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Original Article

Bronchoscopic Management of Recurrent Tracheoesophageal Fistula in Children

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Abstract

Objectives: To evaluate the results of bronchoscopic cauterisation of fistulous tract using bugbee electrode with or without injection of fibrin glue, for recurrent tracheoesophageal fistula (RTEF) in paediatric age group.

Methods: Retrospective analysis of patients of RTEF from 2004 to 2016 undergoing bronchoscopic management was performed. Four patients were identified and they were evaluated with respect to resolution of fistula, and their long-term outcomes.

Conclusion: Bronchoscopic cauterisation of fistula with bugbee diathermy along with application of fibrin glue if performed carefully can resolve RTEF. Author emphasises use of double lumen catheter for injection of fibrin glue and apnea during injection of glue as an important step to be strictly observed while performing bronchoscopic management of RTEF.

Keywords: Recurrent, TEF, Fibrin Glue, Bugbee diathermy

Introduction

Oesophageal atresia and TEF has an overall incidence of 1 in 3000 to 4500 live births. It is one of the most challenging congenital anomalies because of its high morbidity and mortality [1]. RTEF occurs in 5%-15% of patients following oesophageal atresia repair [2]. Re-thoracotomy is technically challenging, and it is associated with significant morbidity, including a re-fistulation rate of 10%-22% [2]. As per literature Endoscopic occlusion of the RTEF with tissue adhesives (fibrin glue, Histoacryl) is reported to be safe and highly effective [2]. Till date only 99 such cases of endoscopic attempts to close RTEF in children are published. Of these, majority was approached from oesophageal side and only few cases of bronchoscopic management have been described.

Materials and Methods

We did a retrospective analysis of all the patients having RTEF after initial repair of congenital type of TEF. From 2004 to 2016 four such patients were identified. Out of these 3 were females and 1 patient was male. These patients were subjected to evaluation by bronchoscopy along with definitive management in the form of diathermy of fistula by monopolar electrode (bugbee) and then injection of fibrin glue. These patients were then evaluated in the immediate postoperative period and then they were on long term follow up for their outcomes (Table 1).

Technique

All the patients after the repair of TEF had either persistent or recurrent lower respiratory tract infections. They underwent water soluble contrast oesophagogram, which confirmed presence of RTEF(Figure1). Then these patients were subjected to the therapeutic bronchoscopy. All procedures were done under general anaesthesia with patients paralysed. An 8.5 Fr integrated cystoscope was used in infants and larger size 13 Fr cystoscope was used in a five-year-old patient. The cystoscope was introduced into the trachea under guidance of laryngoscope. These patients were ventilated through the side channel of bronchscope (Figure 2) Bronchoscopy showed a presence of fistula just proximal to carina in three patients and at a distance of 2 cm from carina in one patient (Figure 3). Then the bugbee electrode was placed through instrument channel of the cystoscope and tip of the bug bee was just introduced into the fistula. The monoploar diathermy at lower

setting of 5-15 was applied and the mucosal lining of the RTEF was cauterized (Figure 4). Then the bugbee was withdrawn along with the scope. The double lumen catheter, which was to be used for injection of glue, was primed and then it was introduced into the trachea. Then by the side of this catheter scope was introduced under the guidance of direct laryngoscopy. The tip of this double lumen catheter was carefully manipulated into the opening of RTEF on tracheal side (Figure 5). Anaesthetists were informed to give apnea and 0.2 to 0.4 ml of glue was injected. After injecting it takes approximately 4 seconds for fibrin glue to solidify. The scope was then negotiated beyond RTEF and gentle ventilation was carried out for few minutes. The scope was finally withdrawn.

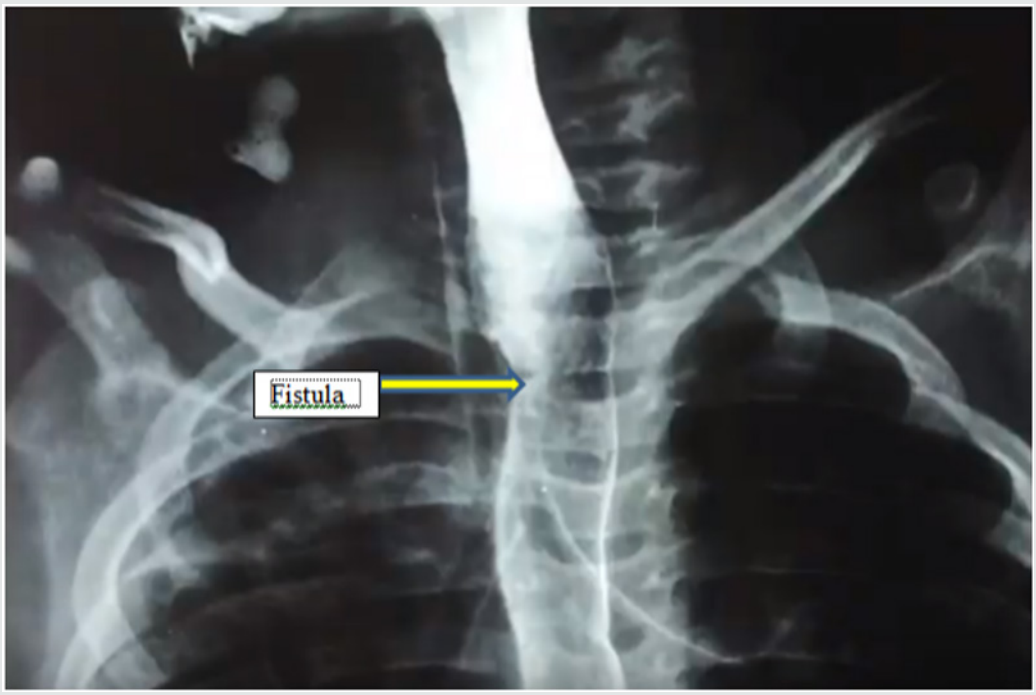

Figure 1.

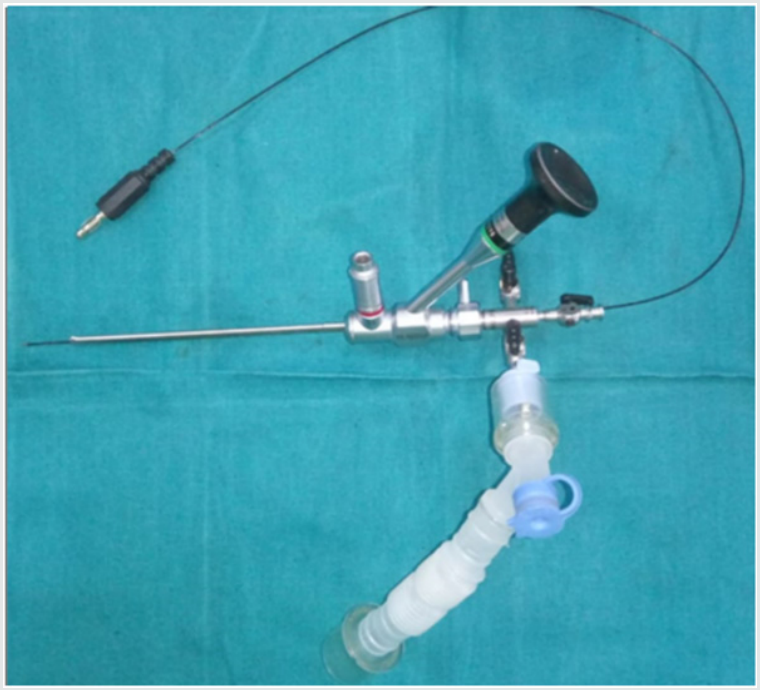

Figure 2.

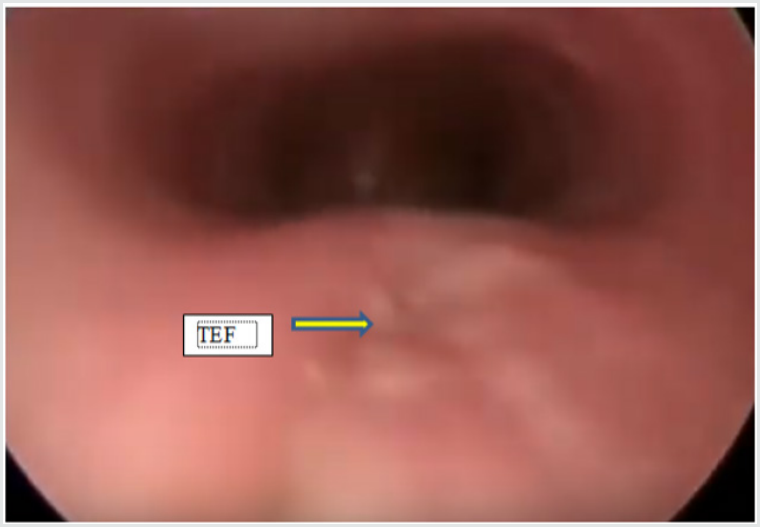

Figure 3.

Figure 4.

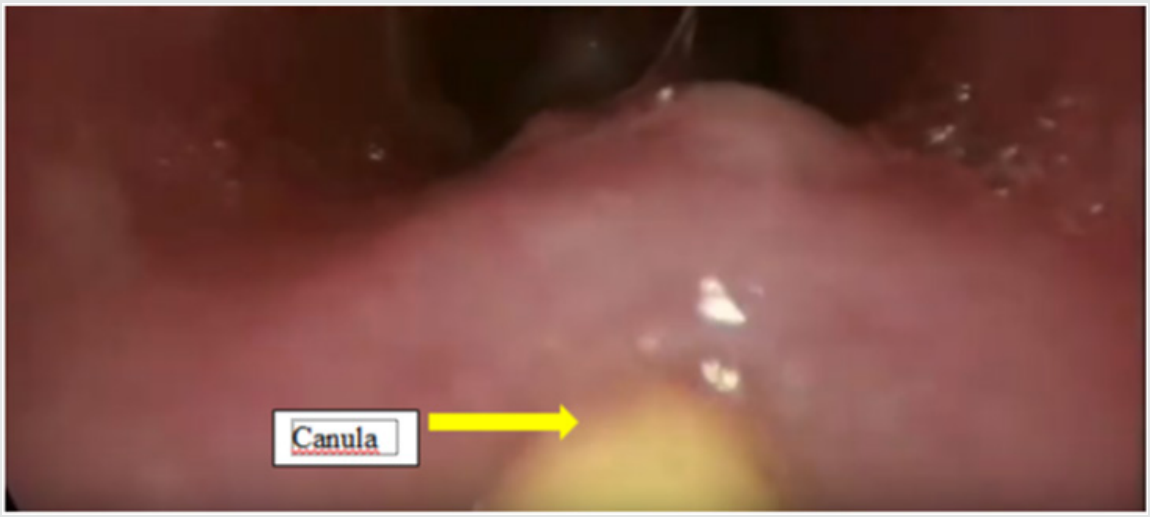

Figure 5.

Post procedure all patients were fed through a nasogastric tube for first two days. Patients underwent contrast swallow study to evaluate status of RTEF. If RTEF was resolved, then the oral feeding was resumed and patients were discharged. All patients received per-operative antibiotics along with proton pump inhibitors.

A. Case 1: A 6 months old female child, who was operated at day 2 of life for congenital TEF with open thoracotomy and ligation of fistula with primary anastomosis, presented with complaints of recurrent lower respiratory tract infection (LRTI) with failure to gain weight. A contrast swallow study was carried out which confirmed a RTEF. She underwent a bronchoscopic evaluation of fistula with only cauterisation of same. Nasogastric tube was kept for two days and feeding was continued through it. Then a dye study was done, which did not demonstrate any fistula and she was started on oral feeds. Patient was clinically alright till fifth day postoperatively when she developed LRTI. A repeat dye study at this time showed a persistent fistula and was advised thoracotomy and ligation of same. But the parents did not agree for same and took discharge against medical advice. She was then lost to follow up since then.

B. Case 2: A two-month-old female child was admitted for pneumonia. She had undergone repair of congenital TEF at day 1 of life. Since 20 days after surgery child had been re-admitted for severe LRTI and then transferred to our institute. On investigating she was found to have a recurrent TEF on contrast study. She was then subjected to bronchoscopic evaluation along with cauterisation and fibrin glue injection by above technique. But only single lumen catheter was used; also, the ventilation was not withheld during injection. So, the proper injection was not achieved. In the postoperative period after 2 days a repeat contrast study showed persistence of fistula. She then underwent a laparoscopic fundoplication with a feeding gastrostomy to prevent reflux and for feeding. Seven days later a thoracotomy with ligation and division of fistula was done. After 5 days of this procedure contrast study showed resolution of fistula. Oral feeds were then started. Patient was on regular follow up, gaining weight and taking oral feeds adequately. At 6 years closure of persistent gastrocutaneous fistula was done.

C. Case 3: A 4months old female child was brought with complaints of failure to gain weight with recurrent LRTI. She was operated on day 2 of life for congenital TEF by thoracotomy and ligation of fistula with primary anastomosis. A contrast study confirmed a RTEF. She then underwent bronchoscopic procedure with diathermy cauterisation with glue application by the technique as mentioned. This time a double lumen cannula was used, and the ventilation was withheld during the procedure. A nasogastric tube was kept, and child fed through it for two days. Dye study after2 days showed no fistula. Patient was then started on oral diet. On follow up patient had symptoms of severe gastroesophageal reflux disease. She then underwent a laparoscopic fundoplication after which she had complete resolution of symptoms. She has been on regular follow up and has no recurrence on follow up even after 5 years.

D. Case 4: A 5-year-old boy presented with complaints of failure to gain weight and recurrent LRTI since 1 year age. He had undergone thoracotomy on day three of life for congenital TEF. On thorough evaluation; contrast swallow dye study demonstrated a recurrent TEF. He underwent bronchoscopy with a 13 French bronchoscope and diathermy cauterisation along with fibrin glue application as per the technique as mentioned earlier. Two days later dye study showed resolution and child was started on full feeds. He is on yearly follow up since then and has no recurrence even after 5 years (Table 1).

Table 1: Details of Patients of present series.

Age	Sex	Procedure	Results		Remarks
			Immediate post op	On follow up	
6 months	F	• Only diathermy. • No injection of glue.	Resolution of symptoms for five days and then re-appearance of symptoms	Lost to follow up	• Discharge against medical advise
2 months	F	• Diathermy plus injection of glue. • Single Lumen Catheter. • No apnea	Persistent of symptoms and recurrence confirmed on day 2	Resolution of symtoms after thoracotomy	• Laparoscopic Fundoplication with feeding gastrostomy. • After one week thoracotomy with repair of RTEF
4 months	F	• Diathermy. • Glue application. • Use of double Lumen Catheter. • Apnea while injection	Resolution	Symptoms free at 5 years follow up	• Laparoscopic fundoplication for severe GERD after management of RTEF
5 years	M	• Diathermy. • Glue application. • Use of double Lumen Catheter. • Apnea while injection	Resolution	Symptoms free at 5 years follow up	• None

Discussion

After primary repair of oesophageal atresia and TEF, reported incidence of RTEF is 5-15 % [1,2]. RTEF present with chronic cough, cough associated with ingestion of food which is also known as Ohno's sign [3], episodes of choking and /or cyanosis associated with ingestion of food, recurrent pneumonia, chronic lung disease and malnutrition should lead to diagnostic suspicion [1]. The diagnosis of RTEF can be confirmed by contrast swallow study or by HRCT (high resolution Computed tomorised scans). However, diagnostic bronchoscopy is the choice of investigation for diagnosis and it can be converted into therapeutic procedure with help of various techniques [1,2].

Although surgical re-explorations are the standard treatment of RTEF; Endoscopic therapy offers a safe and elegant definitive alternative. As per Richter et al and Tzifa et al some patients may require repeated attempts of endoscopic therapy for successful closure of RTEF [2]. Many techniques for endoscopic management of RTEF have been described in children, the details of which are illustrated in Table 2 and are self-explanatory [1,2,4].

As per R.K. Yellapu et al, glue and clipping are considered good for </= 1cm fistula but endoscopic glue therapy is not effective in post infectious fistula [3]. Richter et al reported in 2008, only 37 cases of RTEF treated endoscopically. The first case of endoscopic management was reported in 1970 by Leylailiyeki et al. They reported a case where endotracheal cuff adhesion occurred post usage of histoacrylate and glubran, which was injected from esophageal side. Also, cyanoacrylate has been associated with inflammation and tissue necrosis. These complications have not been reported with the use of fibrin glue.

Authors on review of English literature have found only 99 children with RTEF, who have been managed by endoscopic procedures till date (Table 2). Of these reported patients, majority have been benefitted with help of diathermy along with application of tissue adhesive glue. Majority of these patients were managed esophagoscopically. On review of literature very few cases of bronchoscopic management of RTEF have been reported. Authors have reported 4 patients who developed RTEF after primary repair TEF, which were managed bronchoscopically. Authors in first case had reported use of diathermy alone, which lead to recurrence. In second case, diathermy with injection of glue was carried out but anaesthetists were not instructed to give apnea and glue flew off. In second case, glue injection was carried out using single lumen catheter which further complicates the procedure as there is a possibility of glue to get solidified within the lumen of the catheter. Authors, learning from the experience of first two patients, improvised the procedure in the last two patients with use of a double lumen catheter, where the two chemicals get mixed at the tip of the catheter which is manipulated right at the mouth of fistula and glue after injection solidify within seconds as chemical are mixed. As patients were given apnea when the glue was injected, it stayed at the site of RTEF and solidified. These two patients did well in immediate postoperative period as well as on longer follow up of 5 years. Authors recommend use of a double lumen catheter and apnea during procedure as important steps in bronchoscopic management of RTEF.

Table 2: Literature review of endoscopic management of RTEF in Children.

Author	Year	Number of patients	Management	Obliteration of Fistula
Rangecroft et al.	1984	2	Depithelisation (fulguration) of mucosa of fistula with diathermy (after 5-6 attempts)	2
Al Samarai et al.	1987	1	cyanoacrylate with polidocanol via esophagoscopy	1
Vanderplas et al.	1993	1	Fibrin glue + 30%NACL + polidocanol submucosal(after 4 attempts)	1
Guiterrez et al.	1994	1	Fibrin glue	1
Wiseman et al.	1995	2	De-epithelisation with bronchial brush with fibrin glue(with 1-4 attempts)	2
Willets et al.	1998	22	Endoscopic treatment	22
Bhatnagar et al.	1999	3	2-electrocautery and 1- Nd-YAG laser(1-4 attempts)	3
Hoelzer et al.	1999	1	Fibrin glue	1
NG et al.	1999	1	De-epithelistion with Holmium laser with fibrin glue(3 attempts)	1
Lopes et al.	2003	1	Cyanoacylate Glue with polidocanol	1
Eugene Mc Gharen et al.	2004	1	Diathermy with Fibrin glue	1
Khurrana et al.	2004	6	Diathermy coagulation (1-3 attempts)	
Tzifa KT et al.	2005	10	Diathermy with cyanoacrylate glue	9/10
Linder	2006	4	De-epithelisation with silver nitratebead + glue with esophageal balloon to occlude airflow through fistula(2-5 attempts)	4

San Roman et al.	2006	7	De-epithelisation with diathermy in 4 pts with fibrin glue in all(1-3 attempts)	7
Meier et al.	2007	3	De-epithelisation with bronchial brush with fibrin glue(1-3) attempts	3
Keckler et al.	2008	1	De-epithelisation with plugging by biosynthetic mesh(surgiris) with fibrin glue	1
Guiterrez C et al.	2008	10	Diathermy with Fibrin glue	9/10
Richter et al.	2008	4	Diathermy with Fibrin Glue	4
Sung et al.	2008	3	Chemocauterisation with 50 % Trichloroacetic acid (2-3 attempts)	3
Jai hoon Yoon et al	2009	1	CyanoacrylateGlue	1
Farra jJ et al.	2010	1	Fibrin glue	1
Rakoczy et al.	2010	1	Potassium Titanyl phosphate laser	1
Hosseini et al.	2011	5	Diathermy with cyanoacrylate glue	5
Brigantti et al.	2011	5	De-epithelisation with Dextranomer polymer injected submucosally (2 attempts)	5
Van Neikerk	2012	1	Depithelisation withbrush + plugging with a porcine dermal biological mesh held in place with synthetic absorbable sealant (Durasea)	1
Bennata et al.	2014	1	Stent f/b endoclips	1
Maizlin II et al.	2016	1	Diathermy with Fibrin glue	1
Chun M D et al.	2017	1	Diathermy	1

In endoscopic management of RTEF, use of fibrin glue or cyanoacrylate glue along with diathermy have been mainly described in literature. Gutierrez C reported 10 children with RTEF, who were subjected to bronchoscopy with the use of diathermy and fibrin glue injection. In 9 out of 10 patients, authors could achieve the complete obliteration of RTEF. Of these 5 patients required single session while 4 patients needed second attempt [1]. Fibrin glue (also called fibrin sealant) is a surgical formulation used to create a fibrin clot for hemostasis or wound healing. It is made up of fibrinogen (lyophilised pooled human concentrate) and thrombin (bovine, which is reconstituted with calcium chloride. When these two chemicals are mixed at the site of application it glues them together. Thrombin is an enzyme and converts fibrinogen into fibrin monomers within seconds, giving rise to a three-dimensional gel. There are multiple formulations available in market and in some aprotinin, fibronectin and plasminogen is also added [5,6]. The authors propose bronchoscopic cauterisation along with injection of fibrin glue by the technique described, as the first option to treat RTEF. If RTEF is resolved endoscopically then it can reduce morbidity significantly and it can also reduce the period of hospitalisation.

Conclusion

Authors propose bronchoscopic use of monopolar bugbee electrode for cauterisation followed by injection of fibrin glue on tracheal side of RTEF as a safe and effective method for managing RTEF. Endoscopic management of RTEF can reduce morbidity of the disease significantly.

References

1. Upadhyaya VD, Gopal SC, Gangopadhyaya AN, Gupta DK, Sharma S, et al. (2007) Role of Fibrin Glue as a Sealant to Oesophageal Anastomosis in Cases of Congenital Esophageal Atresia with Tracheoesophageal Fistula. World Journal of Surgery (World J Surg) 31(12): 2412-2415.
2. Willets Willetts IE, Dudley NE, Tam PK (1998) Endoscopic treatment of recurrent tracheo-oesophageal fistulae: long-term results. Pediatr Surg Int 13(4): 256-258.
3. Yellapu RK, Gorthi JK, Kiranmayi Y, Sireesh I (2010) Endoscopic occlusion of idiopathic benign esophago-bronchial fistula Year. 56(4): 284-286.
4. Richter GT, Ryckman F, Brown RL, Rutter MJ (2007) Endoscopic management of recurrent tracheoesophageal fistula. World J Surg 31(12): 2412-2415.
5. Atrah HI (1994) Fibrin glue. BMJ 308(6934): 933-934.
6. Spotnitz WD (2010) Fibrin sealant: past, present, and future: a brief review. World Journal of Surgery 34(4): 632-634.

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