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ORIGINAL RESEARCH

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Risk-adjusted analysis of perioperative outcomes after the Sistrunk procedure

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Abstract

Objectives: Thyroglossal duct cyst (TGDC) is the most common pediatric congenital neck mass. The Sistrunk procedure is the standard method of excision and is associated with low rates of recurrence. This study aimed to review our institution's outcomes following the Sistrunk procedure, specifically the rates of wound complications and cyst recurrence.

Methods: This was a retrospective case series of pediatric patients undergoing the Sistrunk procedure from June 2009 to April 2021.

Results: A total of 273 patients were included. Of these, 139 (53%) patients were male and 181 (66%) were white. The average age at the time of surgery was 7.1 years. The overall cyst recurrence rate was 11%. The most common wound complications were seroma (14%) and surgical site infections (SSIs) (12%). Wound complications were associated with prior history of cyst infection (odds ratio [OR] 1.97, 95% confidence interval [CI] 1.07–3.60, z-test 2.2, $p = .03$). Pediatric surgery was associated with fewer wound complications (OR 0.18; 95% CI 0.05–0.6, z-test −2.78, $p = .005$). However, pediatric surgery operated on fewer patients with a history of cyst infection (36% vs. 55%, $p = .012$). Drain placement and postoperative antibiotics did not affect rates of wound complications.

Conclusions: Prior cyst infection is associated with increased rates of postoperative wound complications. Postoperative antibiotics and drain placement did not significantly affect complication rates.

Level of Evidence: 4.

KEYWORDS

postoperative complications, recurrence, Sistrunk procedure, thyroglossal duct cyst

1 | INTRODUCTION

Thyroglossal duct cysts (TGDCs) are the most common pediatric congenital neck masses. They arise due to failed obliteration of the thyroglossal duct and can occur anywhere along the duct's course.[1,2] Patients typically present with a midline neck mass. Cyst infection is common and may occur in up to 70% of children.[3,4]

Indications for surgical intervention include recurrent infection, cosmesis, fistula formation, and risk of malignancy.[5] Recurrence is a known complication from surgical intervention, and proposed risk

factors for recurrence include method of excision,[6] patient age,[7,8] preoperative cyst infection,[2–4,7,9–12] and cyst histologic features.[7,9,13] The traditional Sistrunk procedure is the gold standard method of TGDC excision, and is associated with lower rates of recurrence and postoperative complications compared to simple cyst excision.[14] Additional modifications to the traditional Sistrunk procedure have demonstrated reduced rates of recurrence.[15,16]

Although there has been extensive research focused on reducing rates of recurrence after the Sistrunk procedure, there is no consensus on the risk factors predisposing for recurrence. In addition, literature on risk factors associated with postoperative wound complications in children after the Sistrunk procedure is limited.

The aim of this study was to perform a large-scale review of pediatric patients undergoing the Sistrunk procedure at our institution. The effect of preoperative cyst infection, surgeon specialty, and drain placement on the rates of postoperative wound complications and cyst recurrence were analyzed.

2 | MATERIALS AND METHODS

This retrospective review was approved by the University of Texas Southwestern Institutional Review Board (STU 2019-1666). Patients less than 18 years old who underwent the Sistrunk procedure between June 2009 and April 2021 were identified by searching for current procedural terminology codes 60280 "excision of thyroglossal duct cyst or sinus" and 60281 "excision of thyroglossal duct cyst or sinus, recurrent" in the electronic medical record database. Only patients who underwent removal of the central portion of the hyoid bone, resection of the suprahyoid soft tissue along with the cyst, were included. Children who underwent simple cyst excision or had non-TGDC pathology were excluded.

Patient demographic details at the time of surgery were collected from the electronic medical record (Epic Systems Corporation, Verona, WI, USA). Physician notes from our institution were reviewed for data collection. These included: age (years), height (cm), weight (kg), body mass index (BMI) (kg/m^2), BMI percentile, sex (male/female), race (Black or African American, White, Asian, more than one race, other), and ethnicity (Hispanic/non-Hispanic). Preoperative variables collected included: age at presentation (years), history of cyst infection (yes/no), number of courses of antibiotics for cyst infection, history of cyst incision and drainage (I&D) (yes/no), history of previous excision (yes/no), presence of a fistula or sinus tract (yes/no), maximum cyst size on imaging (cm). Intraoperative variables included: admission status (inpatient vs. outpatient), active infection at time of surgery (yes/no), surgeon specialty (otolaryngology/pediatric surgery/other), intraoperative cyst rupture (yes/no), estimated blood loss (EBL) (mL), and drain placement (yes/no). Postoperative variables included: postoperative antibiotic administration (yes/no), admission (yes/no), length of stay (LOS) (days), and cyst recurrence (yes/no). Data were collected and managed using a REDCap electronic database hosted at the University of Texas Southwestern.[17]

TABLE 1 General data.

Number of patients	273
Mean age at surgery, years (SD)	7.1 (4.4)
Median age at surgery, years (IQR)	6.1 (3.6–9.5)
Male sex	139 (53%)
Race	
White	181 (66%)
Black or African American	48 (18%)
Asian	4 (1%)
Other	40 (15%)
Hispanic or Latino ethnicity	137 (50%)
Mean BMI percentile (SD)	65.8 (29.8)
History of cyst infection	139 (51%)
Previous antibiotic treatment	148 (54%)
Previous I&D	24 (9%)
Previous cyst infection	139 (51%)
Fistula or sinus tract present	50 (19%)
Previous cyst excision	22 (8.1%)
Infection at time of surgery	30 (11%)
Surgeon specialty	
Pediatric otolaryngology	220 (81%)
Pediatric general surgery	53 (19%)
Intraoperative cyst rupture	82 (31%)
Drain placement	193 (71%)
Inpatient admission status	187 (69%)
Mean LOS, days (SD)	0.9 (0.9)
Postoperative antibiotics prescribed	133 (49%)
Cyst recurrence after Sistrunk	31 (11%)

Abbreviations: BMI, body mass index; IQR, interquartile range; I&D, incision and drainage; LOS, length of stay.

The primary outcomes of interest were cyst recurrence and postoperative wound complications. Postoperative wound complications included surgical site infection (SSI), seroma, and hematoma. Primary outcome data were collected from review of postoperative clinic and/or ED visit documentation.

2.1 | Statistical analysis

All statistical analyses were performed with Stata (StataCorp. 2021 *Stata Statistical Software: Release 17*, StataCorp LLC, College Station, TX, USA). Univariate analysis was performed using the Pearson X^2 test for categorical variables and the analysis of variance (ANOVA) for continuous variables. We examined the risk of postoperative wound complications using a multivariable logistic regression model that included weight, age, history of cyst infection, history of previous excision, surgeon specialty, and previous cyst recurrence. A forward stepwise regression model of variable that exhibited a p value <.25 in

the univariate model were added and variable with a p value >.05 were sequentially dropped. The Hosmer–Lemeshow test was performed to assess model fit. A p value <.05 was considered significant.

3 | RESULTS

A total of 273 pediatric patients were included for analysis (Table 1). Patients were equally distributed by sex and were primarily white (66%). The mean age at the time of surgery was 7.1 (SD 4.4) years. The mean follow-up time was 7.8 (SD 15.5) months.

At presentation, 139 (51%) patients had a history of cyst infection, and 148 (54%) patients had received one or more courses of antibiotics. A fistula or sinus tract was present in 50 (19%) patients. Twenty-four (9%) patients previously underwent cyst I&D and 22 (8.1%) patients presented with cyst recurrence after previous surgical excision. Thirty (11%) patients presented with an active infection at the time of surgery. One hundred eighty-seven patients (69%) were admitted for observation after surgery. The median LOS was 0.9 (IQR 0–0.9) days.

3.1 | Cyst recurrence

There were 31 cases of cyst recurrence and an overall recurrence rate of 11.4%. Rates of cyst recurrence were higher in patients with preoperative cyst infection but did not reach significance (15% vs. 6.7%, $p = .052$). Rates of cyst recurrence were also significantly higher in those with wound complications compared to those without (30.6% vs. 5.7%; $p < .001$).

3.2 | Prior cyst infection

One hundred and thirty-nine (51%) patients had a history of prior cyst infection at presentation. Patients with a history of cyst infection had a higher average BMI percentile compared to those without (70.2 vs. 61.5; $p = .029$). They were more likely to have a history of prior cyst excision (14% vs. 2%, $p < .001$) and active infection at time of surgery (17% vs. 5%, $p < .001$). Postoperative wound infections were more common in patients with a history of cyst infection compared to those without (18% vs. 5%; $p = .002$). See Table 2 for further characteristics of patients with history of cyst infection.

3.3 | Surgeon specialty

Two hundred and twenty surgeries (81%) were performed by pediatric otolaryngologists 53 (19%) were performed by pediatric surgeons. Otolaryngologists were significantly more likely to operate on patients

TABLE 2 Factors associated with prior cyst infection.

	Prior cyst infection ($n = 139$)	No history of cyst infection ($n = 132$)	p value
Mean age at surgery, years (SD)	7.6 (4.3)	6.6 (4.5)	.063
Mean BMI percentile (SD)	70.2 (28.9)	61.5 (30.1)	.029
Fistula or sinus tract present	47 (34%)	3 (2%)	<.001
Previous antibiotic treatment			<.001
None	3 (2%)	122 (92%)	
1	70 (50%)	10 (8%)	
2	30 (22%)	0 (0%)	
3	17 (12%)	0 (0%)	
4+	19 (14%)	0 (0%)	
Previous cyst excision	20 (14%)	2 (1.5%)	<.001
Surgeon specialty			.012
Pediatric otolaryngology	120 (86%)	98 (74%)	
Pediatric general surgery	19 (14%)	34 (26%)	
Active infection at surgery	24 (17%)	6 (4.5%)	<.001
Intraoperative cyst rupture	54 (40%)	27 (21%)	<.001
Drain placement	108 (78%)	83 (63%)	.008
Discharge antibiotics prescribed	88 (63%)	43 (33%)	<.001
Postoperative wound complication			
Seroma	22 (18%)	12 (10%)	.095
Hematoma	3 (2%)	4 (3%)	.652
SSI	22 (18%)	6 (5%)	.002
Cyst recurrence	21 (15%)	10 (7.6%)	.052

Abbreviations: BMI, body mass index; SSI, surgical site infection.

with prior cyst infection (55% vs. 36%, $p = .012$). In addition, patients operated on by pediatric otolaryngology received significantly more courses of antibiotics prior to their surgery ($p < .001$). Pediatric otolaryngologists were significantly more likely to place a drain ($p < .001$), admit patients after surgery ($p < .001$), and prescribe postoperative antibiotics ($p < .001$). See Table 3 for further characteristics of patients by surgeon specialty.

3.4 | Drain placement

A drain was placed in 193 (71%) cases. Drains were placed more often in patients with a history of cyst infection ($p = .008$) and in cases for recurrent cysts ($p = .008$). The average EBL was higher in cases where a drain was placed (7.8 vs. 5.7 mL, $p = .49$). Drain placement was almost exclusively performed by otolaryngologists (99%; $p < .001$), and patients with a drain were more likely to be admitted postoperatively ($p < .001$). Average LOS was longer in patients with a drain (1.0 vs. 0.5, $p < .001$). Drain placement was not associated with a significant difference in rates of wound complications or cyst recurrence. See Table 4 for further characteristics.

3.5 | Wound complications

Sixty-two patients experienced wound complications postoperatively. Seven patients were reported to have both postoperative seroma and SSI. There were a total of 69 postoperative wound complication events and an overall wound complication rate of 25%. The most common wound complications were seroma (14.1%) and SSI (11.6%), followed by hematoma (2.9%). Rates of SSI were higher with prior history of cyst infection (18% vs. 5%, $p = .002$). Postoperative antibiotics did not affect rates of postoperative wound complications. Of the cases with postoperative complications, 95% were performed by pediatric otolaryngologists ($p < .001$). See Table 5 for further characteristics by wound complications. On multivariate analysis, Sistrunk procedures performed by pediatric surgery (0.18, 95% confidence interval [CI]: 0.05–0.60, $p = .005$) were negatively associated with postoperative wound complications. Prior cyst infection

TABLE 3 Factors associated with surgeon specialty.

	Pediatric otolaryngology ($n = 220$)	Pediatric general surgery ($n = 53$)	p value
Mean age at surgery, years (SD)	7.1 (4.5)	6.9 (4.2)	.81
Mean BMI percentile (SD)	67.0 (30.1)	60.1 (28.3)	.20
Fistula or sinus tract present	42 (19%)	8 (15%)	.53
History of cyst infection	120 (55%)	19 (36%)	.012
Previous antibiotic treatment			.008
None	90 (41%)	35 (66%)	
1	70 (32%)	12 (23%)	
2	25 (11%)	5 (9%)	
3	16 (7%)	1 (2%)	
4+	19 (9%)	0 (0%)	
Courses of previous antibiotics (SD)	1.1 (1.3)	0.5 (0.8)	<.001
Previous cyst excision	21 (10%)	1 (2%)	.07
Active infection at surgery	25 (11%)	5 (9%)	.68
Mean EBL, mL (SD)	8.0 (8.5)	3.8 (3.1)	<.001
Intraoperative cyst rupture	58 (27%)	24 (45%)	.01
Drain placement	191 (87%)	2 (3.8%)	<.001
Inpatient admission status	176 (80%)	11 (21%)	<.001
Discharge antibiotics prescribed	181 (18%)	5 (9%)	<.001
Postoperative wound complications			
Seroma	34 (17%)	0 (0%)	.006
Hematoma	7 (3.4%)	0 (0%)	.25
SSI	25 (12%)	3 (7.9%)	.44
Cyst recurrence	29 (13%)	2 (4%)	.053

Abbreviations: BMI, body mass index; EBL, estimated blood loss; SSI, surgical site infection.

TABLE 4 Factors associated with drain placement.

	Drain placed ($n = 193$)	No drain ($n = 80$)	p value
Mean age at surgery, years (SD)	7.1 (4.5)	6.9 (4.3)	.72
BMI percentile (SD)	66.4 (30.3)	64.4 (28.8)	.65
Fistula or sinus tract present	37 (19%)	13 (17%)	.59
History of cyst infection	108 (57%)	31 (39%)	.008
Preoperative antibiotic treatment			.001
No	77 (40%)	48 (60%)	
1	60 (31%)	22 (28%)	
2	21 (11%)	9 (11%)	
3	16 (8%)	1 (1%)	
4	19 (10%)	0 (0%)	
Previous cyst excision	21 (11%)	1 (1%)	.008
Surgeon specialty			
Pediatric otolaryngology	191 (99%)	29 (36%)	<.001
Pediatric surgery	2 (1.0%)	51 (64%)	
Active infection at surgery	20 (10%)	10 (13%)	.58
Mean EBL, mL (SD)	7.8 (8.1)	5.7 (7.5)	.049
Intraoperative cyst rupture	56 (30%)	26 (33%)	.66
Inpatient admission status	152 (79%)	35 (44%)	<.001
Mean LOS, days (SD)	1.0 (1.0)	0.5 (0.5)	<.001
Discharge antibiotics prescribed	118 (61%)	15 (19%)	<.001
Postoperative wound complications			
Seroma	26 (15%)	8 (13%)	.71
Hematoma	7 (4%)	0 (0%)	.11
SSI	22 (12%)	6 (10%)	.56
Cyst recurrence	26 (14%)	5 (6%)	.09

Abbreviations: BMI, body mass index; EBL, estimated blood loss; LOS, length of stay; SSI, surgical site infection.

(odds ratio [OR] 1.97; 95% CI: 1.07–3.60, $p = .03$) was a significant risk factor for postoperative wound complications (Table 6).

4 | DISCUSSION

4.1 | Postoperative complications

This study was a large retrospective review of risks associated with wound complications and recurrence after the Sistrunk procedure in children. The Sistrunk procedure is the gold standard method of excision of TGDC in children and is reported to have the lowest rates of recurrence and few complications. Overall, our rates of recurrence and wound complications were similar to those previously reported.[11,13,18]

In a systematic review of 1172 adults and children, Gioacchini et al reported rates of recurrence after the Sistrunk ranging between 4% and 9%, with an average recurrence rate of 6%. The rate of minor complications was low (6.1%), and the most common complications were seroma, SSI, and wound dehiscence.[14]

In this study, the overall rate of postoperative wound complications was 25%. Our rate of complications is higher than that reported in the systemic review by Gioacchini, but falls within the range of complication rates of 3.03%–39.4% in literature specific to the Sistrunk procedure in pediatric patients.[19–25] The most common complications were seroma and SSIs, which is consistent with the literature.[16,21,23,26–29] Most importantly, we found that the history of infection significantly predicted wound complications.

The overall rate of cyst recurrence was 11% and it falls within the recurrence range of 0% to 16% in literature specific to the Sistrunk procedure in children.[2,8,12,13,16,19–21,24,27,30–32] Cyst recurrence was higher with a history of cyst infection but did not reach significance. The higher incidence of prior cyst infection may account for the higher rate of wound complications and recurrence seen in this study.

The association between preoperative cyst infection and complications including recurrence is likely due to increased difficulty in resection due to disruption of tissue planes, granulation tissue, and scarring. Our findings are similar to those of Ross et al. who found that preoperative infection was a significant predictor of both postoperative complications and cyst recurrence.[11] A history of more than

TABLE 5 Factors associated with wound complications.

	Wound complication	No complication	*p* value
Mean age at surgery, years (SD)	7.4 (4.8)	7.0 (4.3)	.50
BMI percentile (SD)	68.6 (31.4)	65 (29.4	.44
Fistula or sinus tract present	15 (24.2%)	35 (16.7%)	.18
Prior cyst infection	41 (66%)	98 (47%)	.008
Preoperative antibiotic treatment			.007
No	20 (32%)	105 (50%)	
1	26 (42%)	56 (27%)	
2	4 (7%)	26 (12%)	
3	8 (13%)	9 (4%)	
4	4 (7%)	15 (7%)	
Previous cyst excision	2 (3%)	20 (10%)	.112
Surgeon specialty			
Pediatric otolaryngology	52 (95%)	161 (76%)	<.001
Pediatric surgery	3 (5%)	50 (24%)	
Active infection at surgery	9 (15%)	21 (10%)	.29
Mean EBL, mL (SD)	8.1 (9.4)	7.0 (7.5)	.30
Intraoperative cyst rupture	22 (36%)	60 (29%)	.29
Inpatient admission status	51 (82%)	136 (65%)	.008
Mean LOS, days (SD)	1.1 (0.9)	0.8 (0.9)	.04
Discharge antibiotics prescribed	34 (55%)	99 (47%)	.27
Drain placement	50 (81%)	143 (68%)	.05
Cyst recurrence	26 (14%)	5 (6%)	.09

Abbreviations: BMI, body mass index; EBL, estimated blood loss; LOS, length of stay.

TABLE 6 Multivariable logistic regression model of wound issues after Sistrunk procedure.

Variable	OR	Std Err	*z*-test	*p* value	95% CI
Otolaryngologist	1.00	Reference	–	–	–
Pediatric surgeon	0.18	0.11	−2.78	0.005	0.05–0.60
Previous infection					
No	1.00	Reference			
Yes	1.97	0.61	2.20	0.03	1.07–3.60

Note: $N = 271$; pseudo $R^2 = 0.06$; Pearson X^2 goodness of fit $= 0.07$.
Abbreviations: CI, confidence interval; OR, odds ratio.

two episodes of infection, abscess formation, and cellulitis have been associated with cyst recurrence.[9,13] Revision operations have been associated with increased rates of postoperative infections.[33] No significant difference was observed between primary and revision sistrunk. Wound complications were, however, associated with cyst recurrence in this study, which is consistent with findings by Rhof et al.[18] It is possible that postoperative SSIs and seromas could be due to residual thyroglossal duct elements filling the surgical site with mucus rather than a true bacterial surgical infection. Of note, prescription of postoperative antibiotics did not significantly reduce rates of wound complications in this study.

This study also found significant differences in practice patterns between pediatric surgeons and pediatric otolaryngologists. Pediatric otolaryngologists were more likely to place a drain and admit postoperatively. The decision to admit at our institution, during the study period, was primarily based on surgeon preference. No difference was observed in rates of recurrence between specialties. However, pediatric otolaryngologists had significantly more wound complications in this study. Pediatric otolaryngologists were significantly more likely to operate on patients with a history of cyst infection and previous cyst excision, which could explain the difference in wound complications and recurrence rates. The effect of surgeon specialty on Sistrunk outcomes in the literature is varied. Previous studies have reported a difference in rates of recurrence by specialty, with rates ranging between 4.0%–9.1% for pediatric otolaryngologists and 27.3%–30.1% for pediatric surgeons.[11,23] Other studies have found no difference in rates of complications or recurrence by surgeon specialty.[21]

Drains are often used to prevent complications after surgery, but recently their efficacy has been debated. In this study, drain placement was not associated with a reduction in wound complications or

cyst recurrence, which is consistent with the current literature.[16,21,22,34] Drains were almost exclusively placed by pediatric otolaryngologists, and patients were more likely to be admitted postoperatively if a drain was placed. Our findings are consistent with a retrospective review by Pool et al. that reported that drain placement was significantly associated with surgery by pediatric otolaryngologists and overnight observation, and was not associated with lower rates of seroma, infection, or recurrence.[21] In this study, pediatric surgeons did not routinely use drains but had similar postoperative outcomes as pediatric otolaryngologists who commonly used drains. Thus, routine drain placement does not appear to reduce postoperative complications or recurrence rates.

The observational and retrospective nature of this study is an important limitation. Record-keeping was inconsistent in terms of clinical detail at patient presentation. Variation in surgical technique could impact postoperative complications, but we were unable to further classify patients by type of Sistrunk procedure performed (traditional vs. modified) given our small sample size and inconsistent documentation present in the operative reports. Follow-up period was difficult to assess as most patients were seen 2–3 weeks after surgery for their postoperative visit and subsequently discharged from the clinic if there were no concerns. Delayed complications and recurrence could be underestimated. Several patients had no follow-up after surgery, and patients may have sought care for complications or recurrence elsewhere which would result in an underestimation of results and compromise the internal validity of our study.

5 | CONCLUSIONS

The Sistrunk procedure is the standard method of excision for TGDCs and has historically been associated with lower rates of recurrence when compared with simple excision. Wound complications were significantly associated with a prior history of cyst infections. Drain placement and postoperative antibiotics did not change wound complication rates after the Sistrunk procedure. Thus, surgeons should reconsider routine placement of drains and prescription of antibiotics postoperatively. These findings should assist clinicians in preoperative family counseling and surgical planning for pediatric patients undergoing the Sistrunk procedure.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

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