Management of Congenital Tracheosophageal Atrasia and Fistula: A preliminary Bi-Cenre Study in Nigeria.

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Background: ***Esophageal atresia and tracheosophageal fistula are congenital abnormalities of the esophagus. The incidence is about 1 in 2,400 to 4,500.It can occur in association with conditions of the VACTERL group. The diagnosis is confirmed by the failure of a nasogastric tube to get to the stomach. Surgery is the mainstay of treatment. There has been remarkable improvement in the outcome following surgery in developed countries. This cannot be said of low resource and developing countries where significant morbidities and mortalities are recorded. This paper highlights the early postoperative complications seen from two centers in Nigeria. It also raises questions that stimulate further research***
Methods: ***This was a retrospective study done in two tertiary teaching hospitals in Nigeria over a one year period (2012). It was a preliminary work to evaluate the differences in management of neonates with tracheosophageal fistula (TEF) from the two centers over the same period.***
Results: ***Two centres were compared.Centre A managed 4 patients and Center B managed 5 patients.Staged repair was done in centre A and primary repair in Centre B.***
Conclusion: ***The outcome of management using primary repair and staged repair were comparably the same.***

Keywords: Atresia, Fistula, Tracheoesophageal, Congenital

Introduction

Congenital tracheosophageal fistula (TEF) is one of the less common developmental anomalies but it has received considerable attention and is one of those anomalies which can be successfully treated by surgery[1].Esophageal atresia (EA) can occur alone or with an abnormal connection (TEF).The great majority of patients with this malformation have both EA/TEF[1].

TEF occurs in about one in every 3,000 live births. There are no figures of the incidence of this malformation in Nigeria yet. This condition is slightly more common in males than females[2]. Postnatal diagnosis of EA is confirmed by the failure to pass a firm nasogastric tube into the stomach. On chest x-ray the tube is seen curling in the upper esophageal pouch[2]. The surgical management of EA/TEF has evolved over the last 60 years associated with progressive improvement[3]. The survival rate of infants with TEF improved dramatically towards the end of the twentieth century. Corrective surgery for EA/TEF is well established and survival rate of over 90% can be expected[2].Jacqueline et al in their series reported a reduction in mortality from 61% (1947-1968) to 11% (1994-2000)[4].This however cannot be said for developing countries [2,4]. The outcome of management of EA/TEF in low resource countries is not encouraging[2].

Most infants with EA/TEF undergo repair in early infancy with division of the TEF and primary anastomosis[5].In many cases, anastomosis may be delayed with placement of a cervical esophagostomy and a feeding tube, until sufficient esophageal esophageal growth occurs to allow anastomosis. Neonatal esophageal stretching has been reported to allow primary anastomosis in infants with long gap[6]. Some risk categorization classifications are popular in determining the outcome of infants treated for EA/TEF.These include the Waterson as well as Spitz risk categorization[5]. The birth weight and the presence or absence of pneumonitis is important variables in determining the outcome. The authors in this series report the management of infants with EA/TEF in two Nigerian tertiary centers. The paper highlights the variation in approach to treatment and the early

postoperative challenges. This paper is a preliminary report as more work and research is still on-going in this area.

Patients and Methods

This study was a retrospective one done in two tertiary teaching hospitals in Nigeria over a one year period in 2012. It wass a preliminary work aimed at evaluating the differences in management of neonates with tracheosophageal fistula (TEF) from the two centers over the same period.The case notes of the patients were retrieved from the medical records department of the two centers and relevant data extracted. These included the name, sex age at presentation, birth weight results of investigations and the treatment offered. Offered..

The Irrua Specialist Teaching Hospital, Irrua was considered as Center A while University College Hospital, Ibadan was labeled Center B.

Center A had a total of 4 patients while Center B had 5 patients managed for TEF over the same period. Data was analyzed using simple statistical with SPSS version 10.

Results

In this study a total of 9 (nine) neonates with TEF from both center were analyzed. Center A had 4 patients while Center B had 5 patients. The M: F ratio in Center A was 3:1. The M: F ratio in Center B was 3:2. The ages at presentation of patients from Center A were 6 days, 2 days, 9 days and 6 days with an average age of 5.7days. The ages at presentation of patients from Center B were 2 days, 1 day, 2 days, 1 day and 3 days with an average age at presentation of 2 days.

Table 1. Presenting Symptoms

Symptoms	Center A	Center B
Regurgitation	4 (100%)	5 (100%)
Drooling of Saliva	4 (100%)	5 (100%)
Choking	2 (50%)	3 (60%)
Cyanosis	1 (25%)	2 (40%)
Abdominal distention	1 (25%)	2 (40%)
Dyspnea	NIL	1 (20%)

Table 2. Complications Following Surgery

Complications	Center A (Patients)	Center B (Patients)
Wound Infection	2	2
Sepsis	2	-
DIC	1	-
Retraction of Oesophagostomy	1	1
Dislodgement of Feeding Tube	2	-
Cardiac Arrest	-	2
Subcutaneous Emphysema	-	1

The birth weight of the patients from Center A were 2.8kg, 3.0kg, 2.6kg, and 3.5kg while that from Center B were 3.6kg, 2.6kg, 3.2 kg, 3.0kg and 4kg respectively.The neonates presented with the following symptoms as shown in Table 1. All the patients from both centers had chest and abdominal radiographs with a nasogastric in –situ. This showed curling of the tube in the proximal esophageal

pouch with failure of the tube to reach the stomach. There was gas shadow in the stomach in 1 (25%) patient from Center A and 2 (40%) from Center B.

Table 3. Mortality Table

Center	Patient	Weight	Age	Types of Surgery	Cause of Death
A	1	2.8Kg	6 Days	Feeding jejunostomy and venting gastrostomy	Sepsis
	2	2.6Kg	9 DAYS	Feeding jejunostomy and venting gastrostomy	DIC
B	1	3.6Kg	2 DAYS	Thoracotomy and anastomosis	Cardiac Arrest
	2	2.6Kg	1 DAY	Thoracotomy and anastomosis	Cardiac Arrest

Other investigations which included echocardiogram and abdominal ultrasound were done to detect any coexisting congenital abnormalities in all the patients. Hematologic investigations were also done for the neonates in both centers.There were no associated congenital anomalies noted.The blood picture were normal in both centres. All the 5 patients from Center A and 4 of the patients in Center B underwent surgery. The fifth patient in Center B died before surgical intervention. The patient had signs and symptoms of aspiration pneumonitis with associated respiratory difficulties.

All the neonates from Center A had feeding jejunostomy, venting gastrostomy and a cervical esophagostomy. The definitive surgery was done after individual assessment of the patients and when considered fit. Four of the patients from Center B had surgery, 3 (60%) had extra- pleural posterolateral thoracotomy with primary anastomosis of the esophagus.while the fourth patient had feeding gastrostomy, esophageal banding and cervical esophagostomy. The post-operative care in both centers recorded significant morbidities as shown in the Table 2.

Discussion

Congenital EA/TEF is common congenital anomalies affecting 1 in 2,400 to 4,500 individuals [5]. . A combination of history and radiological findings will usually assist in a quick diagnosis. The cardinal symptoms are regurgitation of feeds, drooling of saliva, choking and failure of a nasogastric tube to reach the stomach. These findings were observed in both groups of patients analyzed in this series .

The male to female ratio was roughly same for both groups, with a male preponderance in keeping with global findings[2,3,4].The average age of presentation from Center A and Center B were 5.7 days and 2 days respectively. The likelihood of aspiration pneumonitis is greater for older neonates, thus affecting outcome of treatment as seen in the only patient that was not operated in this series who eventually died before surgery.(There is no mention of pneumonia in the result and how did it affect your outcome?.

The average weight of the patients in Center A was 2.9kg and 2.7 kg for Center B which was comparable. Weight and pulmonary function was used by Waterston[7] in categorizing patients into various risk groups. The weight of the patients from both groups in the absence of pulmonary problems was classified as Waterston A. EA/TEF was confirmed in all the patients using radiographs

with plain nasogastric tube in place showing curling of the tube. Radiopaque tubes are preferred, though not commonly available in many centers in Nigeria. Endoscopy is not routinely considered due to unavailability and paucity of expertise.

Staged repair was done for 4 of the patients in this series (Center A).They initially had a feeding jejenustomy for administration of feeds, cervical esophagostomy and venting gastrostomy. Staged repair for EA/TEF allows for growth of the neonate and natural lengthening of the esophagus. Gastrostomy alone[8] for feeding was the most frequent procedure used prior to 1939.Gastrostomy feeding may hasten death by increasing the regurgitation of gastric content through the patent TEF. Gastrostomy with esophageal banding can be done to reduce the chances of gastroesophageal reflux disease (GERD) in patients with TEF.Feeding jejunostomy was done in this group of patients to reduce GERD.A venting gastrostomy helps to achieve aspiration of gastric contents that could reflux into the esophagus in the presence of TEF (Gross Type C, Vogt's Type 3b).This procedure is still subject for additional research.

Primary repair was done for 3 patients (Center B) via an extrapleural right posterolateral thoracotomy.Anastomosis was done in one layer.Haight[9] described a two-layered telescoping anastomosis which is not currently popular. Significant fewer strictures occur with single –layer technique[10].

Major early postoperative complications were observed in both groups of patients analyzed as shown in Table 2.Wound infection was observed in both groups that had primary repair and staged repair. Sepsis and disseminated intravascular coagulopathy (DIC) were observed in patients who had staged repair.The patients that had sepsis had excoriation of the skin around the gastrostomy site and also wound infection.Management of sepsis in neonates can be challenging as they may not exhibit the characteristic features as seen in adults. Two patients of those that had primary repair had postoperative cardiac arrest few hours after surgery. .

Management of EA/TEF in these centers had been extremely demanding for both the parents and surgeons. Most of the parents are indigent and are not always able to provide needed support at all times. This is compounded by the limited facilities at the disposal of the Surgeons. These are factors that can adversely affect proper care of neonates with EA/TEF.

Conclusion

The management of neonates with EA/TEF is a highly intensive one .The choice of the surgical options depends largely on the surgeons preference and department policy as seen in this review.It was observed from this review that both primary and staged repair recorded significant morbidities and mortalities.This is an ongoing work and the outcome is expected to improve when patients present early and when requisite tools for adequate care are available.

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