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Esophageal stricture as a complication in post operative neonates with congenital esophageal atresia with tracheoesophageal fistula

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ABSTRACT

Introduction: Esophageal Atresia (EA) with Tracheoesophageal fistula (TOF) is one of the commonest congenital anomalies encountered in the pediatric surgery with commonest postoperative complication as esophageal stricture (ES) and gastroesophageal reflux disease (GERD). The aim of this study was to identify factors associated with ES formation in EA with TOF during post operative period.

Materials and Methods: This cross-sectional study was carried out in pediatrics surgery department. A total 30 neonates with EA with TOF who were diagnosed clinically and radiologically were included while pure atresia cases were excluded. The distance between upper and lower esophageal pouch was noted with ruler intraoperatively. Post operative soluble contrast dye study was done to detect leaks. If leak was not present then ratio of upper and lower esophageal pouch width (UPW: LPW) was measured. During follow up, ES and GERD were diagnosed clinically and radiologically**.** The data was analyzed using MS-Excel and SPSS software and p value < 0.5 was taken as significant.

Results: The measurement of gap between upper & lower pouch was < 2cms and >2cms in 12(40%) and 18(60%) patients respectively. The ES was found in 9(30%), GERD in 1(3.3%) and both ES and GERD in 6(20%) patients. The ratio of UPW: LPW found to be more than 1.5, equal to 1.5 and less than 1.5 in 12,2 and 1 patient respectively with ES. Dilatation for ES was required in all those with UPW: LPW >1.5 which was statistically significant. The postoperative UPW:LPW ratio showed statistically significant correlation with the intraoperative gap between upper & lower pouch and anastomosis under tension.

Conclusion: The ES had significant correlation with gap between upper & lower esophageal pouch more than 2 cm and anastomosis under tension. The ratio of UPW: LPW ≥1.5 needed dilatation for ES during follow up.

Keywords: *Esophageal Atresia, Esophageal stricture, Neonate, Tracheoesophageal fistula*

1. INTRODUCTION

Esophageal Atresia (EA) with Tracheoesophageal fistula (TOF) is the commonest congenital anomalies encountered in pediatric surgery practice. It is characterized by fistulous connection between esophagus and trachea along with atretic esophagus. The major advances in neonatal and surgical care led to more than 95% survival for many neonates and [1] majority of today's neonates will reach into adulthood [2,3]. Also, the burden of mortality should not be on EA or TOF alone, but also on its associated findings such as chromosomal anomalies, cardiac anomalies with or without extremely low birth weight (ELBW) [4,5].

A significant number of complications can occur despite excellent long-term survival and the commonest one is esophageal stricture (ES) defined as presence of symptoms such as dysphagia, recurrent respiratory problems and narrowing of esophagus on endoscopy or contrast esophagography. ES formation depends on various factors such as tension during anastomosis, number of layers in anastomosis, suture material, gap, gastro-esophageal reflux disease (GERD) and anastomotic leak. It has been found in various studies that in infants who have been operated for EA or TOF and who have history of previous anastomotic leakage, recurrent respiratory infections or feeding problems should be investigated for ES [2]. Clinically significant stricture can be treated by dilatation either via antegrade or retrograde bougienage. Many strictures respond to one to three dilatations, but many strictures resistant to repeated dilations may require resection and re-anastomosis, esophageal replacement, or even anti-reflux procedure, if associated with GERD [6].

At our institute, patients operated for EA or TOF were followed up with contrast esophagograms when indicated and esophageal dilatation was done accordingly. Considering the above factors responsible for stricture formation, this study was conducted with the aim to identify factors associated with ES formation in operated cases of EA with TOF during post operative period at our institute.

2. MATERIALS AND METHODS

This cross-sectional study was conducted in pediatrics surgery departments of the tertiary care hospitals from 31st January 2018 to 1st January 2022 after approval from the Institutional Ethics Committee. A total of 30 neonates with EA with TOF who have been operated were included in our study after obtaining written informed consent from parents / guardians. Neonates with EA (without TOF) were excluded. All these neonates were followed up for the period of one year.

The clinical diagnosis of EA with TOF was done by red rubber tube test by two consultants who examined the patient independently. The radiological investigations like X- ray chest with red rubber tube was done to confirm EA and presence or absence of gastric or bowel gas and was considered to exclude pure atresia (Figure1). The distance between upper and lower esophageal pouch was noted with ruler on operation table. The special consideration was given to cases who had undergone Lividiti's myotomy or anastomosis under tension. Post operative soluble contrast dye study was done and presence or absence of leaks was noted, and if no leak, then discrepancy in diameter between upper and lower esophageal pouch was noted as a ratio between width of upper and lower pouch i.e. UPW: LPW (Figure 2).

During follow up, proper history taking and examination was done and if needed, radiological investigation such as barium meal was done to see stricture and GERD. If symptomatic stricture was present, then need of esophageal dilatation was considered and size of dilator was noted which would just fit without bleeding. If evidence of GERD present, then number of patients responding to fundoplication was considered. The follow-up was done at 1 week, 1 month, 3 months, 6 months and at 1 year of age.

In every follow-up, the points noted were a) Weight and height measurements b) Any history of vomiting after feeds, and if present, then time interval between feeds and vomiting was noted. C) In case of multiple episodes of vomiting, contrast study was done to note the ratio of width of upper pouch to that of lower pouch and to look for the evidence of GERD. d) If necessary, the doppler study was done to confirm the presence of GERD. All the findings were confirmed by at least one of the consultants who initially assessed the patient on presentation.

The data was analyzed using MS-Excel and SPSS statistical software. Appropriate descriptive statistics were applied to data like frequency, mean with standard deviation and p value calculated by Chi-square test where p value < 0.5 was significant.

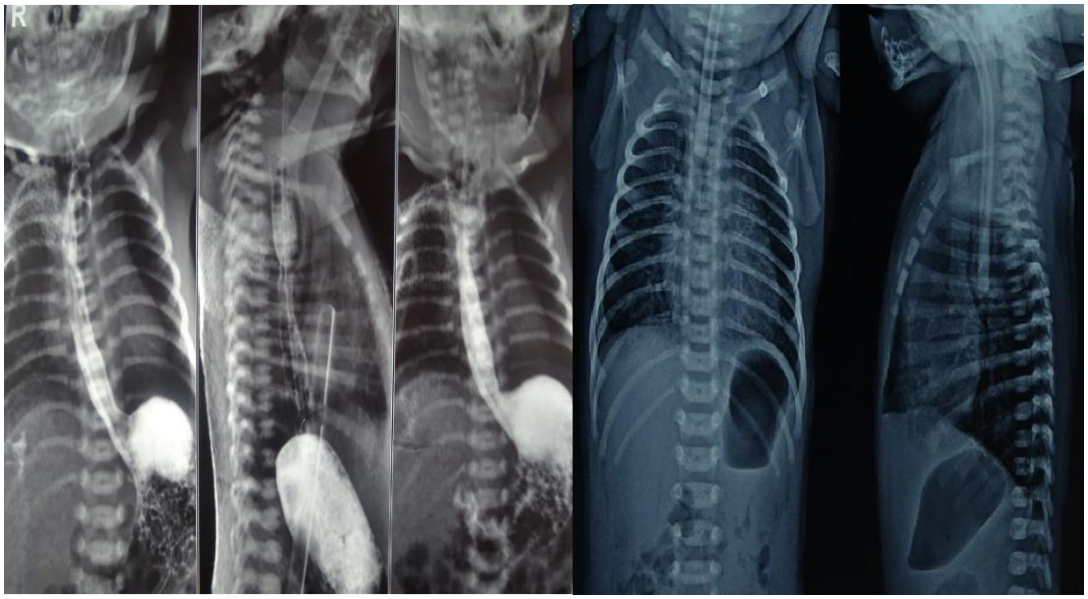

Figure 1: The diagnosis of esophageal atresia with trachea-esophageal fistula.

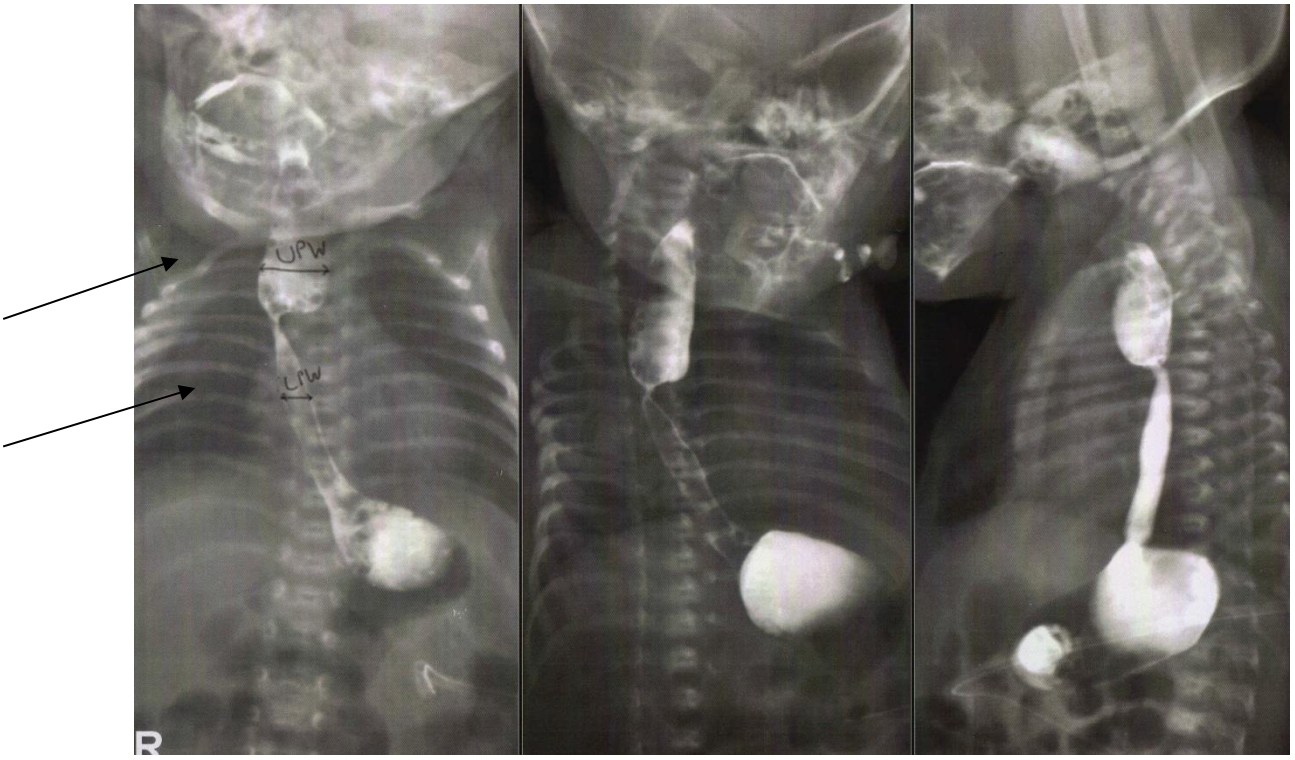

Figure 2: A ratio between width of upper and lower esophageal pouch i.e. UPW: LPW (UPW: upper pouch width, LPW: Lower pouch width)

3. RESULTS

This study was conducted on 37 patients of EA with TOF, who were operated at our institute. Out of these, 5 patients died during the postoperative period due to associated cardiac anomalies & 2 patients were lost follow up. Hence, out of 37 patients, 30 were included in the present study and followed up till 1 year of age. The postoperative outcome and complications were studied and observed.

Out of 30 patients ,7(23%) and 23(77%) presented in first 24 hours and after 24 hours respectively while17(56.66%) were females and 13(33.34%) were males. Thirteen patients (43.33%) were diagnosed antenatally with common finding of

polyhydramnios and 7 of these presented within 24 hours after birth and 6 presented within 2 days after birth. Twenty five (83.33%) patients presented with frothing from mouth and 4 of them also had respiratory distress due to late presentation at age of 4 days. But all these patients had history of non-passage of orogastric tube beyond certain distance (range 8-10 cms) from upper lip. Findings in all these patients were consistent with blind ending proximal pouch and distal fistulous communication suggestive of 'Type- C' fistula. The distribution of patients according to various associated congenital anomalies were shown in Table 1.

Congenital anomalies	Number(N)	Percentage (%)
Anorectal malformations	4	33
Cardiovascular system	6	50
Duodenal atresia	1	8
Down's syndrome	1	8
Total	12	100

Table 1- Distribution of patients according to various associated congenital anomalies

Eight (26.66%) patients were posted on the same day of presentation, 5(16.67%) patients were posted on the next day of presentation, and 17 (56.67%) patients were posted with gap of 1 day in between. The 8 patients who were posted on the same day of presentation and 5 patients, who were posted on the next day of presentation were antenatally diagnosed cases. The measurement of gap between upper & lower pouch was < 2cms in 12(40%) patients & >2cms in 18(60%) patients. Lividiti's myotomy was done in total 3(10%) patients. All these 3 patients had gap length of more than 2.5cms. The anastomosis was done under tension in 18(60%) patients and without tension in 12(40%) patients. All these 18 with anastomosis under tension had gap length of more than or equal to 2cms and those without tension had gap length less than 2cms.

Postoperative leak occurred in 5(16.66%) patients. Feeding Jejunostomy was done in all these patients and leak stopped spontaneously with time but stricture developed in 4 of them as diagnosed on follow up. All patients showed a gap between upper and lower pouch of more than or equal to 2cms. In 2 out of 5 patients, Lividiti's myotomy was done. Fourteen (46.66%) had symptoms on follow up after a period of 3-6 months. The distribution of patients according to types of symptoms on follow up was shown in Figure 3.

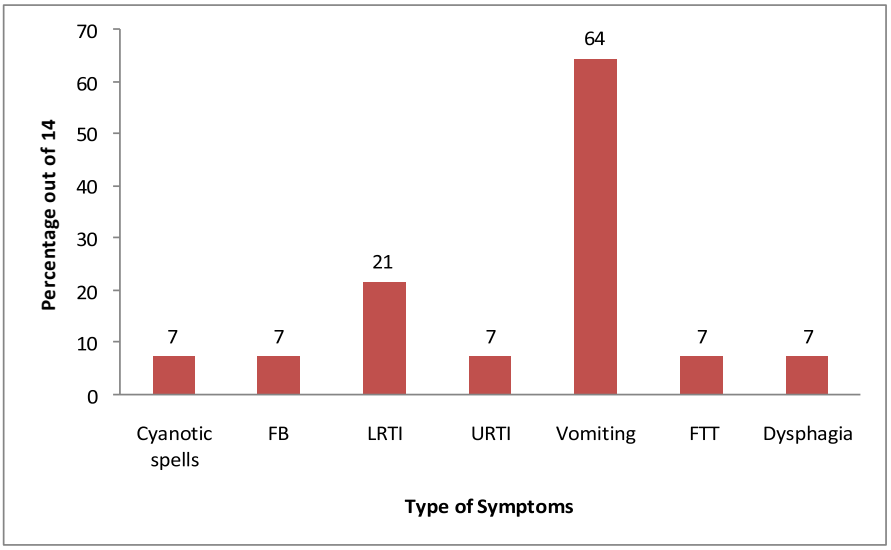

Figure 3: Distribution of patients according to types of symptoms on follow up

Sixteen patients (53.33%) were clinically evaluated and were advised dye study for the complications like ES and/or GERD. The ES was found in 9(30%), GERD in 1(3.3%) and both Stricture and GERD in 6(20%) patients. Out of 15 esophageal stricture patient dilatation was required in 13 patients only. The number of dilatations varied such as single dilatation was done in 7 (out of 13), 2 times in 5 patients and 3 times in 1 patient.

The novel parameter studied in our study based on ratio of UPW: LPW on dye study found to be more than 1.5 in 12, equal to 1.5 in 2 and less than 1.5 in 1 patient out of 15 esophageal stricture patients. Dilatation was required in all those with UPW: LPW >1.5 and in 1 (out of 2) with ratio equal to 1.5. Almost all patients (12/14) with UPW: LPW >1.5 presented with symptoms. There was statistically significant association between UPW: LPW ratio and dilatation. (p=0.004). The postoperative UPW: LPW was correlated with the intraoperative gap between upper & lower pouch. The Pearson's correlation coefficient was 0.41 which showed a positive correlation. It means that as the gap increases, the postoperative UPW: LPW ratio will increase. Out of 15 cases of stricture, 6 had GERD on dye study of which 1 responded to medical management & dilatation. However, 5(83%) patients had persistent symptoms and needed fundoplication. In 1(17%) patient with stricture, although GERD was not documented on dye study, fundoplication was done on clinical suspicion. The patient had good outcome after the surgery. The occurrence of esophageal stricture as a postoperative complication in patients of EA with TOF was compared with all the clinical and surgical parameters studied to see for any statistical significance (Table :2)

Sr. No.	**Parameters**	**p-value**	**Statistical significance (Yes/No)**
1.	Age	0.19	No
2.	Gender	0.27	No
3.	Gap	0.025	Yes
4.	Anastomosis under tension	0.025	Yes
5.	Myotomy	0.55	No
6.	Postoperative leak	0.14	No

Table2: Association of esophageal stricture with clinical and surgical parameters

4. DISCUSSION

EA with TOF is the commonest congenital surgical anomaly encountered in pediatric clinical and surgical practice. A significant number of complications can occur despite excellent long-term survival for infants with EA with TOF and commonest one are ES and GERD. In this study, 30 operated cases of EA with TOF were followed up post-operatively, keeping in mind proneness of these patients to various complications specially ES.

In the current study, out of 30, 7 patients (23%) presented within 24 hours after birth and 23 patients (77%) presented after 24 hours and it was found that those who presented after 24 hours of birth had complication like aspiration pneumonitis. The patients were referred to us mainly from rural areas & home deliveries, where there is lack of awareness and diagnostic facilities. So, this might be reason behind majority of delayed presentations in our study. Twenty-five patients presented with frothing from mouth while four of them also had respiratory distress due to late presentation at age of 4 days. Harmon C M et al [(7)] observed the usual clinical sign was excessive salivation followed by respiratory distress and inability of passage of orogastric or nasogastric tube. Findings in all the patients in present study were consistent with blind ending proximal pouch and distal fistulous communication suggestive of 'Type- C' fistula and these findings were confirmed on thoracotomy. On the other hand, in Losty P et al [(8)] found Type- C variety constitutes 85%.

In the present study, intraoperative measurement of gap length was > 2cms in 30% & less than or equal to 2 in 70% patients. However, study done by Thakkar H et al [(9)], gap length was >2cms in 15% and less than or equal to 2cms in 85%. The circular myotomy was done in 3 patients with long gap (37.5%). Similarly, in a study by Amit Singh et al [(10)] for sharing their experience regarding Lividiti's circular myotomy, found that myotomy was required in 35% of the cases with long gap. The anastomosis was done under tension in 18 out of 30 patients (60%) and without tension in 40% of patients. All these 18 with anastomosis under tension had gap length of more than or equal to 2cms and those without tension had gap length less than 2cms. On the contrary, in a study done by Thakkar H et al [(9)], anastomosis under tension was found in those with gap >3cms.The postoperative anastomotic leak was found in 16% (5/30) which was almost equivalent to the leak rate of 11.5% found in the study of Friedmacher et al [(2)], but in another study done by Charles Wynn Acher et al [(11)] for long term outcomes, 26% patients had leak. In search of association of gap between upper & lower pouch and anastomotic leak, all of those who developed leak in postoperative period, had gap length of more than or equal to 2 cms. Out of these, 80% (4/5) landed up with stricture. Similar result was found in another study done by Charles Wynn Acher et al [(11)] in which 74% of those with leak developed ES. However, in a study done by Thakkar H et al [(9)] for measurement of outcomes in relation to intraoperative gap length, leak occurred in only 5.5% of those with gap >2cms and stricture occurred in 50% of these. This

increased leak rate in our study may be due to small sample size in our study. Out of 3 patients with myotomy, 66% developed leak. In a study by Amit Singh et al [11], leak developed in 18% patients. The feeding jejunostomy (FJ) was done in all cases of anastomotic leak (5/5) and all these leaks (100%) closed spontaneously with stricture in 4 of them, whereas in a study done by Bawa M et al [12] for knowing the role of FJ in major anastomotic disruptions in EA, it was found that 60% of the patients were discharged successfully on FJ & there was healing of the anastomosis. This variation in outcome may be due small sample size in our study and larger sample in other.

ES was found in 15 (50%) patients, GER in 7(23.3%), both ES and GERD in 6(20%). I Friedmacher F et al [2] observed ES in 71.9% of symptomatic children & in another study done by Allin B et al [13] for outcomes at one year post anastomosis, stricture rate was 39%. In current study, GERD was found in 7 out of 30 patients (23%) & 6 of these (86%) of these patients had associated anastomotic stricture. In the study done by Okata Y et al [14], incidence of postoperative GER ranged from 27-75% . In a study done by Friedmacher F et al [2], GERD along-with anastomotic stricture was found in 85.5% and fundoplication was required in 44%.

In current study, esophageal dilatations were required in 13 of 15 patients who developed ES. Number of dilatations varied such as single dilatation was done in 7 (54%), 2 times in 5 patients (38%) and 3 times in 1 patient. Out of 7(with single dilatation), gap length was more than or equal to 2 cms in 6 of these patients (6/7). In 5 patients in which 2 times dilatation was done, gap length was <2cms in 2/5 and > /=2cms in 3/5. Mean number of dilatations done in our study were 1.7(approx.2), whereas in a study done by Davidson J et al [15] or their experience in pediatric esophageal dilatations, mean number of dilatations done were 3. In an article by Liveque D et al [16] an average 1-15 dilatation may be required in each patient with ES. Fifty percent of stricture will improve within 6 months, but 30% will persist and will require repeat dilatation. It was also observed in present study that total of 17 out of 30 patients (57%) had decreased growth rate, out of which 15 had stricture esophagus and 2 had associated cardiac anomalies.

In present study, Nissen's fundoplication was done in total of 6 patients (20%), in which GERD was documented in 83% (5/6) of patients and 1 patient had recalcitrant stricture. In a study done by Friedmacher F et al [2], GERD along-with anastomotic stricture was found in 85.5% and fundoplication was required in 44% of these. In our study, GERD patients were refractory to medical management, so the rate of fundoplication was high.

In our study, we studied the novel parameter based on ratio of UPW:LPW on dye study. The dilatation was needed in 86% (13/15) & mean number of dilatations done was 2. Also, out of 13 patients, 12 had UPW: LPW >1.5 and 1 had UPW: LPW equal to 1.5 and 2 patients who did not require dilatation, 1 of them had ratio <1.5. On contrary, in a study done by Leah Leibovitch L et al [17], stricture requiring dilatation was found in 47.8%. In another study done by Allin B et al [18], median number of dilatations required in the first year was 3. So, it can be derived that out of all symptomatic patients i.e. 14, stricture with UPW: LPW was > 1.5 in 86% i.e. 12/14 and all of them required dilatation. Although there is no description of parameter such as UPW: LPW in literature anywhere, but in a study done by Landisch RM et al [19] for utilization of stricture indices to predict dilatation, a parameter known as Upper pouch esophageal anastomotic stricture index (U-EASI) less than or equal to 0.39 on esophagogram is predictive of need of dilatation. The postoperative UPW: LPW was correlated with the intraoperative gap between upper & lower pouch. It means that as the gap increases, the postoperative UPW: LPW will increase. Stricture formation depends on various factors such as tension during anastomosis, number of layers in anastomosis, suture material, gap, GERD, and anastomotic leak.

5. CONCLUSION

The ES as a postoperative complication had significant correlation with gap between upper & lower pouch more than 2 cm and anastomosis under tension. The ratio of UPW: LPW on contrast study 1.5 needed dilatation for ES in follow up period and hence, holds promise for future.

REFERENCES

[1] Puri P. Esophageal atresia and tracheoesophageal fistula. In: *Newborn Surgery*.3rd ed. CRC Press; 2011:387.

[2] Friedmacher F, Kroneis B, Huber-Zeyringer A. Postoperative Complications and Functional Outcome after Esophageal Atresia Repair: Results from Longitudinal Single-Center Follow-Up. *J Gastrointest Surg*. 2017;21(6):927-35. doi:10.1007/s11605-017-3423-0

[3] Thompson A, Thakkar H, Khan H, Yardley IE. Not all neonates with oesophageal atresia and tracheoesophageal fistula are a surgical emergency. *J Pediatr Surg*. 2019;54(2):244-46. doi:10.1016/j. jpedsurg.2018.10.074

[4] Masuya R. Predictive factors affecting the prognosis and late complications of 73 consecutive cases of esophageal atresia at 2 centers. *Pediatric Surgery International*. https://doi.org/10.1007/s00383-018-4326-1

[5] Zani A, Wolinska J, Cobellis G, Chiu PP, Pierro A. Outcome of esophageal atresia/tracheoesophageal fistula in extremely low birth weight neonates (<1000 grams). *Pediatr Surg Int*. 2016;32(1):83-88. doi:10.1007/s00383-015-3816-7

[6] Coran AG, Adzick NS, Krummel TM, Laberge JM, Shamberger RC, Caldamone AA. Congenital anomalies of the Esophagus. In: *Pediatric Surgery*. Vol.2. 7th ed. 893.

[7] C Harmon C M, Arnold G. Direct aspiration of secretions into the trachea may occur with either a proximal or a distal tracheoesophageal fistula. Journal of Pediatric Surgery.1997;32(10):1401-1529

[8] Losty P D, Baillie Colin. Esophageal Atresia and trachea-esophageal fistula. In Newborn surgery, 2nd edition,2023:337-352

[9] Thakkar HS, Cooney J, Kumar N, Kiely E. Measured gap length and outcomes in oesophageal atresia. *J Pediatr Surg*. 2014;49(9):1343-46. doi:10.1016/j.jpedsurg.2014.03.021

[10] Singh A, Bajpai M, Bhatnagar V, Agarwala S, Srinivas M, Sharma N. Effect of number of associated anomalies on outcome in esophageal atresia with or without tracheoesophageal fistula patient. *Afr J Paediatr Surg*. 2013;10(4):320-322. doi:10.4103/0189-6725.125430

[11] Acher CW, Ostlie DJ, Leys CM, Struckmeyer S, Parker M, Nichol PF. Long-Term Outcomes of Patients with Tracheoesophageal Fistula/Esophageal Atresia: Survey Results from Tracheoesophageal Fistula/Esophageal Atresia Online Communities. *Eur J Pediatr Surg.* 2016;26(6):476-480. doi:10.1055/s-0035-1570103

[12] Bawa M, Menon P and K. L. N. Rao. Role of feeding jejunostomy in major anastomotic disruptions in esophageal atresia: A pilot study. *Journal of Indian Association of Pediatric Surgeons.*

[13] Allin B, Knight M, Johnson P, Burge D; BAPS-CASS. Outcomes at one-year post anastomosis from a national cohort of infants with esophageal atresia. *PLOS One*. 2014;9(8):e106149. Published 2014 Aug 25. doi:10.1371/ journal. pone. 0106149

[14] Okata Y, Maeda K, Bitoh Y, et al. Evaluation of the intraoperative risk factors for esophageal anastomotic complications after primary repair of esophageal atresia with tracheoesophageal fistula. *Pediatr Surg Int*. 2016;32(9):869-73. doi:10.1007/s00383-016-3931-0

[15] Davidson JR et al. Pediatric Esophageal Dilatations: A Cross-Speciality Experience. *Journal of Laparoendoscopic & Advanced Surgical Techniques.* DOI: 10.1089/lap.2019.0592

[16] D. Lévesque, R. Baird, J.-M. Laberge; Refractory strictures post-esophageal atresia repair: what are the alternatives? *Diseases of the Esophagus*, Volume 26, Issue 4, 1 May 2013, Pages 382–387, https://doi.org/10.1111/dote.12047

[17] Leibovitch L, Zohar I, Maayan-Mazger A, Mazkereth R, Strauss T, Bilik R. Infants Born with Esophageal Atresia with or without Tracheo-Esophageal Fistula: Short- and Long-Term Outcomes. *Isr Med Assoc J*. 2018;20(3):161-166.

[18] Allin B, Knight M, Johnson P, Burge D; BAPS-CASS. Outcome at one-year post anastomosis from a national cohort of infants with esophageal atresia. *PLOS One*. 2014;9(8): e106149. Published 2014 Aug 25. doi:10.1371/ journal. pone. 0106149

[19] Landisch RM, Foster S, Gregg D, et al. Utilizing stricture indices to predict dilation of strictures after esophageal atresia repair. *J Surg Res*. 2017; 216:172-78. doi:10.1016/ j.jss.2017.04.024