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Original article

Experience of oesophageal atresia management in a tertiary Bangladeshi hospital

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Summary. *Objectives: Background and aim:* This study has been conducted to determine the clinical presentations and surgical outcome of patients with OA admitted in a tertiary level hospital in Bangladesh. *Methods:* A prospective analysis was conducted for 32 patients with confirmed OA between July 2007 and June 2015. Of them surgical correction was done in 25 cases. The other 7, in whom surgery could not be done, were excluded from the study. *Results:* Of the 25 cases, that had been operated, 15 were full term and 52% were more than 48 hours old. Type-C OA with TOF was the commonest type (96%). 24 cases were operated through Right Thoracotomy. Of them 52% were done by Single Lung (left) ventilation. 'Division of the fistula' with 'end to end anastomosis' was done in all of them. Only 1 patient had a Gastric pull up surgery. Of the 25 cases, 16 survived (64%). In follow up all the patients were found thriving and showed no problem in swallowing. *Conclusion:* Our study which shows 64% survival is a great achievement for us but not satisfactory in international standard. However, the experience and encouragement that we are gathering, will guide us to achieve better result in near future. (www.actabiomedica.it)

Key words: oesophageal atresia, tracheo-oesophageal fistula, management of OA

Introduction

Oesophageal Atresia (OA) is not an uncommon problem in children. It is frequently associated with Tracheo-Oesophageal Fistula (TOF) and other congenital anomalies. As a fact it is one of the most complex birth defects dealt by the Paediatric surgeons. Its survival can be used as an index for the status of neonatal surgical care because of its highest mortality rate due to problems in respiratory care and surgical technique failure (1). OA correction is regarded as the symbol of neonatal surgical expertise, while evolution of OA treatment from simple ideas to staged and robotic operation has elapsed over 60 years (2-3). Still the overall survival rate remains poor except in some very developed countries. This advancement is possible due to the improvement of overall neonatal care by creating readily available modern Neonatal Intensive Care Units (NICU) and development of neonatal anaesthesia. Now a day's only the presence of associated major congenital anomalies determines the chance of survival (4). In our country, the incidence of OA is not low but as there is incomplete national data of the congenital diseases, the real picture is still unknown to us. Very few hospitals have so far published data on OA management due to its extreme poor result.

At the Square hospital, taking the advantages of its modern NICU and Anaesthesia facilities, we conducted this study to determine the clinical manifestations and surgical outcome of patients admitted with OA with TOF.

Materials and methods

It was a prospective study conducted between July 2007 to June 2015 in the Paediatric Surgery department of a tertiary level private hospital in Dhaka,

Bangladesh. The study protocol was approved by the 'Research Ethics Committee' of the hospital. Parents of the patents also gave informed consent prior to their inclusion in the study. In this period total 32 patients were admitted in the Neonatal Intensive Care Unit (NICU) of the hospital with features of OA. Diagnosis was suspected by the observing the common features of OA like history of maternal polyhydramnios, baby's excessive salivation, chocking on feeding, respiratory tract infection and failure to put an Oro-Gastric (O.G.) tube into the stomach. OA was confirmed by taking a chest x ray after putting a radio opaque marked Oro gastric tube in the oesophagus, which showed it's arrest in midline of upper chest. Associated TOF was confirmed by seeing presence of gas shadow in the abdomen in x ray. Echocardiography, bleeding and sepsis profiles were done in all cases. Relevant other investigations were also done in isolated conditions to diagnoses any particular associated anomalies. Among the 32 cases admitted, we could do surgery in 25 cases. In the rest 7, surgery could not be carried out due to associated severe cardiac anomalies, fatal sepsis or inability of the parents to bear the expenses of treatment. So, those 7 cases had been excluded from the study.

Results

Of the 25 patients who had been operated, most of them (52%) came late that is after 48 hours of birth as they were born outside (Table 1). This delay was due to late decision making from the attending doctors or the family. However, the duration of hospital stay of the patients varied from 2 days to 64 days (mean - 24 days).

Most of the children were mature in the present study. Only 10 of them were preterm (Table 2). The birth weight varied from 1700 gm to 3040 gm (mean-2170 gm) (Table 2).

Table 1. Age incidence at the time of admission: (n-25)

Time of Admission (in hours)	Number (%)
<24 hours	08 (32)
24 to 48 hours	04 (16)
>48 hours	13 (52)

Table 2. Maturity at birth and Birth weight of the patients: (n-25)

	Number (%)
Maturity	
Full term	15 (60)
Pre term	10 (40)
Birth Weight	
>2500 gm	11 (44)
2000 to 2500 gm	07 (28)
<2000 gm	07 (28)

In our series 24(96%) patients had 'Type-C' Gross Anatomical type of Oesophageal atresia with Trachea Oesophageal fistula (lower end). Only one baby (04%) had isolated Oesophageal atresia with no fistula (Gross' Type-A). Associated other congenital anomalies were present 65% cases among them Cardiac anomalies were commonest and were serious in many cases (Table 3). However, many patients of the series had more than 1 associated congenital abnormality. One of them had the VACTERL association. She had vaginal atresia in addition.

All the children who had OA with TOF were operated after a few days preparation in NICU which included controlling the dehydration, sepsis and nutrition. A team of neonatologist helped. Anaesthesia was given by a team of anaesthetist expert in paediatric anaesthesia. Single left lung Endotracheal tube ventilation was given in 52% cases which helped the surgeons to do surgery much comfortably as the right lung remained paralyzed. In the rest 48%, conventional both ventilation was given (Table 4). Right open thoracotomy by intercostals space (extra pleural) was the approach. The gap between the two ends was also variable. Most of our cases had 'short' or 'intermediate'

Table 3. Associated Congenital anomalies

Anomalies	Number (%)
Cardiac (ASD, PDA, MR, LR shunt etc)	14 (54)
Vertebral (Hemivertebra, Scoliosis)	06 (24)
Anorectal (ARM) (1 high, 1 low varieties)	02 (08)
Limb deformity	01 (04)
Genital (vaginal atresia)	01 (04)
Renal Agenesis (single)	01 (04)
Post. Urethral Valve (PUV)	01 (04)

Table 4. Types of Endotracheal tube ventilation for GA: (n-24)

Types of Ventilation	Number (%)
Single Left Lung	13 (52)
Both lungs	11 (48)

Table 5. Gap between two ends of Oesophagus and use of chest drains: (n-25)

	Number (%)
Type	
Short gap (<1 cm or 1 vertebral body)	13 (52)
Intermediate gap (1 to 3 cm)	11 (44)
Lon gap (>3cm)	01 (04)
Water seal Chest Tube	
Given in right thorax	18 (72)
No chest tube	07 (28)

Table 6. Early Post operative complications and Causes of death of the patients

	Number
Complications	
Pneumonia	05
Sepsis	08
Anastomotic leakage	03 (1 major, 2 minor)
Tracheomalacia	02
Ventilator dependency	01
	Number (%)
Causes of Death	
Sepsis	08 (72)
Major anastomotic leakage	01(14)
Anaesthetic complication	01 (14)

• Some patients had more than one early post operative complications.
• Some patients had more than one causes of death.

type of gaps (Table 5). The babies who had associated TOF had the gap of this nature. In all cases, division of the fistula and primary single layer end to end anastomosis could be done using 4/0 Monocryl. A Fr.10 Nasogastric tube was passed through the anastomosis in all cases as a stent and early feeding. They were kept for 10 days in all cases. But the patient with no TOF had a very long gap. As it could be diagnosed preoperatively, a life saving 'Cervical Oesophagostomy with feeding gastrostomy' was done without any thoracotomy. This child had an Oesophageal replacement surgery by 'Gastric pull up' elsewhere at the age of 1 year. Water seal Chest tube (right) was introduced in 18 (72%) cases and kept for 7 days in average (Table 5).

The associated anomalies were managed mostly conservatively. In high ARM a Sigmoid colostomy and then PSARP was done at 1 year of age. The low ARM was managed by Anoplasty. Cystoscopic fulguration of PUV was done in the same sitting of OA surgery. The vaginal atresia will corrected later.

Post operative care in a well equipped NICU was as important as the surgery. Among the 25 cases 17 (68%) needed ventilator support in some stages after surgery. However, all the 7 cases that died in the series needed ventilator support. Rest 5 patients needed no ventilation after surgery and all of them survived. There was limited post operative complications noted in our series among them sepsis was commonest early complication (32%) and was lethal in many cases (Table 6). The sepsis was due to pneumonia or nosocomial in nature.

Of the 25 cases operated, 16 (64%) survived and could go home. 9 patients died after surgery, one on the OT table and the rest in the NICU. The major cause of death was uncontrollable Sepsis (72%). (Table 6). The comparison between our survival and internationally acceptable survival is shown in Table 7.

We have tried to follow up the patients as longer as possible. However, all of them were thriving well. None had any problem in swallowing liquids or solids.

Table 7. Comparison between our survival and international survival (Waterston risk group)

Groups	Features	Accepted survival	Our survival
A.	Birth weight >2500 gm & otherwise healthy	100%	70% (7 of 10)
B.	Birth weight 2000-2500 gm & well or higher weight with moderate associated anomalies (non cardiac anomalies plus PDA, VSD, ASD)	85%	71% (5 of 7)
C.	Birth weight <2000 gm or higher with severe associated cardiac anomalies	65%	57% (4 of 7)

Follow up contrast Oesophagogram after 3 months showed good continuity of the oesophaguous with no functional stenosis in all cases. Domperidone and Ranitidine were advised orally for at least 3 years to prevent reflux and its related complication. The child, who needed gastric pull up, developed acute intestinal obstruction due to bands and adhesions after 6 months of surgery. He needed laparotomy and now doing fine.

Discussion

The first anatomic description of OA with TOF was reported by Thomas Gibson in 1697 (5). In 1943, Haight and Towsley reported the first survivor after a primary anastomosis (6). At that period that were a few survivors after surgery. Now a day the incidence of surviving has increased due to modernization of NICU and Anaesthesia. Still in our country various socio economic factors plays role in the result as most of the deliveries are carried at home and hence the diagnosis and transfer to a tertiary centers are too delayed. In our study most of the patient came after 48 hours of birth. During this period they did not get any supportive treatment which definitely had detrimental effect on the result. However Tandon and his co workers have did not recognize the age at the time of admission as a bad prognostic factor (1).

Prematurity and low birth weight in the patients of OA is problem as the physiology of the child does not develop properly and there is always increased chance of sepsis. However it is a global problem. Many authors consider birth weight <2500 gm as a high risk factor. But Spitz et al. does not regard weight as a contra indication for primary repair (7). In Tandon's series there survivor with lowest birth weight was 1800 gm (1) but in our series it was 1700 gm. The full maturity of that child might help him to survive. Fortuitously we had most of our patients in full term.

In most of the case in our study, the defect was Type C and was consistent to the Gross Anatomical Classification (Upper end blind and lower end fistula) as the commonest type (8). In about 65% of our cases had associated congenital anomalies which a little bit higher than other international studies like 60% in Hassab et al. (9), 59% in Saing et al. (10), 47% in Spitz et al. (7) and 52.4% in Rokitansky et al. studies (11). Cardiac anomalies were the commonest and a risk factor like others. Incidentally, we got a true VECTREL association (Vertebral, Anorectal, Cardiac, Tracheo-oesophageal, Renal and Limb defects) in our series. It had additional vaginal atresia. However, she survived after surgery .The ARM has also been corrected.

Preparation of the baby before going to surgery is always an important part of management. Most of these babies have aspiration pneumonia due to repeated vomiting which leads to sepsis. They also suffer from severe dehydration, malnutrition and disorders in bleeding profile. If these are not corrected meticulously before surgery, the result becomes very poor. The need for NICU is essential in this stage. On the other hand if the surgery is delayed for a long period there is chance of chemical pneumonitis due to reflux of gastric juice into the lungs through the TOF. It is very critical to decide the exact time of surgery to achieve the best result. The decision taken together by the surgeon, neonatologist and anaesthesits after repeated careful evaluation can solve the problem.

Role of neonatal anaesthetist with vast experience and knowledge in this field is another way to proceed for good result. Our anaesthetist could give Single left lung endo tracheal tube anaesthesia which almost paralyzed the right lung. As a result the surgeon could easily dissect and do anastomosis without damaging the right lung by retraction. We made the end to end anastomosis in single layer with monofilament 4/0 Monocril to present stricture. Trans anastomotic stent was given in oesophagus in all cases to provide early feeding. We found it safe and cost effective without any added complication. Moriarty et al. has also reported in our favor (12).On the other hand Tandon et al. did not find any extra benefit in terms of survival (1).

Post operative care in NICU is also an important part of management. In 12 cases in our series needed ventalatory support. The entire patients died in the series were in the ventilator. On the other hand those who did not need any ventilator at any stage survived uneventfully. Some centers routinely prefer to keep the child in ventilator for about 48 hours after surgery to support the damaged lungs. But we did not do this due to probable ventilator related complication and also for cost effectiveness.

Our overall survival in our series was 64%. The major cause of mortality was due to uncontrollable sepsis in post operative period. If we compare this result with internationally accepted results like Waterston risk group (13) it will look gloomy for Group A patients where there is 100% survival. But in case of Group B and C patients, our achievements are satisfactory. On the other hand if we compare our result with the other centers of our country it looks extra ordinary (personal observation).

Recently many advanced centres of the world have started thoracoscopic repair of Oesophageal atresia. It represents a natural evaluation in the operative correction of this complicated congenital anomaly and can be safely performed by experienced endoscopic surgeons. The results are comparable to the babies undergone repair through a thoracotomy (14). Thoracoscopic repair of OA can avoid the musculoskeletal problems following thoracotomy. So, they are considered as a better procedure. But none of our cases were done thoracoscopically due to lack of facilities.

To our knowledge no paediatric surgical centers of our country have ever published any data of their experience on OA with TOF probably due to very poor outcome. The other developing and under developed countries must have the similar experience.

Conclusion

Like other developing and under developed countries of the world, the overall result of OA with TOF surgery is still very poor in our country. Early diagnosis of the disease and associated other anomalies and referring them to the proper centre is needed for good result. But lack of a modern NICU in our country is the prime factor for poor result though other different socio economic conditions also contribute the ultimate result. Our study shows 64% survival is an extra ordinary achievement for us but not fully satisfactory in international standard. However, the experience and encouragement that we have gathered, will guide us to achieve better result in near future.

References

1. Tandon RK, Sharma S, Sinha SK, et al. Esophageal atresia: Factors influencing survival – Experience at an Indian tertiary centre. J Indian Assoc Pediatr Surg 2008; 13(1): 2-6.
2. Myers N. Esophageal atresia: The epitome of modern surgery. Ann R Coll Surg Engl 1974; 54: 277-87.
3. Ashcraft KW, Holder TM. The story of esophageal atresia and trachea-esophageal fiatula. Surgery 1969; 65: 332-40.
4. Spitz L, Keily EM, Morecroft JA, Drake DP. Oesophageal atresia: At-risk groups for the 1990s. J Pediatr Surg 1994; 29: 723-5.
5. Gibson T. The anatomy of human bodies epitomized. 6th ed. Awnsham and Churchill1697: London.
6. Haight C, Towsley HA. Congenital atresia of the esophagus with trachea esophageal fistula: Extra-pleural ligation of fistula and end to end anastomosis of esophageal segment. Surg Gynecol Obstet 1943; 76: 672-5.
7. Spitz L, Kiely E, Brereton RJ. Esophageal atresia: Five year experience with 148 cases. J Pediatr Surg1987; 22: 103-8.
8. Gross RE. The surgery of infancy and childhood. Philadelphia: Saunders1953: 76.
9. Hassab MH, Swilem MA, AL-Gobair F. Profile and outcome of esophageal atresia in a rapidly developing area. Kuwait Med J 2002; 34: 12-5.
10. Saing H,Mya GH,Cheng W. The involvement of two or more systems and the severity of associated anomalies significantly influence mortality of esophageal atresia. J Pediatr Surg 1998; 33: 1596-8.
11. Rokitansky A, Kolalkaya A, Bichler B, Mayr J, Menardi G. Analysis of 309 cases of esophageal atresia for associated congenital malformations. Am J Perinatol 1994; 11: 123-8.
12. Moriarty KP, Jacir NN, Harris BH,Lathaw LA, Robertson FA, Cromblehome TM. Transanastomotic feeding tube in repair of esophageal atresia. J Pediatr Surg1996; 31: 53-5.
13. Watreston DJ, Carter RE, Aberdeen E . Oseophageal atresia: Tracheo-oesophageal fistula: A study of survival in 218 infants. Lancet 1962; 819-22.
14. Holcomb GW, Rothenburg SS, Bax KMA et al. Thoracoscopic repair of esophageal atresia and tracheoesophageal fistula: A multi-institutional analysis. Annals Surgery 2005; 242(3): 422-30.

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