, Chien et al2 reported on 20 patients who had been observed over a 3-year period (incidence of 7.6%); the investigators recommended conservative management for 1 month before pursuing operative management. Eighty percent of their patients did not improve and went on to surgical repair.9
Ketcham et al\textsuperscript{10} and Watters and Russell\textsuperscript{4} advocated a slightly longer period of conservative management—2.5 months and 4 to 8 months, respectively. Seventy-five percent of the patients in Watters and Russell’s study failed conservative management. Kubba et al\textsuperscript{14} recommended proceeding directly to surgical repair on all symptomatic type 1 PLCs. Rahbar et al\textsuperscript{15} recommended surgical management based on (1) clinically apparent aspiration with feeding, (2) severity of pulmonary status, (3) imaging studies, (4) presence or absence of other significant comorbid conditions predisposing to aspiration, (5) findings on upper aerodigestive endoscopy, and (6) poor response to medical management and feeding therapy.\textsuperscript{15}

Bakthavachalam et al\textsuperscript{1} reported 59 patients with type 1 PLCs. Fifteen children were managed with endoscopic repair (after a trial period of approximately 6 months of conservative management), and 44 children were successfully managed conservatively. From those data, the investigators recommended medical management in children under the age of 18 months whose aspiration could be adequately managed with compensatory strategies such as thickening liquid oral intake. These patients were noted to have resolution of their aspiration around the same age regardless of how early they received the diagnosis or underwent endoscopic repair. The children whose aspiration did not resolve by the age of 18 months were unlikely to show spontaneous resolution and therefore would benefit from surgical intervention.

Multiple surgical techniques to repair type 1 PLCs have been described. The least invasive of these techniques involves injection of material such as Gelfoam into the laryngeal cleft to increase the vertical height of the interarytenoid mucosa.\textsuperscript{21} Open surgical techniques (laryngofissure and lateral pharyngotomy) have largely been replaced by endoscopic repair of type 1 PLCs.\textsuperscript{9,21,26} Endoscopic repair has primarily used of a CO\textsubscript{2} laser either (1) to create microflaps within the cleft that are reaproximated with multiple sutures or (2) to denude the mucosal margins of the cleft and allow for suture apposition of the mucosal edges.\textsuperscript{1,4,9,10,15,17} Both techniques are reported to have high success rates.\textsuperscript{1,4,9,10,15}

Our success rate is 76%. This is comparable to the findings of other published series: Rahbar et al\textsuperscript{15} reported 80% overall improvement, Watters and Russell\textsuperscript{4} reported 100% symptom resolution following repair, Bakthavachalam et al\textsuperscript{1} reported 73% improvement of the severity of aspiration, and Ketcham et al\textsuperscript{10} reported symptom resolution in 57% (laser approximation) and 78% (microflap) (not significantly different). The determination of a successful outcome following type 1 PLC repair remains somewhat variable given the inhomogeneity of patient populations, comorbidities, periods of observation and medical management, and objective evidence versus subjective perception of improvement. In this study we define improvement as resolution or improvement of laryngeal aspiration based on the findings of the postoperative VFSS or FEES. Aspiration is the most common indication to pursue surgical management of a type 1 PLC, and both VFSS and FEES are suited to diagnose the presence of aspiration.\textsuperscript{10,11,16,17} Eighty-nine percent of our patients were evaluated with VFSS at an average of 8 weeks postoperatively. We feel that this time frame is adequate for the resolution of any postoperative edema within the supraglottic larynx that may affect handling of thin liquids. It is unclear whether the timing of the postoperative VFSS or FEES examination influenced the overall result. However, all patients who underwent a postoperative FEES or VFSS during their hospital stay were evaluated by the surgeon and a speech and language pathologist in outpatient follow-up and would have had a follow-up instrumental examination of swallowing as clinically indicated. Repeat examinations were not needed in any of the children who demonstrated resolved aspiration after repair in this study. Johnston et al\textsuperscript{27} showed persistent aspiration in 7 of 24 patients (29%); associated comorbidities included hypotonia, tracheo esophageal fistula, gastrostomy dependence, and syndromic diagnosis. In our study, 13 patients (24%) were persistent aspirators postoperatively and 9 of these had 2 or more comorbidities. Nearly all of the 9 had developmental delay, and other comorbidities included subglottic stenosis, seizure disorder, syndromic diagnoses, and tracheomalacia. One patient in this study had Opitz Frias syndrome, and this patient’s aspiration improve after surgery. There were 3 children with tracheo esophageal fistula, 2 of whom improved postoperatively.

There is some controversy regarding the need for postoperative intubation after PLC repair. Chien et al\textsuperscript{9} recommended postoperative intubation for 1 to 10 days to allow for wound healing. Koltai et al\textsuperscript{13} and Holinger et al\textsuperscript{24} advocated no postoperative intubation. Ketcham et al\textsuperscript{10} kept their patients who underwent the laser reapproximation technique intubated, while not requiring intubation for those who underwent the microflap technique. Watters et al\textsuperscript{17} recommended close airway observation in the PICU overnight and a floor observation the second postoperative night. Watters’...
feeding regime after the child is fully awake.\textsuperscript{4,17} Sandu and Montier\textsuperscript{22} recommended postoperative noninvasive ventilation. Leboulanger and Garabedian\textsuperscript{2} reported initiation of postoperative oral feeding after 7 to 14 days. At our institution, the postoperative management protocol has transitioned from postoperative observation in the PICU to an overnight observation in a floor status room with continuous pulse oximetry. To our knowledge, our report is the first that demonstrates similar surgical outcomes with no increase in postoperative complications in patients who were managed in 2 different monitoring settings following type 1 PLC repair. Both groups had a similar clinical presentation and comorbidities. Incorporation of a speech and language pathologist to assist in safe postoperative resumption of the feeding is very important. Most children remained on their own individualized preoperative feeding regime until postoperative VFSS or FEES confirmed the absence of aspiration on thinner liquids. Six patients in the Obs PLC group underwent endoscopic type 1 PLC repair and supraglottoplasty for concurrent laryngomalacia. There were no increased complications or increased laryngeal edema in these patients postoperatively. The presence of laryngomalacia in children with laryngeal clefts has been described in several previous studies.\textsuperscript{1,2,12,16} The management of the supraglottoplasty airway without postoperative intubation is a controversial subject and one that deserves further investigation.

After surgery, patients were admitted for at least 24 hours to either the lower acuity Obs PLC group or the higher acuity PICU PLC group. More patients were admitted to observation (n = 33) than PICU (n = 21) depending on surgeon preference. Given that the comorbidity profiles of the 2 groups were very similar with no discrepancy in postoperative complications, the main difference in the management of these patients is the cost of hospitalization.

An average 24-hour stay in the surgical observation unit at our institution totals $2433, compared with $5941 for the same duration in the PICU. Triaging postoperative patients to a lower acuity unit translates to an estimated cost savings of $3508 per day, with no observed increase in postoperative complications.

While there was a trend toward a shorter length of stay in the observation PLC group, this did not reach statistical significance. Limitations of this study include those inherent to a retrospective review. We concede that selection bias may have determined which patients were selected to observation in the PICU. However, review of the data did not demonstrate any statistically significant difference with respect to medical comorbidities in these groups. The follow-up time for each patient was not entirely standardized. However, all patients were managed per our standard protocol, which calls for instrumental evaluation of swallowing 4 to 6 weeks after the procedure. While this is the largest series to date of operatively repaired type 1 PLCs, this remains an overall uncommon condition; thus, the study population may not be large enough to detect a complication in patients in either the Obs PLC group or the PICU PLC group. A strength of this study was the use of the same surgical technique for all patients who underwent repair of their type 1 PLC.

**Conclusion**

Type 1 PLCs can be managed medically. However, in children who require surgical intervention, the endoscopic surgical repair of a type 1 PLC is successful and has a low complication rate. We feel that these patients may be safely managed postoperatively without intubation, nasogastric feeds, or mandatory ICU monitoring. This may allow for a significant cost reduction while maintaining a high level of patient safety.

**Author Contributions**

Nathan S. Alexander, acquisition and analysis of data, drafting, final approval, accountable; Judy Z. Liu, acquisition and analysis of data, drafting, final approval, accountable; Bharat Bhushan, analysis of data, drafting, final approval, accountable; Lauren D. Holinger, conception, critical revision, final approval, accountable; James W. Schroeder Jr, conception, critical revision, final approval, accountable.

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