

Congenital Upper Gastrointestinal Structural Anomalies in Infants with Special Reference to Oesophageal Anomalies at Tertiary Care Institute in Central India

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Abstract:

Background:

A wide spectrum of congenital anomalies may affect the upper gastrointestinal tract, including anomalies of the esophagus (e.g., atresia, fistulas, webs, duplications, vascular rings), stomach (e.g., congenital gastric outlet obstruction, duplications), and duodenum (e.g., atresia, annular pancreas, duplications, malrotation).

Method: - A descriptive study was done prospectively and approximately 46 patients were included for a period of 15 months. All infants with congenital gastrointestinal anomalies who were admitted in paediatric medicine or surgery ward in Sanjay Gandhi Memorial Hospital and Gandhi Memorial Hospital.

Result: - In the present study, the incidence of low birth weight babies was approximately 54.35% according to the WHO definition but in normal Indian population, the incidence of low birth rate (< 2.5kg) is 13.04% of all live births (World health report, 1995) among the 31 patients of esophageal atresia and tracheo-esophageal fistula, the sex distribution showed that male patients were more than the double of female patients.

Conclusion: - CT and MR imaging are unsuitable for general screening because of the need for patient sedation and monitoring, but they do provide superb anatomic detail and added diagnostic specificity. They are especially useful in demonstrating esophageal duplications and vascular rings as well as associated abnormalities.

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Introduction

The Global Burden of Disease study concluded that birth defects (also called congenital malformations, birth defects, or birth defects) are the fifth leading cause of

death for children under the age of five worldwide. [1] This equates to approximately 500,000 deaths annually from birth defects, 97% of which occur in low and middle income countries

(LMICs). In fact, this may underestimate the true number of deaths due to the underdiagnosis of newborns with birth defects who die in the community and the lack of death certificates in many low- and middle-income countries. [2] Not only is mortality high among low-income groups, but micronutrient deficiencies, infections, and teratogens during pregnancy also contribute to higher prevalence, increased cases, and lack of prenatal testing to prohibit abortion. increase. [3 4]

Incidences of each range from 1 in 2000 to 1 in 5000, collectively accounting for up to 40% of emergency neonatal surgeries, and associated mortality can exceed 50% in many LMICs. [5–8]

Congenital anomalies of the upper gastrointestinal tract are a significant cause of morbidity in children, overall prognosis of congenital structural anomalies of the gastrointestinal is very good and has improved with improved medical management and the advent of newer surgical modalities.

In 2010, the World Health Assembly passed a resolution recommending "where possible prevention, screening programs, care and ongoing support for children with birth defects and their families".² Prevention is paramount. The Sustainable Development Goal 3.² aims to end preventable deaths of newborns and children under the age of 5 years by 2030.⁵ One-third of infant deaths are due to birth defects, and this clearly cannot be achieved without accelerating efforts to provide surgical care for children.⁶ In fact, studies have shown that such care is highly cost-effective in terms of extending lifespan when disability is taken into account.⁹ Yet, neonatal and paediatric surgical care remains a low priority on the global health agenda.⁹ The goal is to identify factors that can be corrected. This is important to support advocacy and global health prioritization and to inform future

intervention studies aimed at improving outcomes.

Aims and objectives

- * To know the incidence of various upper gastrointestinal structural anomalies at tertiary care institute in Central India.
- * To know the surgical outcome of various upper gastrointestinal structural anomalies.
- * To know the possible perinatal risk factors.
- * To know the other associated congenital anomalies

Material and methods

A Descriptive Study was done prospectively and Approximately 46 patients were included. All infants with congenital gastrointestinal anomalies who were admitted in paediatric medicine or surgery ward in Sanjay Gandhi Memorial Hospital and Gandhi Memorial Hospitals associated with Shyam Shah Medical College, Rewa (M.P.) during the period of 1st April 2021 to 30th June 2022 (15 Months) with following inclusion and exclusion criteria

Inclusion Criteria-

All infants including neonates who were come for Emergency admission in Department of surgery(casualty) or admitted at SNCU OR NICU in Department of Paediatrics or Paediatric surgery outdoor or indoor, for various upper gastrointestinal structural anomalies were included.

Exclusion Criteria-

Predominantly those with non surgical management.

Patient Data Collections :

All infants who are having congenital gastrointestinal structural Anomalies Attending Paediatric Surgery OPD and patient were admitted in SNCU or NICU in Department of paediatrics or Emergency

admission in Department of surgery (casualty) were included in this study. All the details of patients was entered in predesigned structured proforma. A patient who are undergoing initial conservative managements and surgical interventions were recorded.

Investigation Details:

Patient was investigated for HB, TLC, DLC, LFT, RFT, Blood Sugar, Urine Routine and microscopic, Blood Grouping,

BT, CT, PT-INR, X Ray chest, USG Abdomen. The other investigations like MRI and CT scan of abdomen and thorax, Gastrograffin, Barium meal, barium meal follow through, barium swallow will be done if required.

Statistical Analysis : After collection and compilation of data, appropriate statistical tests were used to analyse the data.

Results

Table 1: Neonates Gender

SN	Gender	No of Cases	Percentage
1	Male	29	63.0
2	Female	17	37.0
Birth Weight of Neonates			
1	1.0 to 1.5 kgs	25	54.35
2	1.5 to 2.0 kgs	14	30.44
3	2.1 to 3.0 kgs	06	13.04
4	>3.0 kgs	01	2.17
Length of Pregnancy (Gestational age)			
1	Preterm	39	84.78
2	Term	5	10.87
3	Post term	2	4.35
APGAR Score			
1	5 in 1 minute, 6 in 5 minute	32	69.56
2	6 in minute 7 in 5 minute	14	30.44

From the observed data, the incidence of congenital anomalies was observed to be more male infants 29 (63.0%) cases and 17 (37.0%) were female infants. The incidence of congenital anomalies was observed to be more when the birth weight was less than 1.5 kg (54.35%). Hence weight <1.5 kg is a significant risk factor for congenital malformations. (Table-1)

Table 2: Complications of Neonates

SN	Complications	No of Cases	Percentage
1	Frothing respiratory distress	31	67.39
2	Unable to accept feeding vomiting after feeding distended abdomen since birth	06	13.04
3	Non bilous vomiting and distension	05	10.86
4	Excessive salivation since birth bluish after each feed attempt difficulty in breathing, coughing after feeding, abdominal distension after birth 24hrs of birth	03	6.52
5	Abdominal distension, non-bilious vomiting	01	2.17
	Total	46	100.0

The above table were found for complication 31 (67.39%) in frothing respiratory distress and 6 (13.04%) cases were found in unable to accept feeding vomiting after feeding, 5 (10.86%) cases were found in non-bilious vomiting and distension. (Table-2)

Table 3: Anomalies of Infants

SN	Anomalies	No	%
1	No anomalies	31	67.39
2	Congenital Heart Disease	12	26.08
3	Anorectal malformation	1	2.17
4	Anorectal malformation with arm with bifid scrotum	1	2.17
5	Down Syndrome	1	2.17
Infant according to x-ray chest and abdome			
1	NG tube coiled	34	73.91
2	Dilated stomach	04	8.69
3	Double bubble	06	13.05
4	Not significant	01	2.17
5	Pyloric web	01	2.17
Antenatal USG			
1	Polyhydramnios	36	78.26
2	Single umbilical artery	01	2.17
3	Double bubble sign	02	4.34
4	Normal	01	2.17
5	No record	06	13.05

The maximum anomalies of infants were found in 31 (67.31%) no anomalies and 12 (26.08%) were found in CHD, 1 of each was found in anorectal malformation, down syndrome and anorectal malformation with arm with bifid scrotum. 34 (73.91%) NG tube curled in x-ray chest and abdomen, 4 (8.69%) dilated stomach and 6 (13.05%)

were found in double bubble, 1 of each was found in not significant and pyloric web. 36 (78.26%) polyhydramnios in antenatal USG, 6 (13.05%) were found in no record and 2 (4.34%) were found in double bubble sign, 1 of each was found in single umbilical artery and normal antenatal USG. (Table-3)

Table 4: Diagnosis of Upper congenital anomalies

SN	Diagnosis	No	%
1	Oesophageal atresia upper blind pouch with lower fistula	19	41.30
2	H type tracheo oesophageal fistula	08	17.39
3	Duodenal atresia	06	13.05
4	Pyloric Stenosis	05	10.86
5	Oesophageal atresia with TEF with right bronchooesophageal fistula	04	8.69
6	Pure oesophageal atresia	03	6.52
7	Pyloric web	01	2.17
Surgical procedure done			
1	Posteriolateral Thoracotomy with ligation of fistula with end to end anastomosis with right ICD placement	12	26.08
2	Decompressive gastrostomy	04	8.69
3	Diversion cervical esophagostomy end feeding gastrostomy	01	2.17
4	Right posterolateral thoracotomy with primary repair oesophageal defect	01	2.17
5	Cervical oesophagostomy with feeding jejunostomy	01	2.17

6	Right posterolateral thoracotomy with ligation of bronchoesophageal fistula lower part with closure of both oesophageal end with feeding tube with ICP placement	10	21.73
7	Laparotomy duodenostomy with transverse colostomy with drain placement	04	8.69
8	Laparotomy duodeno jejunostomy	02	4.34
9	Laparotomy ladd's band excision (Kimura's) duodeno jejunostomy	01	2.17
10	Pyloromyotomy	05	10.86
11	Kimura's duodenostomy with end feeding gastrostomy	01	2.17
12	Kimura's duodeno duodenostomy with left side colostomy descending sigmoid loop (complete type 2)	01	2.17
13	Right posterolateral thoracotomy with ligation of fistula end to end long lap moderate tension oesophageal anastomosis with ICD	02	4.34
14	Right posterolateral thoracotomy with ligation of fistula with feeding gastrostomy with cervical oesophagostomy (long segmental cap)	01	2.17
Postoperative complications			
1	Pneumonitis septic shock	21	45.65
2	Anastomotic leak pneumothorax	12	26.08
3	No complications	12	26.08
4	Structure after 1 month	01	2.17

The maximum number of cases were found in 19 (41.30%) Oesophageal atresia upper blind pouch with lower fistula, 8 (17.39%) were found in H type tracheo oesophageal fistula and 6 (13.05%) were found in Duodenal atresia, 5 (10.86%) were found in pyloric stenosis, 4 (8.69%) were found in Oesophageal atresia with TEF with right bronchoesophageal fistula, 3 (6.52%) were found in pure oesophageal atresia, 1 was found in pyloric web. 12 (26.08%) posteriolateral thoracotomy with ligation of fistula with end to end anastomosis with right ICD placement, 10 (21.73%) were

found in right posterolateral thoracotomy with ligation of bronchoesophageal fistula lower part with closure of both oesophageal end with feeding tube with ICP placement, 5 (10.86%) were found in pyloromyotomy, 4 (8.69%) each were found in decompressive gastrostomy and laparotomy duodenostomy with transverse colostomy with drain placement. 21 (45.65%) pneumonitis septic shock, 12 each (26.08%) were found in anastomotic leak pneumothorax and No complications. 1 was found in structure after 1 month.(Table-4)

Table 5- Outcomes

SN	Outcomes	No	%
1	Expired	31	67.39
2	Discharge	15	32.61
Mortality with Sex (n=31)			
1	Male	17	54.83
2	Female	14	45.17
Mortality with anomalies (n=31)			
1	Oesophageal atresia with pneumothorax	20	64.51
2	Tracheoesophageal atresia	08	25.80
3	Pure oesophageal atresia	02	12.90
4	Duodenal atresia	01	3.22

The outcome were found in 31 (67.39%) expired and, 15 (32.61%) were discharge in

after postoperative period. 31 out of 46 cases in 17 (54.83%) were found in male

and, 14 (45.17%) were female. 20 (64.51%) were found in oesophageal atresia with pneumothorax, 08 (25.80%) were found in tracheoesophageal atresia, 02 (12.90%) were found in pure oesophageal atresia and 01 (3.22%) were duodenal atresia. (Table-5)

Discussion

Esophageal atresia (EA) is defined as an interruption in the continuity of the oesophagus with or without fistula to the trachea. It is considered the most common congenital anomaly of the esophagus [10]. Some define 2 cm as a cut-off point, others classify the gap into short (1 cm), intermediate (2.5–3.0 cm) and long (>3 cm), others define a gap more than 3–3.5 cm as long, while still others recommend an esophageal replacement if the gap exceeds the length of six vertebral bodies. There is also a lack of uniformity in the methods used to measure the gap.

In the present study, among the 31 patients of esophageal atresia and tracheoesophageal fistula, the sex distribution showed that male patients were more than the double of female patients. Similar male predominance has been reported by Kronemer et al and Arora M et al. [11-12] This may be due to the reason that in our society male child's health is given preference over the female child's health.

Only 45.0% of our patients reached the tertiary centre within 24 hours. Arora et al, reported similar kind of data i.e. 40.7% cases reached within first 24 hours in spite of 75% deliveries at hospital. [12] Agarwal et al in AIIMS hospital reported that the patients presenting within 24 hours were 44% and similar data (46%) was also obtained by Sharma AK et al. [13-14] Traveling long distance, poor transport system and poverty are also the contributory factors.

Mortality was high among patients who reached the tertiary centre late i.e. >24 hours. This is similar to the observations reported by Arora M et al. [12] In our setup,

there is no facility of ultrasonography in primary health centre, sub-center or in antenatal clinic and lack of awareness of this anomaly in medical and paramedical workers. The early diagnosis of EA with TEF is possible if obstetrician suspect the anomaly in antenatal period in the pregnancy associated with polyhydramnios (incidence is 0.5-1% in normal pregnancies). These pregnancies should be taken as high-risk pregnancies and should be kept under observation and regular follow up throughout the pregnancy to facilitate early diagnosis and early referral of EA with TEF patient to the better centre. The role of ultrasonography in prenatal diagnosis of EA with TEF is limited because of less accuracy. Sparey C et al reported 50% of accuracy and 42% accuracy was observed by Robert K et al. [15-16]

In the present study, the incidence of low birth weight babies was approximately 54.35% according to the WHO definition but in normal Indian population, the incidence of low birth rate (<2.5 kg) is 13.04% of all live births (World health report, 1995).¹⁷ The incidence of <2 kg babies was 30.44% in normal population and 13% in the present study which is comparable with Arora M et al they reported the incidence of less than 1.8 kg as 15%. [12]

The reasons for the high incidence of 54.35% of low birth weight found in the present study are many, such as rural background population, adolescent pregnancies, higher incidence of severe anemia, maternal malnutrition, multiple pregnancies and antenatal infections which are mostly related to poverty. The mortality in the low birth weight patients is quite high than the babies weighted more than 2.5 kg but it was statistically not significant.

The results are comparable with those reported by Arora M et al. [12] Almost 2/3 of patients had clinical or radiological evidence of pneumonia (67.39%). The presence of pneumonia grossly affects the

survival. When we compared our data with that of study conducted by Agarwal S et al we observed that survival has improved over the past few years probably because of better preoperative and post-operative care and potent newer antibiotics.¹³ Pneumonia in EA with TEF patients is mainly due to aspiration of saliva, preoperative breast feeding, and reflux from fistulous communication with trachea. So, these can be prevented with vigorous upper pouch section during transportation and early diagnosis before feeding may decrease the incidence of pneumonia and increase the survival of EA with TEF patients.

The total number of major associated anomalies was 26.08% and minor associated anomalies were 2.17% in the present study. In comparison to Arora M et al, they found 21% of major associated major anomalies and 33% of minor associated anomalies and the overall incidence of associated anomalies patients in the present study was 33.61% as of 35% incidence reported by Arora M et al, 49% by Rejjal A et al and 50% by Dwayne C et al. [12,18-19] These data are comparable with the present study. The system wise distribution of associated anomalies is comparable with Rejjal A et al. [18] In comparison to Rejjal A et al, we have higher incidence of GI tract anomalies lower incidence of genitourinary tract anomalies. [18] Survival rate was 32.61% and mortality rate was 67.39% which is comparable with Arora M et al.¹² There are various reasons for higher mortality e.g. late presentation of patients at the centre managing these problems, poor transportation facilities for sick neonates, concentration of trained man power only in the major metropolitan cities, lack of adequate infrastructure at most of the centers in India.

The current series of ELBW infants with EA/TEF is one of the largest reported in the literature. Although the number of ELBW infants born with EA/TEF is small, accounting for only 3% of all

newborns with EA/TEF, morbidity and mortality are very high, most of them with associated congenital Gender abnormalities have been added to premature birth. This reflects the poor survival reported in the literature for ELBW infants without EA/TEF in general [20-24]. In the present series, the survival rate for ELBW and EA/TEF infants is 50%. This is much lower than the survival rate recently reported at our institution (91% at 3 years) [25].

Prenatal ultrasound was less definitive in this series of patients. Scans were normal in 2 fetuses, showed polyhydramnios in 2, and were nonspecific in her other four. The prenatal diagnosis rate of EA using ultrasound may be low in early pregnancy, but may be higher in late pregnancy when other features such as small gastric bubbles and a dilated cecal esophageal pouch are visualized. may be accurate²⁶. Given the preterm birth rate in this patient series, most had only their first prenatal scan, which could explain the low ultrasound yield.

This condition raises ethical question regarding the management of these infants, but given the very poor prognosis, no further surgical treatment is recommended after the diagnosis has been confirmed by chromosome analysis and parental counseling. may not be justified [27]. One patient was born with mosaic trisomy 14. This is a very rare chromosomal abnormality and to our knowledge has never been reported in association with EA/TEF. Patients with mosaic trisomy 14 have growth and psychomotor retardation, dysmorphic facial features, congenital heart defects, and cryptorchidism [28]. Although the life expectancy of these patients has not been studied, about two-thirds of people with trisomy 14 mosaicism survive relatively free of serious medical problems since childhood [28]. Attempts were therefore made to cure this child, but the end result was poor.

The surgical approach to infants with ELBW and EA/ TEF is variable and should be individualized. One option is to perform TEF ligation.

Postpone esophageal repair until the infant is older and gains weight, possibly in combination with a gastrostomy for feeding. Another option is to perform immediate TEF ligation and esophageal anastomosis. In any case, it is important to ligate the TEF promptly as there is a risk of aspiration and death if left untreated. Preterm infants with severe respiratory distress syndrome require constant positive pressure ventilator support, which can lead to a fatal cascade of severe gastric distension, further exacerbating ventilation with possible gastric rupture. It can cause tension pneumoperitoneum [29]. In some of these cases, TEF ligation dramatically improves the infant's respiratory status, allowing primary esophageal repair to proceed safely.

The strategy we used at our institution was to delay esophageal anastomosis unless the patient was stable enough to tolerate it. Of the 4 survivors, 3 had delayed anastomoses and 1 underwent primary repair. All three of her patients who died were treated with her TEF ligation with a delayed anastomosis plan. However, none of these deaths were due to delayed final repair or complications associated with the initial esophageal surgery. In two of these patients, the decision to discontinue aggressive treatment with family consent was due to bilateral intraventricular hemorrhage in one and neurological complications due to multiple cerebellar and cerebral hemorrhages in the other. based on a very poor clinical outcome. Surgical complications typical of EA/TEF patients, including thoracic duct injuries, anastomotic leaks and/or strictures, are well managed. Tracheomalacia and gastroesophageal reflux disease, known to

be associated with EA/TEF, have also been successfully treated.

Ultimately, all but one of the survivors with the shortest follow-up will be fully orally ingested.

Regardless of the surgical approach, one should not underestimate that these infants suffer all the complications of premature birth. In fact, all survivors in this series had severe morbidity associated with early pregnancy. (28 weeks gestation), 3 very preterm infants (28–32 weeks gestation), and 2 moderately preterm infants (32–33 weeks gestation). gestational weeks) [30]. However, also the number and severity of associated anomalies play an important role for survival.

Conclusion

CT and MR imaging are unsuitable for general screening because of the need for patient sedation and monitoring, but they do provide superb anatomic detail and added diagnostic specificity. They are especially useful in demonstrating esophageal duplications and vascular rings as well as associated abnormalities. Some of these anomalies can remain asymptomatic, in which case diagnosis is the result of incidental findings at routine examination for other conditions in adulthood.

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