The Role of Injection Laryngoplasty (IL) in Treating Deep Interarytenoid Notch (DIN) Associated Dysphagia in Young Children

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ABSTRACT

BACKGROUND

Deep Interarytenoid Notch (DIN) is the mildest form of laryngotracheoesophageal cleft defect and is frequently found in young children with dysphagia and aspiration. Treatment guidelines are not defined. Injection laryngoplasty (IL) is a surgical procedure injecting polymer gel into the tissue around the defect. Our objective was to evaluate the efficacy of IL in pediatric populations with severe dysphagia and aspiration.

METHODS

We conducted a pilot retrospective chart review of DIN patients under 36 months who underwent IL at PCH. Severity of dysphagia before and after IL was measured using modified barium swallows (MBS) (scale 0-10) and documented symptoms. Statistical analysis was done using paired two sample t-test with a p value of 5 percent.

RESULTS

Patients with initial MBS above double honey (7) improved an average of 2.6 ±1.38 points while those with initial scores below 7 did not see a statistical change in MBS. At the end of the study period, the final MBS scores for both groups were statistically similar.

CONCLUSIONS

IL treatment for DIN associated dysphagia results in improvement of MBS scores and symptoms in toddlers with severe aspiration. Careful timing of IL impacts morbidity. Future prospective controlled studies are necessary to evaluate the role of IL and medical interventions in thickener wean and clinical improvement.

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INTRODUCTION

Laryngotracheoesophageal clefts are a rare aero-digestive pediatric malformation with an annual incidence of one per ten thousand to one in twenty thousand but seen in high frequency in specialized airway centers [1-3].

Presenting symptoms are varied but most commonly are dysphagia and aspiration, GERD, chronic cough, recurrent pneumonia, and feeding problems [1-3]. Deep interarytenoidal notch (DIN) is a variant of laryngotracheoesophageal clefts defined as a notch-shaped defect limited to the supraglottic interarytenoid area, and is considered the mildest form of laryngotracheoesophageal defect. Management for patients presenting with DIN is currently divided into thickener weaning until the patient outgrows the defect and/or surgical bulking of the affected area with surgical grade gels [4]. One commonly employed surgical treatment modality is injection laryngoplasty (IL) which is a minimally invasive procedure where gel is injected into the tissue surrounding the defect acting as a filler to occlude the defect [5-7]. This protocol is commonly utilized in children 1-3 years age where other surgical repairs would be invasive or technically limited due to the underdeveloped airway [8-9]. However, despite the frequent utilization of IL in this patient population the clinical impact on dysphagia with aspiration of DIN treated with IL is poorly studied [8-10]. The Aerodigestive clinic at PCH follows 120 patients with DIN and our goal was to evaluate radiologic and clinical outcomes of patients with DIN and to identify methods for stratifying risk versus benefit in potential candidates for IL in children 1-3 years with dysphagia and aspiration.

METHODS

This retrospective chart review study was conducted by collecting the records of children who had undergone injection laryngoplasty at the Phoenix Children's Hospital Aerodigestive Clinic during the past two years. As part of the inclusion criteria, patients had to be between one and three years of age and have been diagnosed with a deep interarytenoidal notch at the Aerodigestive center on video laryngoscopy or modified barium swallow. Additionally, to be included in the study group, patients had to have undergone at least two radiologist reviewed modified barium swallows during treatment; one occurring prior to the injection and one occurring within six months after injection.

Modified barium swallow studies included in this analysis were conducted at PCH. Patients were fed a spoonful of water thickened to a recognized standard thickener level (i.e. nectar, honey, double honey) and mixed with a barium tracer. The swallow was observed radiographically and patients were fed progressively thinner preparations until microaspiration was noted by the radiologist. The modified barium swallow was graded based upon the last thickness that no aspiration occurred at.

In addition, patient demographic information was collected from the electronic patient record including: age, sex, weight at initial visit, and if the patient was a preterm birth which is a known risk factor for tracheoesophageal malformations. Individual patient visits were also reviewed to ascertain what symptoms that the patient was experiencing both prior and post treatment.

Patients were divided into two groups: patients who had a reduction in modified barium swallow score as a result of injection laryngoplasty, and those that did not. These patient groups were compared to evaluate for differences between the populations present at initial consult that could serve as a predictor of eventual treatment success.

Analysis was performed using paired t-test of pretreatment and posttreatment modified barium swallow scores in Microsoft Excel. Additionally, the presence or absences of various symptoms were evaluated using chi-squared tables also done in Excel.

RESULTS

Patient demographics were collected at the time of initial consult with the Phoenix Children's Hospital Aerodigestive clinic. The patients were subdivided into two populations: patients with a decrease in their modified barium swallow study score at 6 months post injection and those that saw no change or an increase in modified barium swallow score. These two patient populations were entitled benefit and no benefit respectfully.

In Table 1 the patient demographics and calculated p values for the two populations are listed graphically. For this analysis statistically significant variability between these populations would be set at a p value of 0.05.

In Table 2 the entire study population was taken as a whole. The initial modified barium swallow score and last modified barium swallow score done in a six month period after treatment with injection were taken and compared using a paired two sample t-test.

In Table 3 the modified barium swallow at initial presentation for both study populations (benefit vs. no benefit) are compared using a two sample t-test. Mean modified barium score for the benefit population is measured at 8.15 versus 5.4 for the no benefit population. This difference was statistically significant at a p value of 0.0003.

In Table 4 the modified barium swallow scores measured within six months after intervention with injection laryngoplasty for the two study populations are compared with a two sample t-test. The mean modified barium swallow score for the benefit population was measured at 5.5 versus 6 for the no benefit population. The difference measured between these populations was not statistically significant at a p value of 0.56.

In Table 5 the reported symptoms as a fraction of each study population before and after intervention with injection laryngoplasty are presented. P values for these figures were calculated using a Chi-squared table. The percent reporting symptoms after intervention decreases (or remained the same in the case of feeding refusal) but was only a statistically significant change in the cases of gagging and coughing reported in the no benefit populations.

In Table 6 the raw data for the modified barium swallow scores is presented. Data is sorted by patient identifier and by months prior to or after intervention by injection laryngoplasty denoted by the T-# nomenclature. The initial column is the first pre-intervention score documented for a patient, and the final column is the last documented modified barium swallow score within the six month post treatment window. The change column is the initial column minus the final column.

Figure 1 shows the scoring schema used to convert the radiologist read of the modified barium swallow studies into the raw data presented in Table 6. On the scale, the thinnest possible mixture using the thickener agent is rated a 1 and the scale incremental increases to a thickness of double honey. Patients who aspirate at double honey score a 10. Patients not requiring any thickener whatsoever earn a zero.

Mean (std dev)		Benefit n = 13	No Benefit n = 10	All n = 23	P value B vs NB
Age in Months		20.2 (5.8)	24.0 (8.0)	21.9 (6.9)	0.22
Sex	Female	6	5	11	0.85
	Male	7	5	12	
Weight in Kg		10.9 (2.3)	11.8 (1.6)	11.3 (2.0)	0.31
% Preterm		0.31	0.10	0.22	0.23

Table 1: Patient Demographics, Benefit vs No Benefit Populations

ALL PATIENTS: BEFORE VS AFTER t-Test: Paired Two Sample for Means			
Change in MBS Score	*Baseline*	*Final*	Improvement
Mean	6.956522	5.73913	1.217391304
Variance	3.952569	3.474308	
Observations	23	23	
Pearson Correlation	0.450643		
Hypothesized Mean Difference	0		
df	22		
t Stat	2.887979		
P(T<=t) one-tail	0.004269		
t Critical one-tail	1.717144		
P(T<=t) two-tail	0.008538		
t Critical two-tail	2.073873		

Table 2: All patients, MBS scores before versus after IL

STARTING MBS SCORES FOR THE TWO GROUPS t-Test: Two-Sample Assuming Unequal Variances		
Did IL help this patient?	***Did not help***	***Helped***
Mean	5.4	8.153846154
Variance	2.488888889	1.807692308
Observations	10	13
Hypothesized Mean Difference	0	
df	18	
t Stat	-4.421363217	
P(T<=t) one-tail	0.000164762	
t Critical one-tail	1.734063607	
P(T<=t) two-tail	**0.000329525**	
t Critical two-tail	2.10092204	

Table 3: Benefit versus No Benefit starting MBS score comparison

FINAL MBS SCORES: HELPED VS DID NOT HELP t-Test: Two-Sample Assuming Unequal Variances		
Did IL help this patient?	***Did not help***	***Helped***
Mean	6	5.538461538
Variance	3.111111111	3.935897436
Observations	10	13
Hypothesized Mean Difference	0	
df	20	
t Stat	0.589072604	
P(T<=t) one-tail	0.281203942	
t Critical one-tail	1.724718243	
P(T<=t) two-tail	**0.562407885**	
t Critical two-tail	2.085963447	

Table 4: Benefit versus No Benefit final MBS score comparison

Percentage Reporting Symptoms						
Group Period	Benefit (n=13) Initial	 After IL	p value	No Benefit (n=10) Initial	 After IL	p value
Choke	0.38	0.23	**0.39**	0.60	0.40	**0.37**
Gag	0.31	0.15	**0.35**	0.60	0.10	**0.02**
Vomit	0.46	0.31	**0.42**	0.60	0.50	**0.65**
Feeding Refusal	0.23	0.00	**0.06**	0.20	0.20	**1.00**
Cough	0.46	0.23	**0.22**	0.80	0.20	**0.01**

Table 5: All patients, symptoms reported pre-IL versus post-IL

Patient	Initial	T-12	T-11	T-10	T-9	T-8	T-7	T-6	T-5	T-4	T-3	T-2	T-1	T+1	T+2	T+3	T+4	T+5	T+6	Final	Change
1	5				5			5						5						5	0
2	9	9						7						7						7	2
3	7			7			7							7		7		5		5	2
4	7		7		3			7	7	7				7			4	7	5	5	2
5	7						7			7				9	9				7	7	0
6	9						9					7		7				7		7	2
7	7				7					7	5		7	5				0		0	7
8	7							7						9						9	-2
9	7				7		7	7					5	4			7			4	3
10	7											7		6					6	6	1
11	10						10		10				10	9	7					7	3
12	7				7						6			6		5			5	5	2
13	3		3						3	5	5			4	4					4	-1
14	10									10				7		7			7	7	3
15	5					5				6			6	7						7	-2
16	9		9					9						7	7					7	2
17	5				5						7			5						5	0
18	7		7									7		4			7			7	0
19	5			5	7				6					6						6	-1
20	7		7		7				7					7	7				5	5	2
21	7						7	10						7						7	0
22	10									10				7					7	7	3
23	3			3						3				3						3	0

Table 6: Patient MBS scores raw data

Ultrathin	*1*
Thin	*2*
½ nectar	*3*
¾ nectar	*4*
Nectar	*5*
¾ honey	*6*
Honey	*7*
¾ double	*8*
Double	*9*
npo	*10*

Figure 1: MBS scores

DISCUSSION

Overall, the twenty-three patients included in this retrospective chart review averaged an improvement of 1.2 points on the modified barium swallow thickener scale (figure 1). However, on further analysis it became apparent that some patients were benefiting from intervention with injection laryngoplasty while others were seeing no improvement.

When the study population was subdivided into patients with eventual benefit (n=13) and patients with no benefit (n=10) or worsening modified barium swallow score it became apparent that these two patient populations looked very different at initial presentation. Table 3 quantifies the disparity at initial consult. The no benefit group had an initial average modified barium swallow study score of only 5.4 and a maximum score of seven. In comparison, the group that saw benefit had an initial average score of 8.2 with a minimum score of 7. From this comparison it is evident that the initial severity of disease was very unequal in the two study populations with the population that saw benefit having more serious disease.

Furthermore, Table 4 compares the endpoints of the two study populations. In this table patient's last modified barium swallow study done after intervention with injection laryngoplasty were compared. Here we see that patients that did benefit from the injection had an average score of 5.5 versus an average of six seen in the no benefit population. Analysis shows that the two study populations are no longer statistically different from each other in regard to swallow study.

In addition to modified barium swallow study data the patient's charts were reviewed for reported symptoms. Table 5 summarizes these findings for the two study populations. Both populations saw either a decrease or no change in the percentage reporting the various symptoms, however, only the no benefit population had a statistically significant change in reporting frequency after intervention. Interestingly this seems to contradict the apparent lack of improvement in the modified barium swallow scores seen in the study group bringing into question the reliability of patient reported symptoms as a marker for clinical improvement.

CONCLUSIONS

Injection laryngoplasty can be a helpful treatment for the correction of aspiration as a result of deep interarytenoid notch in young children. In particular, greatest improvement is seen in patients who present with thickener requirements of at least honey thick.

Additioanlly, modified barium swallow correlation with reported clinical symptoms was not a reliable marker of radiographic severity of aspiration.

STUDY LIMITATIONS

As a retrospective study data recording is inherently limited in a few key ways. Patient symptoms were collected by reviewing patient notes and the accompanying review of symptoms. Providers were not told at the time of the patient interaction that this data would be reviewed for symptom resolution and data may have been omitted from the patient record. Additionally, no standardized protocol was in place at the time of intervention and as such the frequency of modified barium swallows was inconsistent between patients.

FUTURE DIRECTIONS

We purpose a pilot prospective study looking at MBS clinical symptoms correlation in children 1-3 years of age and dysphagia with oral aspiration using current treatment protocols. Prospective analysis of IL versus IL and rapid thickener wean protocol feeding therapy study. The ultimate goal is to achieve a treatment modality that results in decrease need for thickener in treatment of pediatric dysphagia and reduction of radiation exposure and associated dysphagia morbidities.

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