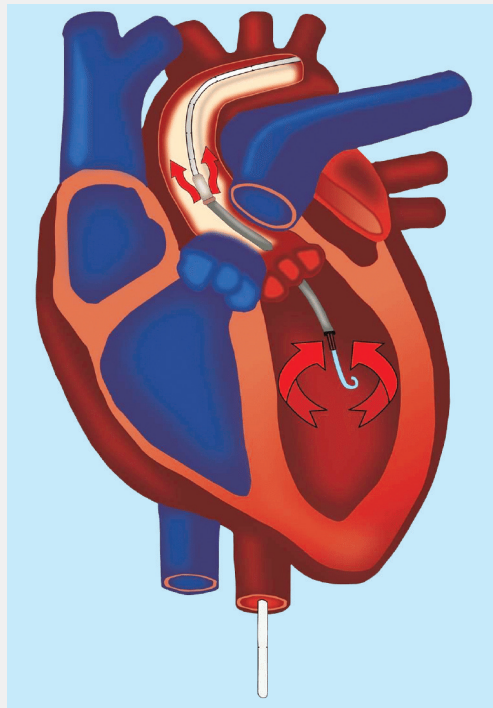


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**Editorial**

*"Paediatric Cardiac Intervention in Bangladesh"*



Bangladesh Institute of Child Health



Dhaka Shishu (Children) Hospital

# DHAKA SHISHU (CHILDREN) HOSPITAL JOURNAL

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**EDITORIAL**

# Paediatric Cardiac Intervention in Bangladesh

Manzoor Hussain

## Introduction

Technological innovations have greatly advanced treatment of cardiovascular disease in both children and adults with congenital heart disease (CHD). The field of pediatric cardiac interventions has witnessed a dramatic increase in the number and type of procedures performed in the past 2 to 3 decades. Interventional therapy has become an acceptable alternative treatment for many CHD, including closure of atrial septal defects (ASDs), muscular ventricular septal defects (VSDs), patent ductus arteriosus (PDA), dilatation of stenotic valves (aortic and pulmonary) and dilatation of stenotic vessels [branch pulmonary arteries, coarctation of the aorta (CoA)]. The era of intervention in pediatric cardiology started in 1966 by Rashkind et al<sup>1</sup> with the advent of balloon atrial septostomy & by Portsman et al<sup>2</sup> in 1967 with non-surgical PDA closure by Ivalon plug. Since then lot of innovations/bioengineering modifications of different devices with animal studies were done.

Congenital heart surgery, together with transcatheter interventions, has resulted in a marked improvement in survival for those with critical CHD. Some interventions performed in the first weeks of life to optimize hemodynamics and prevent end-organ injury associated with delayed diagnosis. With the advent of prostaglandin therapy for duct dependent lesions, many previously lethal congenital heart conditions that present with severe hypoxemia, shock, and acidosis in the newborn period are now survivable and can be palliated.<sup>3</sup> Timely recognition of critical CHD could improve outcome, so it is important to identify and evaluate strategies to enhance early detection.

Surgical method of treatment of CHD is an established method. But non-surgical device closure is an

alternative mode replacing the surgical method.<sup>4</sup> Surgical method has the more morbidity and mortality than the transcatheter device closure of some of the CHDs. Surgery is subjected to complication of cardio pulmonary by pass (CPB) which not infrequently leads to perfusion injury to brain and other vital organs.<sup>5</sup> In some cases where the percutaneous approach is difficult or the patient still requires repair of other associated cardiac anomalies, a hybrid approach can be implemented with its obvious advantages to the patient.<sup>6</sup>

CHD occurs in 5-8 out of every 1000 livebirths.<sup>7,8</sup> Like other developing countries, Bangladesh is facing a multitude of health problems and CHD is one of them. Improvement of diagnosis and orientation or awareness among general pediatrician and early referral to pediatric cardiologists has resulted in an increase of reported prevalence of CHD in Bangladesh.<sup>9,10</sup> Many of them are critical CHD which requires surgery or catheter intervention in the early period of life and responsible for more deaths than any other type of malformation.<sup>11</sup> Without early recognition, diagnosis and treatment, vast majority of infants with CHD die in their first month of life in developing countries.<sup>12,13</sup>

Percutaneous cardiac interventions is an emerging life saving procedure for critically ill children with CHD. These techniques complement surgical interventions. Variations in anatomy, patient size, and general condition at presentation add to the challenge of carrying out these catheter interventions.

## Bangladesh situation

In developed and developing countries there is a wide gap regarding pediatric cardiac care. Absence of pediatric cardiac centers, presence of cardiac centers only in large cities and absence of specific health

1. Professor & Head, Department of Paediatric Medicine, & Paediatric Cardiology, Bangladesh Institute of Child Health (BICH) and Director, Dhaka Shishu (Children) Hospital.

care policies in various countries are the reasons for this variation.<sup>14</sup> Scarcity of pediatric cardiac care in public hospitals, expense of treatment in private hospitals, lack of resources and trained personnel in this field and lack of awareness are the major reasons due to which treatment for CHD is currently out of reach for a majority of children in Bangladesh.<sup>15</sup>

Catheter based procedures for CHD has emerged as a valuable alternative to medical & surgical therapy in Bangladesh since 1998 when a balloon atrial septostomy was performed on a transposition of great arteries (TGA) case in cath lab of CMH, Dhaka.<sup>16</sup> Since then there is significant advancement in the field of pediatric cardiac interventional treatment in Bangladesh. There are several milestone achievements of the paediatric cardiology unit of CMH, Dhaka. They did first ever balloon atrial septostomy in 1998, pulmonary valvuloplasty in an infant in 1999, PDA coil occlusion in 2000, ASD device closure in 2001, aortic balloon in critical AS in 2001, CoA balloon in newborn in 2001, PDA device closure in 2003, VSD device closure in 2004, PM VSD Closure in 2006, PDA Stenting in 2006, VSD coil occlusion in 2007, coronary fistula closure in 2010, pulmonary valve replacement with Melody valve in 2012.<sup>17</sup> Fatema et al<sup>18</sup> also reported 1020 patients with secundum ASD closure with device since 2000 to 2013 using Amplatzer, Lifetech, Figulla, Cookon and some other Chinese devices without any complication. Existence of multiple simple congenital heart lesions is a commonly encountered problem in paediatric cardiac practice. Multiple interventions can be done on these lesions safely. One years experience of multiple intervention reported at CMH, Dhaka showed excellent outcome with ASD+PDA, VSD+PDA, PDA+CoA, ASD+VSD, ASD+VSD+PDA.<sup>19</sup>

Since 2005, National Institute of Cardiovascular Diseases (NICVD) started their journey with a PDA device closure. There is marked advancement in cardiac interventions in NICVD also. They are doing PDA device and PDA coil closure, ASD device closure, balloon pulmonary valvuloplasty (BPV), balloon aortic valvuloplasty (BAV), coarctation balloon angioplasty, mitral valvuloplasty (PTMC), balloon atrial septostomy successfully.<sup>20</sup> Salam et al<sup>21</sup> at NICVD showed that combined interventional treatment in suitably selected cases may be feasible and safe alternative to surgery in many CHD like

PDA+CoA, PDA+PS, ASD+PS, ASD+PDA, AS+PDA, AS+CoA, multiple ASD and ASD+MS.

The paediatric cardiology unit of Dhaka Shishu (Children) Hospital is the first and pioneer in pediatric cardiology in Bangladesh. It started functioning back in 1988 to provide treatment, control and prevention of childhood cardiac problems and the unit was enriched and started cardiac intervention and cardiac surgery since 2012. More than 300 cardiac surgery and 400 cardiac interventions were already been done. Rima et al<sup>22</sup> reported life saving balloon atrial septostomy for TGA, balloon angioplasty (BA) of native CoA, balloon dilation of the aortic valve (BAV) for AS, balloon valvuloplasty for pulmonary valve stenosis and PDA stenting at Dhaka Shishu (Children) Hospital Paediatric Cardiac Center. They are also doing PDA device and PDA coil closure, ASD and VSD device closure.

BSMMU also performing device closure in CHD.<sup>23</sup> National Heart Foundation and Research Institute started device closure of PDA, ASD, VSD, balloon aortic valvuloplasty, pulmonary valvuloplasty, MAPCA coiling, balloon atrial septostomy and ballooning of CoA from 2014.<sup>24</sup>

### Conclusion

Paediatric cardiac interventions has witnessed a marked improvement in recent years. It has become an acceptable alternative treatment for many CHD. We have to take the challenges of carrying out these transcatheter interventions in Bangladesh.

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## SPECIAL ARTICLE

# Carbide in Fruits: The Misdeed is on

Probir Kumar Sarkar

Fruits are known to be foods of heaven. Fruits are widely distributed in nature, usually consumed raw and considered as nature's best gift for human beings. Everyone knows that fruits are delicious, that is why, all generations of people are fond of them very much. Being a part of a balanced diet, fruits play a vital role in human nutrition by supplying the necessary growth regulating factors essential for maintaining normal health. It has been said that God first separated the salt water from fresh, made dry land, planted a garden, made animals and fish- all before making a human. God made and provided what we need before we were born. Of them fruits are the best and more powerful when eaten raw. Nevertheless much of way remains to be made for better knowing the impact of the fruits and their components, on the health and the prevention of the principal chronic diseases. Beyond the consumable part of the fruits, an emphasis are also put on the by-products, such as the fruit peels, that could represent precious layers for food, medicinal or cosmetic purposes. But these heavenly foods are not that heavenly in our country. Now a day's fruits are deliberately being contaminated by chemicals causing serious health hazard.

The fruits and vegetables may require few days to ripen after maturation and this short period seriously limits its commercialization in distal markets. So the unripe fruits are collected and they are ripped by using different chemicals as fruit ripening agent. All incidents of food adulteration are united by a common and noble motive- the reduction of post harvest loss. In 2009, the Food and Agriculture Organization estimated that 40-50 percent of the global crop of fruit and vegetables is lost annually to spoilage or waste. In a world where hunger is still commonplace, the scale of this loss is concerning.<sup>1</sup> In such an environment, individual distributors able to implement innovative, cost-effective ways to reduce the loss of the products they handle are rewarded both by increased earnings and reduced

uncertainty. This should result in the adoption of practices that are beneficial all. The problem is that the easiest solutions are also the most harmful to humans.<sup>2</sup>

### What is natural ripening?

Fruit ripening is a highly coordinated, genetically programmed and an irreversible phenomenon involving a series of physiological, biochemical and organoleptic changes, that finally lead to the development of a soft edible ripe fruit with desirable quality attributes. Excessive textural softening during ripening leads to adverse effects upon storage. Carbohydrates play a major role in the ripening process, by way of depolymerization leading to decreased molecular size with concomitant increase in the levels of ripening inducing specific enzymes, whose target differ from fruit to fruit. Whether fruit ripen on the plant or after harvest, the general changes associated with ripening process is softening of fruit, change in colour and development of characteristic aroma and flavor. There is also reduction in sourness and increase in sweetness of the fruit. Usually fruit produce ethylene gas, a plant hormone, naturally that ripens the fruit.

### What is artificial ripening?

Unsaturated hydrocarbons such as acetylene, ethylene etc. can promote ripening and induce colour changes effectively. Although the cosmetic quality of such artificially ripened fruits was found to improve, organoleptic quality was impaired especially when harvested fruits are subjected to treatment without considering their maturity status. Besides, the quantity of ripening agent required to induce ripening for better cosmetic quality, including appearance will be much more than conventional dose, when properly mature fruits are not used for such purposes.

### What is calcium carbide?

With the development of food trade the fruits are sent to distant places, requiring several days in

### Identification of Calcium Carbide ripened Fruits

Qualitative Parameters	Types of Fruits	
	Artificially ripened fruit by Calcium carbide	Naturally ripened fruit
Weight per fruit	Fair	Good
Texture	Not very attractive but uniformly coloured	Attractive but not uniformly coloured
Aroma	Good	Best
Firmness	Fair	Fair
Taste	In-core sour, mildly pleasant	Sweet pleasant
Self-life	Shorter, black blotches appear on the skin of the fruit in two to three days	Longer

ordinary or refrigerated transportation and only firm and mature fruits are least damaged during marketing. The fruits are ripened at the destination markets before retailing and hence artificial ripening has become essential. The most commonly used chemical for artificial ripening is Calcium Carbide ( $\text{CaC}_2$ ) and is popularly known as “Masala”, though banned under The Bangladesh Pure Food Ordinance, rules 1959 and The Bangladesh Pure Food (Amendment) Act, 2005 made thereunder.

Calcium Carbide is colourless when pure, but grayish-white to black in colour otherwise, with garlic like odour. When it reacts with water, it produces acetylene gas (popularly referred to as carbide gas) which is an analogue to ethylene and quickens the ripening process. Calcium Carbide contains traces of arsenic and phosphorus hydride.

$\text{CaC}_2 + 2\text{H}_2\text{O} = \text{C}_2\text{H}_2 + \text{Ca}(\text{OH})_2$  and said to have the effect as ethylene the natural ripening hormone. However, acetylene is not nearly as effective for ripening as is ethylene and acetylene is not a natural hormone as ethylene. A strong reactive and corrosive chemical  $\text{CaC}_2$  has carcinogenic properties and is used in gas welding. Being cheap and easily available in the local markets  $\text{CaC}_2$  is indiscriminately being used in preference to other recommended practices of inducing ripening in fruits.

#### Effects of Calcium Carbide on Fruit Quality

Fruits ripened with Calcium Carbide are overly soft, are inferior in taste and flavour. They also have a shorter self life. The fruit ripened with Calcium Carbide may develop uniform attractive surface colour, but the tissue inside would not be ripe or may remain green or raw. When Calcium Carbide is

used in very raw fruit, the amount of chemical needed to ripen the fruit has to be increased. This results in the fruit becoming even more tasteless, unhealthy and possibly toxic.<sup>3,6</sup>

#### Potential Health Hazards Associated with Calcium Carbide

Calcium carbide, a dangerous corrosive chemical used as ripening agent for mangoes, bananas, litchis, jackfruits and other fruits also. Packets of calcium carbide powder are kept in the container of fruits where in contact with moisture, acetylene gas is produced and acts as a ripening agent.  $\text{CaC}_2$  contains traces of arsenic and phosphorus hydride and the acetylene gas has carcinogenic properties.<sup>3</sup> The early symptoms of arsenic or phosphorus poisoning include vomiting, diarrhea with or without blood, sore throat, cough, shortness of breath, burning sensation of chest and abdomen, thirst, weakness, difficulty of swallowing, irritation or burning of the eyes and skin to ulceration. Acetylene gas may affect the neurological system by inducing prolonged hypoxia which causes headache, dizziness, mood disturbances, sleepiness, mental confusion, memory loss, cerebral edema and seizure. In the long run,  $\text{CaC}_2$  adversely affects vital organs such as the liver and kidney resulting in organ failure and/or cancer of various organs.<sup>3,4</sup>

#### Precautions to be taken by the consumers

- Wash the fruits thoroughly before consumption under portable running water for few minutes, so that the chemicals are washed away.
- While eating fruits, it is better to cut the fruit into pieces, rather than consuming them directly.
- As far as practically possible, peel off the fruits before consumption.

- Even better option is to buy mature fruits directly from growers and to ripen it at home in warm temperature in contact with one ripe fruit.

### Conclusion

The first ripening of fruits means they may contain various harmful properties. Bangladesh pure food ordinance, 2005 & Bangladesh Pure food act, 2013 prohibits use of any poisonous chemical like calcium carbide, formalin, pesticides or toxic colour/ flavour in any food that may cause harm to human body.<sup>7,8,9</sup> The Bangladesh high court recently issued some directives to the government to stop this practice and some administrative drives are also been taken. Despite this legal prohibition, chemically treated fruits are selling openly in all markets and ripening agents are also available. Department of environment need to be activated with proper authority, strength and administrative actions should be reinforced more vigorously. Above all, due attention should be given to build awareness among traders and consumers.

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## ORIGINAL ARTICLE

# Balloon Atrial Septostomy - An Essential Lifesaving Intervention in Neonate with Critical Cyanotic Heart Disease: Experience of Dhaka Shishu (Children) Hospital Paediatric Cardiac Center

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## Abstract

**Background:** The balloon atrial septostomy (BAS) using fluoroscopy or 2D-ECHO is an accepted effective palliative treatment for cyanotic congenital heart disease (CHD) due to inadequate intra-cardiac mixing. BAS improves the hemodynamics in a variety of compromised circulations like D transposition of great arteries (d-TGA) with intact IVS, Tricuspid or Mitral atresia, Hypoplastic left heart syndrome (HLHS) with restricted interatrial communication until definitive surgery can be attempted. Atrial septostomy (AS) is indicated in some patients with RV failure and associated PH in whom medical therapy has failed.

**Objective:** The study was undertaken to find out the immediate outcome of cyanotic children who required BAS to allow adequate intra-cardiac mixing of blood.

**Methods:** This prospective study was conducted in the paediatric cardiac intensive care unit (PCICU) of Dhaka Shishu (Children) Hospital from July 2014 to October 2016. Total 27 cyanotic children required urgent BAS during the study period. Clinical parameters, SPO<sub>2</sub>, echocardiographic data, cathlab data & outcome were recorded. Statistical analysis was done by using SPSS version 15.

**Results:** BAS was done in 27 patients mostly for d-TGA with PFO/small secundum ASD±small PDA. Mean age for BAS patients were 23 days±13.66 days and mean weight 2.79±0.59 Kg. The diameter of the atrial communication increased from 1.6±0.36 mm to 4.78±0.77 mm (p<0.0001). Oxygen saturations increased significantly at discharge from 40.21±9.6 % to 65.7±9.6% (p<0.0001). Mean pressure gradient between left & right atrium decreased from 7.7±8.12 to 1.08±1 mmHg (p<0.0001). Complications encountered were sepsis in 10(37%), transient cardio-respiratory arrest during the procedure in 2(7.41%) patients and renal impairment in 1(3.70%) patients. Overall mortality was 11.1%.

**Conclusion:** BAS is safe and effective palliative procedure for different cyanotic CHD with good immediate results in our institution.

**Key words:** Balloon Atrial Septostomy, cardiac intervention.

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## Introduction

In the current era, congenital heart surgery allows for repair or palliation of nearly all types of congenital heart malformations. Congenital heart surgery, together with transcatheter interventions, has resulted in a marked improvement in survival of critical CHD. d-TGA is one of such critical CHD characterized by a short life and, with a few exceptions, death occurs in the first 6 months.<sup>1</sup> Early palliative treatment is desirable to keep the child alive long enough for future surgery to become possible. BAS has dramatically improved life expectancy for infants with complete d-TGA. BAS was first described by Rashkind and Miller<sup>2</sup> in 1966 and remains an important interventional procedure in the palliation of certain forms of CHD. It is a closed technique of rupturing the foramen ovale by repeatedly withdrawing a balloon tipped catheter from the left atrium to the right atrium to achieve adequate intracardiac mixing of blood & to increase effective pulmonary flow. After its introduction in 1966 the procedure was performed under the control of fluoroscopic imaging. Recently, cross sectional echocardiography has been used to help position the balloon catheter during the procedure to increase speed in performing the procedure, to reduce risk of trauma to atrioventricular valves and other vital structures, and to reduce exposure to radiation.<sup>3, 4</sup> The aim of the work is to review and evaluate our experience over last 2 years for BAS in cyanotic infant mostly  $\alpha$ -TGA with restricted interatrial defect with intact interventricular septum.

## Materials and Methods

This prospective study was conducted in the paediatric cardiac intensive care unit (PCICU) of Dhaka Shishu (Children) Hospital from July 2014 to October 2016. The diagnosis was confirmed in all patients by 2-dimensional echocardiography with Doppler and color flow Doppler. The equipment used for the echocardiographic assessment was (Vivid q, GE healthcare) with 12 MHz and 6 MHz probe incorporating color flow, pulsed wave and continuous wave Doppler. Therapy for all patients was to begin with prostaglandin E<sub>1</sub> in a dose of 0.05-0.1 nanogm/kg/min to maintain ductal patency if PDA was present. The decision to perform the BAS was made based on the clinical findings of hypoxia, followed by the echocardiographic confirmation of restrictive atrial septal defect/patent foramen ovale, intact interventricular septum in patient with d-TGA.

Z-5 septostomy catheter (NuMED, Inc., Hopkinton, NY) has been used for septostomy. It is a 50-cm long dual lumen catheter with a 13.5 to 0.5-mm diameter noncompliant balloon made of polymeric nylon material with a maximal capacity of 2 cc at the distal end. It has an end-hole that will accommodate a 0.021 guide-wire. The inflated geometry of the balloon is a sphere. There is a radiopaque imaging band in the middle of the balloon for accurate positioning in the left atrium. The catheter tip is angled at 350 to facilitate entry into the left atrium.<sup>5</sup>

The protocol we followed has been previously reported.<sup>6-7</sup> The procedure was done in the catheterization laboratory under fluoroscopic as well as echocardiographic guidance. All patients except one, the procedure was done in the catheterization laboratory using both echocardiographic and fluoroscopic guidance. Reason behind using cath lab was to do coronary angiogram to accurately inform surgeon of coronary artery anomaly. Venous (femoral) access is obtained with the 6F size sheath. A 4.1 F right coronary catheter is then advanced through the sheath up to the right atrium and through the atrial communication over a terumo guidewire 0.018" in the left atrium or left upper pulmonary vein. The terumo guide wire was then exchanged with 0.018" road runner wire. The balloon catheter is advanced over the wire into the left atrium. The standard sub-costal view was mainly used for appropriate positioning of the balloon across the atrial septum to avoid any potential complication is of crucial importance. The balloon is then fully inflated in the left atrium then pulled into the right atrium using a rapid and forceful jerk. The forceful jerk/pull motion should be stopped at the right atrium inferior vena cava junction. The catheter should be pushed back to the mid right atrium then deflated as rapidly as possible. The deflated catheter is advanced to the left atrium and the procedure is repeated until adequate atrial communication is achieved and no resistance is felt during passage of the inflated balloon across the defect. At the end of the procedure the balloon is deflated and pulled outside the body. Before & after the procedure right & left atrial pressure was taken. Finally aortic root angiogram done antegradely using Berman angiographic catheter or Right coronary catheter in laidback view, LAO, AP view to delineate coronary artery. The procedure was done under general anesthesia. Clinical and hemodynamic profile,

associated cardiac and noncardiac structural anomalies, procedural details and complications, immediate outcome were recorded. No Balloon rupture was reported. There was one procedural failure.

Data were statistically analyzed with the use of the Statistical Package for Social Science program (SPSS version 15.0 for windows, Chicago, IL). The descriptive statistical analysis of the quantitative variables was carried out by calculating the median, mean and standard deviations. Paired t test was used to compare the values of oxygen saturation and atrial septal defect diameter and mean interatrial pressure gradient before and after the procedure. Statistical significance was achieved when P was <0.05.

## Results

Twenty seven patients with congenital heart diseases (92% DTGA with restricted ASD/PFO with intact interventricular septum±PDA, 3.8% mitral atresia, 3.8% tricuspid atresia) underwent BAS. Three more patients BAS was done as an additional procedure in PDA stenting was excluded from the study. Small / tiny PDA was present in patients 16 patients (59%). Mean age for BAS patients were 23.9 days±13.8 days (Fig.-1) and mean weight 2.8±0.59 Kg (Fig.-2). Male and female ratio was 8:1 (Fig.-3).

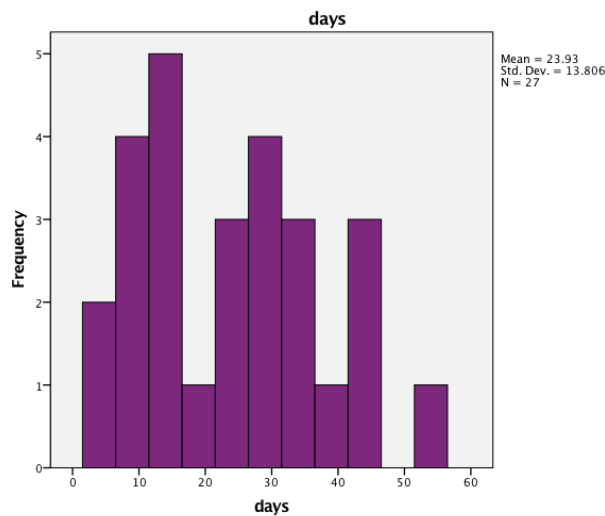


Fig 1 Distribution of age at procedure

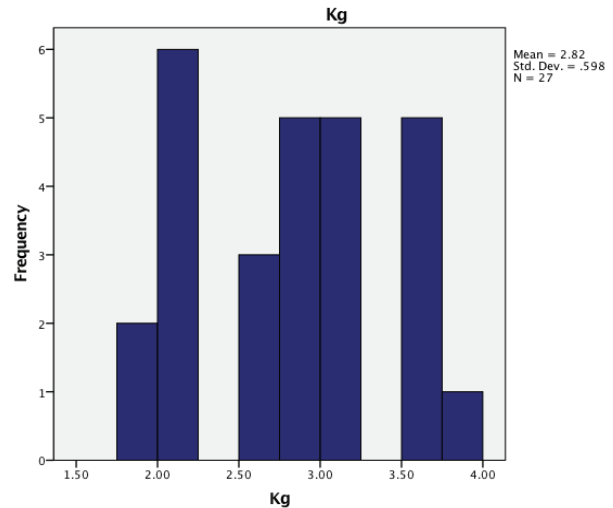


Fig 2 Distribution of weight at procedure

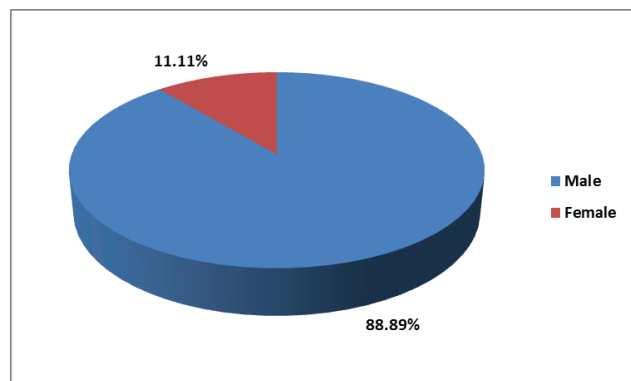
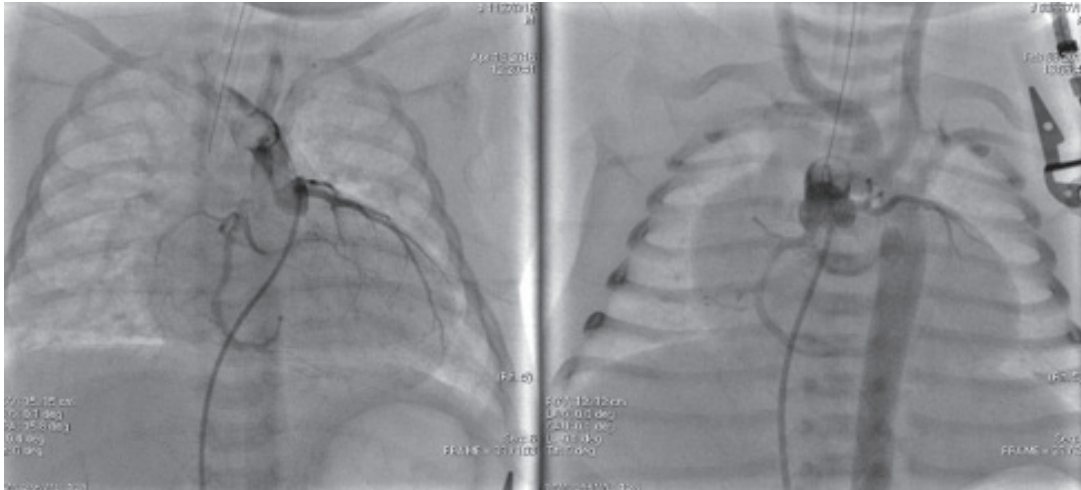


Fig 3 Sex distribution among the study cases

History of Maternal diabetes found in 4 patients (14.8 %) & prematurity observed in 4 patients (14.8 %). Associated noncardiac structural anomalies found in 5 neonates (18.5 %) which includes anorectal malformation (3), hemihypertrophy (1), tongue tie (1) [Table-I].

Table I		
Associated conditions of the cases who underwent BAS (multiple response)		
	Frequency	%
Maternal diabetes	4	14.8
Prematurity	4	14.8
Noncardiac structural anomalies	5	18.5
Drug used during pregnancy other than vitamins	6	22.22
Fever with rash	2	7.41
Maternal hypertension	2	7.41
Family history of CHD	3	11.11



**Fig 4** Aortic root angiogram shows Anomalous coronary artery (LCX from RCA) in d-TGA

The diameter of the atrial communication increased from  $1.6 \pm 0.35$  mm to  $4.788 \pm 0.89$  mm (p value 0.000). Oxygen saturations increased significantly just after the procedure & at discharge from  $41.23 \pm 10$  % to  $82.29 \pm 11.34$  % (p value 0.000) and at discharge from

$40.48 \pm 9.5$  % to  $66.28 \pm 9.8$  % (p value 0.000). Mean pressure gradient between left & right atrium for patients done in the catheterization laboratory decreased from  $7.6 \pm 7.7$  to  $1.07 \pm 1$  mmHg (p value 0.006) (Table-II).

**Table II**

*Distribution of pre and post procedure data (n=27)*

Parameters		Mean $\pm$ SD	P value
SpO <sub>2</sub> (%)	Before procedure	$41.23 \pm 10$	0.000
	After procedure	$82.29 \pm 11.34$	
	Before procedure	$40.48 \pm 9.5$	0.000
	At discharge*	$66.28 \pm 9.8$	
Gradient across atrium (mmHg)	Before procedure	$7.6 \pm 7.7$	0.006
	After procedure	$1.07 \pm 1$	
Size of ASD (mm)	Before procedure	$1.6 \pm 0.35$	0.000
	After procedure	$4.88 \pm 0.89$	

\*Three patients died and excluded from analysis



**Fig 5** Echocardiographic pictures of PFO & ASD before & after septostomy

Coronary angiogram could be done in 18 (66.75) patients of BAS & coronary anomalies found in 2 (7.4%) patients (Fig.-5).

No statistically significant correlation was found between age of procedure below 20 days to saturation at discharge more than 60% ( $p > 0.05$ ) [Table-III].

**Table III**

*Association between ages of procedure below 20 days to saturation at discharge more than 60%*

Age	SPO2 >60%	SPO2 <60%	P value
Below 20 days	10	1	0.059
More than 20 days	7	6	

The most common complication was sepsis (37%). Transient cardio-respiratory arrest in 2 patients during the procedure occurs in BAS patient. Other complications encountered were transient bradycardia in 2 and renal impairment in 1 patient. Overall mortality was 11.1% (Table IV).

**Table IV**

*Complications of the cases who underwent BAS*

Complications	Frequency	%
Sepsis	10	37
Cardio-respiratory arrest	2	7.41
Transient bradycardia	2	7.41
Renal impairment	1	3.70
Balloon rupture	0	0
Mortality	3	11.1

## Discussion

The natural history of transposition of the great arteries between 1957 and 1964 was reported by Leibman J et al based on a total of 742 cases. The area of study was the State of California and data were obtained from 14 selected hospital centers. For the whole group the age of death was as follows: by 1 week, 28.7%; by 1 month, 51.6%; and by 1 year, 89.3%.<sup>8</sup>

In neonates with TGA, BAS offers multiple benefits along with added advantages of simplifying the technical aspects of the arterial switch procedure, by allowing efficient left- and right- sided drainage on cardio pulmonary bypass (CPB) with a single atrial cannula. When introduced, the technique of BAS was

the most important single factor influencing survival in patients with TGA. Creating a nonrestrictive atrial communication optimizes mixing at the atrial level, improving systemic arterial oxygen content and cardiac output as well as lowering left atrial pressure. These beneficial effects often lead to a rapid stabilization of these neonates and improve their condition before they undergo a major neonatal operation.<sup>9</sup>

BAS is a life-saving procedure for patients born with d- TGA. The use of prostaglandins, however, has reduced its need in patients with restrictive atrial septal defects with poor mixing of saturated atrial blood whose preductal saturations cannot be maintained in an acceptable range or in those whose surgery will be delayed for some reason.<sup>10</sup> Prolong use of prostaglandin before arterial switch operation renders the tissue friable and put the surgeons on great difficulty in stitching. Baseline SPO<sub>2</sub> was significantly improved immediately after the procedure & at discharge in all our patients with BAS consisting with other studies all over the world.

Size of Interatrial defect increased significantly as well as pressure gradient across left & right atrium decreased significantly shown in our study was similar to Matter M et al.<sup>11</sup>

We agree with Baker et al<sup>12</sup> and Perry et al<sup>13</sup> that full cardiac catheterization is not required for the initial diagnosis or assessment of neonates with d- TGA and BAS is best performed in the neonatal intensive care unit under 2D control. Although most of our babies underwent BAS in cath lab under fluoroscopy and echocardiographic guidance except one who underwent BAS successfully at bedside under Echocardiographic guidance only. This is because we have performed coronary angiogram in most patients to accurately inform surgeon of coronary artery anomaly. In our series coronary angiogram could be done in 16 patients of BAS & significant coronary anomalies found in 2 patients.

In the initial period after the introduction of BAS, a number of neurologic complications were noted primarily relating to balloon rupture and embolization of balloon fragments but, over the period of time, they have been minimized due to introduction of newer equipment and techniques.<sup>14</sup> Recently, some reports have again highlighted this correlation. McQuillen et al<sup>15</sup> have shown that preoperative brain injury occurs in 40% of patients with TGA who

undergo BAS. They have found BAS to be an independent risk factor for acquired preoperative brain injury in term neonates with TGA. In our study we have not observed any neurological complication. Two out of three patients died because of sepsis & one due to sudden cardiac arrest during the procedure.

For a successful BAS thin septum primum is essential. So the procedure yields best results if it performed before 2-3 weeks of age. Leanage R et al showed at all points on the survival curve, patients having balloon atrial septostomy during the first week of life did best, followed by those having balloon atrial septostomy at the age of over 1 month. Patients having balloon atrial septostomy between 1 week and 1 month of age did worst of all.<sup>16</sup> Our study did not find any relationship between age of procedure below 20 days to saturation value over 60% at discharge.

### Conclusion

Till today, total correction by the arterial switch procedure is routine in neonates with d-TGA in developed country. But country like Bangladesh with few dedicated & competent surgical team & lack of logistics, BAS remains a time-honoured procedure for this subset of patients with d-TGA and restrictive interatrial communication. Thus, for the time being, BAS remains a justifiable clinical option in this group of patients. BAS is safe and effective palliative procedure for different CHD with good immediate results in our institution.

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## ORIGINAL ARTICLE

# Immediate Outcome of Pneumonia in 2 to 12 Months Old Infants with Congenital Heart Disease: A Case Control Study

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## Abstract

**Background:** Infants with congenital heart disease (especially with left to right shunt) have repeated respiratory tract infections which gradually evolve to pneumonia. Because of increased awareness of parents and provision of better diagnostic tools, these infants are being identified of having congenital heart disease early in life. But pneumonia remains as a great challenge for their survival before corrective procedure for their cardiac defects.

**Objective:** To ascertain the outcome of pneumonia in 2 to 12 months old infants with congenital heart disease in comparison to the infants without congenital heart disease of same age.

**Methods:** The exploratory case control study was conducted in Dhaka Shishu (Children) Hospital from July 2011 to December 2011. One hundred and thirteen (113) infants aged 2-12 months with pneumonia with congenital heart disease were enrolled who were compared with another 113 age and sex matched infants with pneumonia but without congenital heart disease. Outcome measures compared, were total no. of complications, non-resolving pneumonia, persistent fever, septicemia, duration of hospital stay, death and DORB (discharged on risk bond).

**Results:** Complication during hospital stay in 2-12 months old infants with pneumonia with congenital heart disease was 32% higher than pneumonia without congenital heart disease ( $P=0.001$ ). Among the complications non resolving pneumonia and septicemia were statistically significant ( $P=0.04$  and  $0.016$  respectively). Duration of hospital stay was also significantly longer ( $P=0.001$ ) in cases than control group.

**Conclusions:** Complication of pneumonia in 2-12 months old infants with congenital heart disease were significantly higher than infants of same age without congenital heart disease.

**Keywords:** Pneumonia, Congenital heart disease.

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## Introduction

Congenital heart disease is the commonest of all congenital lesions and is the most common type of heart disease among children.<sup>1</sup> The incidence of congenital heart disease is approximately 8 per 1000 live birth.<sup>2-3</sup> It is an important cause of childhood morbidity and mortality. One third of the patients require intervention in the 1<sup>st</sup> year of life, 75% of the cases survive beyond the 1<sup>st</sup> year of life if properly treated and can lead nearly normal life thereafter.<sup>3,4</sup>

On the other hand, pneumonia is the major cause of death of children younger than 5 years of age worldwide accounting for 4.3 million deaths per year or 10,000 per day.<sup>8-9</sup> Pneumonia is one of the leading causes of morbidity, hospitalization and death in children under five in Bangladesh and many other developing countries.<sup>8</sup> In developing countries like Bangladesh, there are several risk factors for pneumonia. Congenital anomalies including congenital heart disease is one of them.<sup>8,9</sup>

Control program ? includes identification of children with pneumonia by clinical features (rapid respiration and difficulty in breathing) and administration of antimicrobials with a presumption that majority of pneumonia in developing countries are because of bacterial pathogens. For further impact on morbidity and mortality, a thorough knowledge of the determining factors affecting the outcome of the disease is important. Factors associated with increased mortality include young age of the mothers and lack of proper education of the father, young age of the child, late hospitalization with cyanosis, altered sensorium, grunting, associated chest indrawing, hepatomegaly, acute malnutrition, inability to drink, associated loose stools, heart disease, anemia, rickets and lack of breastfeeding.<sup>9</sup>

Infants with congenital heart disease (especially with left to right shunt) have repeated respiratory tract infections which gradually evolve to pneumonia. These children are frequently admitted to hospital for pneumonia and most of the time diagnosed of having congenital heart disease incidentally. Because of increased awareness of parents and provision of better diagnostic tools, these infants are being identified of having congenital heart disease early in life. But pneumonia remains as a great challenge for their survival before corrective procedure for their cardiac defects. This study was conducted to find out the course, complications and prognosis of pneumonia in infants with congenital heart disease.

## Methods

An exploratory case control study was conducted at Dhaka Shishu (Children) Hospital, Dhaka,

Bangladesh from July 2011 to December 2011. 100 Infants aged 2-12 months old with pneumonia and congenital heart disease were compared with another 100 age matched infants who had pneumonia but no congenital heart disease.

Prior to the commencement of this study ethical permission was taken from ethical review committee of Dhaka Shishu (Children) Hospital.

Pneumonia was diagnosed by history (breathlessness, cough, fever, feeding problem etc.); clinical findings (fever, tachypnoea, tachycardia, chest indrawing, crepitations) and confirmed radiologically. Additionally CBC, Blood C/S findings were done to establish etiology.

Congenital heart disease was identified by suggestive history (recurrent chest infection, poor weight gain, feeding problem, cough and respiratory distress); physical findings were presence of cardiac murmur, Presence of cyanosis, feeding difficulty, features of congestive heart failure (tachycardia, tachypnea along with shifting of apex beat and hepatomegaly) and failure to thrive. Investigations were X-ray chest, ECG and confirmed by Echocardiography.

Infants below 2 months of age were not included because outcome of pneumonia in neonates is influenced by other factors. Infants with prematurity, association with other congenital anomaly, chromosomal anomaly, severe acute malnutrition, neurodevelopmental disabilities were excluded.

The cases were enrolled after taking written consent, demographic data was obtained and the child was followed up every 3 days interval up to discharge, referral or death.

The definition of persistent fever for this study included documented daily temperature  $>38^{\circ}\text{C}$  (shell temperature) without any fever free day for  $>10$  days after hospitalization though in few other studies the duration was  $>14$  days.<sup>10</sup> Non resolving pneumonia was defined as cough and fast breathing persisted for  $>10$  days of antibiotic therapy with no or little radiological improvement.<sup>11</sup> Septicemia was defined as SIRS(systemic inflammatory response syndrome) in the presence of or as a result of suspected or proven infection.<sup>12</sup>

All the data were noted in a preformed data sheet with structured questionnaire. Data analysis was done by using SPSS version 12. Unpaired *t* 'test' was applied for continuous data. Chi-square test (without Yates correction) was applied for rest of the categorical data.

## Results

A total of 226 patients were included in this study. They were divided in 3 groups according to their

age. Majority age was found to be 6-10 months in both groups, which was 72 (64.0%) in group I and 58 (52.0%) in group II. The mean age was found  $5.55 \pm 2.18$  months and  $5.86 \pm 3.38$  months in group I and group II respectively. The mean age difference was not statistically significant ( $P > 0.05$ ) between two groups in unpaired t-test.

Male was found 58 (51.0%) in group I and 75 (66.0%) in group II. Female was found 55 (49.0%) and 38 (34.0%) in group I and group II respectively. Male female ratio was 1.5:1. The male female difference was statistically significant ( $P < 0.05$ ) in group II in chi square test. (Table I)

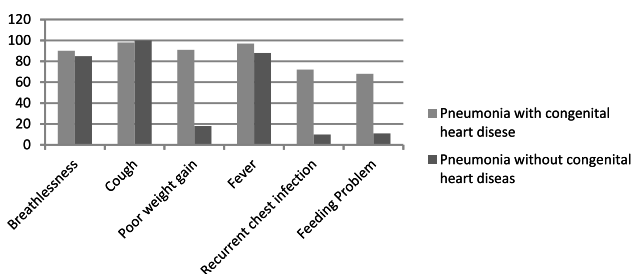
**Table I**

*Age, Sex, Socioeconomic status, exclusive breast feeding & Body weight of the study patients (n=226). (baselin characteristic of case and control)*

Items	Case (n=113)		Control (n=113)		P value
Age in months	n	%	n	%	
2-5	41	36.0	47	42.0	
6-10	72	64.0	58	52.0	
10-12	0	0.0	8	6.0	
Mean $\pm$ SD	$5.55 \pm 2.18$		$5.86 \pm 3.38$		0.441 <sup>ns</sup>
Range (min-max)	(2-9)		(2-12)		
Sex					
Male	58	51.0	75	66.0	0.04
Female	55	49.0	38	34.0	
Socioeconomic status					
Poor	25	22	40	35	0.041
Middle	77	68	67	59	0.186
High	11	10	7	6	0.297
Exclusive breast feeding	29	26	48	42	0.035
Body weight(kg)					
Mean $\pm$ SD	$4.61 \pm 1.43$		$5.38 \pm 2.14$		0.003
Range (min-max)	(4-7)		(5-9)		

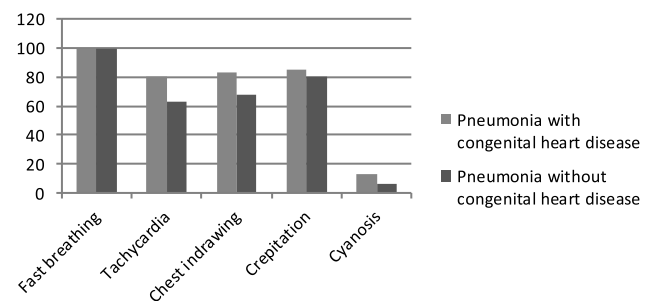
ns=not significant,  $\chi^2$  for qualitative data, t test for quantitative data  $p < 0.05$  =significant

Poor weight gain, fever, recurrent chest infection, feeding problem was statistically significant ( $P < 0.05$ ) between two groups in chi square test. (fig:1)



**Fig 1** Percentage distribution of study patients according to symptoms (n=226)

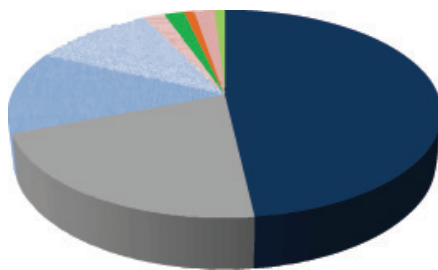
Dyspnea, tachycardia and chest indrawing were statistically significant but other physical findings were not statistically significant ( $P > 0.05$ ) between two groups in chi square test. (Fig.2)



**Fig 2** Percentage distribution of study patients according to signs (n=226)

Chest X-ray of the study patients showed patchy opacity of lung fields in 96(85.0%) in group I and 102(90.0%) in group II. Consolidation was found in 17(15.0%) and 11(10.0%) in group I and group II respectively which was significantly higher in group 1 ( $P=0.008$ ). Pleural effusion was found 2(2.0%) in group II but not found in group I. Cardiomegaly was found in 99(88.0%) cases of group I and 8(8.0%) cases of group II which was statistically significant between two groups in chi square test.

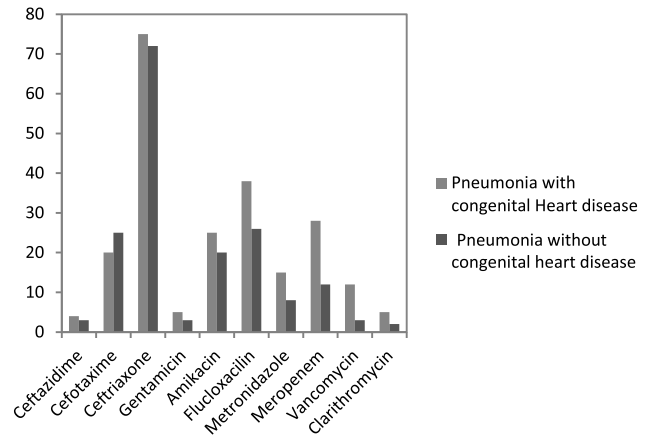
Perimembranous VSD was found in 53 patients (47.0%), 23 (20.0%) had ASD, 14 (12.0%) had PDA, 12 (11.0%) had VSD with pulmonary HTN. Other results are depicted in the figure. (Fig 3)



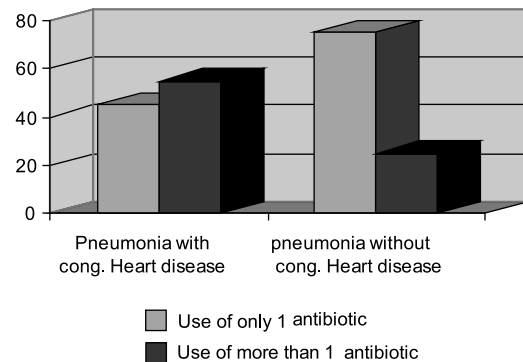
Perimembranous VSD  
ASD  
PDA  
VSD with pulmonary HTN  
Muscular VSD  
VSD with pulmonary stenosis  
VSD with ASD with PAH  
AV canal defect

**Fig 3** Distribution of the study patients according to echocardiographic finding in cong. heart disease ( $N=113$ )

Patients in both case & control groups were treated with various antibiotics. Among them Inj. Meropenem and Inj vancomycin were used in significantly higher number of patients in Group I than those in Group II ( $P$  value 0.004 & 0.016 respectively). Use of more than one antibiotic was found 30.0% higher in group I than group II. The difference was statistically significant ( $P=0.03$ ). between two groups in chi square test: (Fig 4)



**Fig 4** Use of antibiotics in study patients. ( $n=226$ )



**Fig 5** Percentage distribution of study patients according to use of more than 1 antibiotic

Complications during hospital stay were found in 54 cases (48.0%) in group I and 18 cases (16.0%) in group II. Non resolving pneumonia was found in 20(18.0%) and 05(4.4%) in group I and group II respectively which was significantly higher in group I( $P=0.001$ ). Persistent fever was found in 7(6.0%) in group I and 2(2.0%) in group II. Septicemia was found in 16(14.0%) and 8(7.0%) in group I and group II respectively. Heart failure occurred in 30 (26.54%) patients in group 1 and in 5 patients (4.42%) in group 2. Respiratory failure occurred in 2 patients in group 1 (1.76%) and in 1 patient in group II. Complication during hospital stay was 32% higher in group I than group II. Among the complications non resolving pneumonia, heart failure and septicemia were significantly higher in group I than group II ( $P$  value 0.001, 0.001 and 0.04 respectively).

**Table II**  
*Distribution of patients according to complications (n=226)*

Complications	Group I (n=113)		Group II(n=113)		P value
	n	%	n	%	
<b>Complications during hospital stay</b>					
Yes	54	48.0	18	16.0	0.001 <sup>s</sup>
No	59	52.0	95	84.0	
<b>Name of the complications:</b>					
<b>Non resolving pneumonia</b>					
Yes	20	18.0	05	4.4	0.001 <sup>ns</sup>
No	93	82.0	108	95.6	
<b>Persistent fever</b>					
Yes	07	6.0	02	2.0	0.148 <sup>ns</sup>
No	106	94.0	111	98.0	
<b>Heart failure</b>					
Yes	30	26.54	5	4.42	0.001 <sup>s</sup>
No	83	73.46	108	95.58	
<b>Septicemia</b>					
Yes	16	14.0	8	7.0	0.046 <sup>s</sup>
No	97	86.0	105	93.0	
<b>Respiratory failure</b>					
Yes	2	1.76	1	0.88	0.56 <sup>ns</sup>
No	111	98.2	112	99.1	

The mean duration of hospital stay was  $14.5 \pm 10.5$  days in group I and  $11.5 \pm 8.5$  days in group II. Among them cured patients stayed for  $7.5 \pm 3.5$  days and complicated patients stayed for  $17.5 \pm 7.5$  days in group I. In group II cured patients stayed for  $5 \pm 2$  days and complicated patients stayed for  $11.5 \pm 3.5$  days. DORB was found 2(2.0%) in group II but not found in group I. Two patients died (1.8%) in group I and two (1.8%) in group II. Though duration of hospital stay was longer in group 1 in both cured and complicated patients, the difference was not statistically significant in unpaired t test.

Furthermore cured from pneumonia and discharged with medical management with cardiac medicine was found in 95 patients (84.0%) in group I and 99 patients (84%) were cured and discharged without requirement for further follow up in group II. However 16 patients (14%) in group 1 and 10 patients (9.1%) in group II were recovered but not fully cured and discharged on request with oral medication and advised for follow up. These findings were not also statistically significant. Advice for immediate intervention was found in 42(37.0%) patients in group I.

**Table III**  
*Distribution of patients according to outcome variables (n=226)*

Duration of hospital stay	Group I(n=113)		Group II(n=113)		P value
	Days		Days		
All patients					
Mean± SEM	14.5±10.5		11.5±8.5		<sup>a</sup> 0.84 <sup>ns</sup>
Range (min-max)	(4-25)		(3-20)		
Cured patients	7.5±3.5(5-10)		5±2(3-7)		<sup>a</sup> 0.86 <sup>ns</sup>
Complicated patients	17.5±7.5(10-25)		11.5±3.5(8-15)		<sup>a</sup> 0.56 <sup>ns</sup>
Other parameters	Group I (n=113)		Group II(n=113)		P value
	n	%	n	%	
Cured at discharge					
Yes	95	84	99	88	<sup>b</sup> 0.445 <sup>ns</sup>
No	18	16	14	12	
DOR					
Yes	16	14	10	8.9	<sup>b</sup> 0.211 <sup>ns</sup>
No	97	86	103	91.1	
DORB					
Yes	0	0	2	2.0	<sup>b</sup> 0.248 <sup>ns</sup>
No	113	100	111	98.0	
Death					
Yes	2	1.8	2	1.8	<sup>b</sup> 1.000 <sup>ns</sup>
No	111	98.0	111	98.0	

## Discussion

A total of 226 patients were included in this study. Poor weight gain, fever, recurrent chest infection, feeding problems were significantly higher in the cases as these are common problems in infants with congenital heart diseases<sup>3,4,6</sup> Exclusive breast feeding up to 6 months was 16% higher in group II and formula feeding was 22% higher in group I which were statistically significant. This result may be due to the fact that infants with congenital heart disease usually are poorly able to breast feed. Though another study<sup>14</sup> suggest that given support and education necessary to initiate and maintain lactation, mothers can successfully breastfeed their infants with CHD for appropriate duration.

Patients from poor socioeconomic background were significantly higher ( $P=0.041$ ) in group II than group I. This is may be due to many poor patients with congenital heart disease remain undiagnosed and untreated and die after birth or early infancy due to

complication before hospitalization. The mean body weight was significantly lower ( $P>0.05$ ) in group I as infants with acyanotic congenital heart disease mostly suffer from failure to thrive which is shown in most of the studies.<sup>15,16</sup>

Cyanotic spell was not found in any of the groups. Probability is, usually cyanotic spells occur in congenital cyanotic heart disease & there was only 1 pink TOF patient in the study group. Dyspnea, tachycardia and chest indrawing were significantly higher in cases probably because heart failure was more in the case group.

Previous hospitalization of patients due to pneumonia was significantly higher in group I (94%) than group II (57%). Which means infants with congenital heart disease need frequent hospitalization for pneumonia.

Haemoglobin (Hb) level was significantly low ( $P<0.05$ ) in group I. This result was similar to other study<sup>17</sup> which showed high incidence of anemia in

0.5-5 years old infants with acyanotic cong. heart disease. Blood C/S showed 1 case of *S.pneumoniae* & 1 case of *H.influenzae* in group I and 1 case of *streptococcus pneumoniae* in group II.

Pneumonia etiology studies that incorporated viral studies showed that respiratory syncytial virus is the leading viral cause, in the prospective microbiology-based studies, viral causes of pneumonia are identified by rapid diagnostic tests (such as indirect immunofluorescence, enzyme-linked immunosorbent assay, polymerase chain reaction, viral culture on upper respiratory secretions – such as in nasopharyngeal aspirates – or by viral serology in paired samples).<sup>9</sup> These investigations could not be done in this study and so viral or fungal causes of pneumonia were not confirmed.

Patients were treated with various antibiotics. Among them Inj. meropenem and Inj vancomycin were used in significantly higher number of patients in Group I compared with Group II (P value 0.004 & 0.016 respectively). These drugs are usually used as 2<sup>nd</sup> line drugs when there is no improvement with cephalosporin and aminoglycosides because of their extensive coverage against bacteria. As complications like nonresolving pneumonia, heart failure and septicemia were more in group 1 so use of these drugs were also increased in this group. Use of more than one antibiotic was also found significantly higher in cases (30.0%).

Complications during hospital stay were found in 54 patients (48.0%) in group I and 18 patients (16.0%) in group II. Non resolving pneumonia was found in 20(18.0%) and 05(4.4%) in group I and group II respectively which was significantly higher in group 1 (P=0.001). Persistent fever was found in 7(6.0%) patients in group I and 2(2.0%) in group II. Septicemia was found in 16(14.0%) and 8(7.0%) in group I and group II respectively.

Heart failure was significantly higher (26.54%) in cases. Though this heart failure in group I is mainly because of excessive pulmonary blood flow and pulmonary hypertension in congenital heart disease with left to right shunt.

Respiratory failure occurred in 2 patients in group 1 (1.76%) and in 1 patient in group II. The number of patients with respiratory failure was small in both groups because the patients with this complication got admitted directly to ICU or referred to other

hospital for ventilator support from OPD or emergency department.

Complication during hospital stay was 32% higher in group I than group II. Among the complications non resolving pneumonia, heart failure and septicemia were significantly higher in group I than group II (P value 0.001 and 0.04 respectively).

The range of duration of hospital stay was quite wide in both groups because some patients took DORB on 2<sup>nd</sup> day while some patients were complicated and took longer period for recovery. The mean duration of hospital stay was 14.5 days in group I and 11.5 days in group II which were not statistically significant. Cured patients stayed for 7.5± 3.5 days and complicated patients stayed for 17.5±7.5 days in group I. In group II cured patients stayed for 5 ±2 days and complicated patients stayed for 11.5±3.5 days. Two(2) patients were discharged on risk bond in group II but not found in group I. Two (2) patients died (1.8%) in group I and 2(1.8%) in group II. Though duration of hospital stay was longer in group 1 in both cured and complicated patients, the difference was not statistically significant. Ninety five (95) patients (84%) were cured from pneumonia and discharged with medical management with cardiac medicine & advice for further follow up in group I. 99 patients (84%) were cured and discharged without requirement for further follow up in group II. However 16 patients (14%) in group 1 and 10 patients (9.1%) in group II were recovered but not fully cured and discharged on request with oral medication and advised for follow up. These findings were not also statistically significant. Advice for immediate intervention was found in 42(37.0%) patients in group I.

The cause of death in 1 patient in group I was VSD with pneumonia with heart failure with septicemia with multi organ failure. This patient died at ICU after 10 days of admission. Another patient died due to VSD with pneumonia with heart failure on 7<sup>th</sup> day of admission. One(1) patient died in group II due to pneumonia with septicemia with shock on 2<sup>nd</sup> day of admission. Another patient in group II died due to pneumonia with respiratory failure. This patient was admitted to ICU on the day of admission and needed ventilator support and died 5 days after admission. One patient was given DORB on 2<sup>nd</sup> day of admission in group II because of personal reasons.

The strength of the study was its prospective nature and having two groups for comparison. Further studies need to be done to compare between pre-operative and post-operative outcome of pneumonia with congenital heart disease.

### Conclusions

Complication like non resolving pneumonia, heart failure and septicemia in 2-12 months old infants with pneumonia with congenital heart disease were significantly higher than infants of same age with pneumonia but without congenital heart disease. However, days needed for recovery from pneumonia was not significantly different between two groups.

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## ORIGINAL ARTICLE

# Role of early detection of predictors of poor seizure outcomes change the course of neurodevelopment

Shayla Imam Kanta<sup>1</sup>, Rizwanul Ahasan<sup>2</sup>, Humaira Rafiq Quaderi<sup>3</sup>, Naila Zaman Khan<sup>4</sup>

## Abstract

**Background:** Epilepsy is the most common neurological disorder in children. In a previous study in Bangladesh Banu et al (2003) showed three factors, i.e., multiple seizure types, cognitive deficits and abnormal EEGs, to be predictors of poor seizure remission, with a view of developing an appropriate system of management in a resource constrained country like Bangladesh. Another study by Banu et al (2010) included motor disability as another clinical predictor. Both these studies provide important indicators for multidisciplinary services as part of epilepsy management programme.

**Objectives:** To determine if early identification of known predictors of poor seizure outcome influences the course of the seizures and of neurodevelopmental outcomes.

**Methods:** A retrospective study of children attending the Child Development Center, ie, a multidisciplinary service, in Dhaka Shishu Hospital. All children >2 years of age at first attendance to the Epilepsy Clinic between 2010-2012 with follow-up records including EEGs, were included. Patients were categorized in 2 groups, without poor predictors & with poor predictors. Initial seizure semi logy, age, sex, family history, EEG findings, neurodevelopmental profiles were observed and neurodevelopment assessment after stimulation and treatment were compared with initial finding.

**Results:** Total 45 patients were taken according to inclusion & exclusion criteria among them 9 patients were without poor predictors and 36 patients were with poor predictors. Among the patients female were 40% & male were 60%. 82% patients had idiopathic epilepsy and 12% patients had symptomatic epilepsy. Age below 12 months was 15 & above 12 months was 30 family history of epilepsy was found in 6 patients, Birth history was eventful in 21 patient and uneventful in 24 patients, seizure semiology revealed patients age of onset, recurrence rate , immediate post ictal period had significant association with patients with poor predictors. developmental domains in patients with or without poor predictors categorized in normal, mild, moderate and in severe form. In gross motor, fine motor, speech, behavior, and seizure shows significant p value in patients with poor predictors which in subsequent follow up showed much improvement in impairment grading .

**Conclusion:** So early identification of predictors of poor seizure outcome influence the course of the seizures and neurodevelopmental outcomes which subsequently improved due to early stimulation and therapy.

**Key words:** EEG, Epilepsy, Neurodevelopment, RNDA

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## Introduction

Epilepsy is the most common neurological disorder in children. 80% of affected children live in countries with limited resources where 90% of epilepsy is not consistently treated.<sup>1</sup> According to the 2014 definition of Epilepsy by ILAE At least two unprovoked (reflex) seizures occurring greater than 24 hours apart or one unprovoked (or reflex) seizures & a probability of further seizures similar to the general recurrence risk (at least 60%) after two unprovoked seizures, occurring over the next 10 years. Diagnosis of epilepsy syndrome Epilepsy is considered to be resolved for individuals who had an age dependent epilepsy syndrome but are now past the applicable age or those who have remained seizure free for the last 10 years with no seizure medicines for the last 5 years.<sup>2</sup> In Bangladesh epidemiological surveys confirm that seizure disorders are common, one study showing a prevalence rate of 68 out of every 1000 for 'any seizure history' and 9 out of every 1000 for 'any unprovoked seizure', in children aged 2 to 9 years.<sup>3</sup> In a previous retrospective study profile of childhood epilepsy was described where three predictors remained most significant: multiple seizure type ( $p < 0.001$ ), cognitive impairment ( $p < 0.01$ ), and abnormal EEG ( $p < 0.01$ ).<sup>4</sup>

The management of childhood epilepsy in countries with limited resources has several specific problems. These include lack of medical resources, misunderstanding of the nature of epilepsy, stigmatization, large treatment gaps (i.e. a high percentage of the target population not receiving medication but often a high level of use of traditional healers), and a lack of understanding of the priorities of the users by those seeking to provide treatment. All of these issues arise in the management of epilepsy in children in Bangladesh.<sup>5</sup>

So this study is conducted to view whether the predictors of poor outcomes of epilepsy influence the neurodevelopment of a child and to determine if early identification of known predictors of poor seizure outcome influences the course of the seizures and of neurodevelopmental outcomes.

## Materials and method

The study was conducted at the outpatient services of the child development centre in Dhaka Shishu (Children) Hospital. A child-health physician with basic training in child neurology, a developmental therapist trained in physiotherapy, occupational and

speech therapy within a developmental framework, were the core team members of the service. Informed written consent was obtained from all the parents or guardians. Reassurance was given to parents as there was no harmful effect for babies or economic loss. Clearance has been taken from the ethical committee for conducting the study.

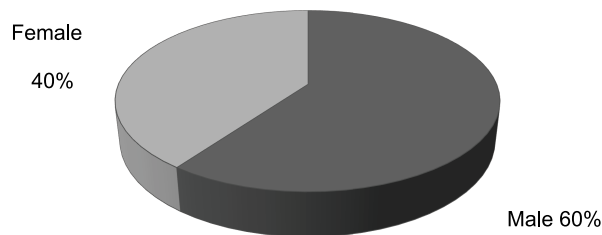
Children aged 1 month to 2 years, presenting with two or more unprovoked seizures with or without motor and D or other co morbidities, were included. General and neurodevelopment examination findings were collected on a pre-coded, structured form. Details of seizures were recorded as 'age at onset', which was categorized as 'early' when unprovoked seizures started at or before 12 months of age, or 'late'; 'seizure type(s)' were categorized as 'single' or 'multiple' type seizures when there was a history of more than one type of seizure such as generalized tonic-clonic and myoclonic or head-drops or absences; 'rate of seizures' was categorized as 'high' when there were one or more attacks per week<sup>4</sup>. Epilepsies were classified for descriptive purposes using the International League against Epilepsy syndromic classification current at the time of the study. Based on examination findings, EEG, seizures were categorized as 'generalized' or 'partial D focal', and epilepsies as 'idiopathic', 'symptomatic or cryptogenic' and 'unclassifiable'. A history of unprovoked, repeated seizures among the siblings, parents, grandparents, and paternal or maternal cousins was coded as a positive family history EEG findings were categorized as 'normal' and 'abnormal' for age and state of arousal. Abnormal brain activities were subcategorized for further descriptive analysis into (1) epileptiform discharges (discharges with spike, sharp, and slow wave complexes) with normal background activities between discharges, (2) non-epileptic background abnormality (diffuse or localized irregular slow waves, excessive beta waves, unorganized non-reactive background activities), and (3) both (epileptiform discharges with background abnormality).<sup>5</sup>

Neurodevelopment assessment was done by RNDA. The RNDA is reliable and valid for identifying specific neurodevelopment impairments including autism spectrum disorders, and when linked to appropriate interventions.<sup>7-9</sup> Total 45 patients with or without poor predictors that are multiple seizure types, abnormal EEG findings, poor cognitive development & positive family history were taken. Children above

2 years of age, No EEG done within assessment period & lost children in follow up, only single assessment were excluded from the study.

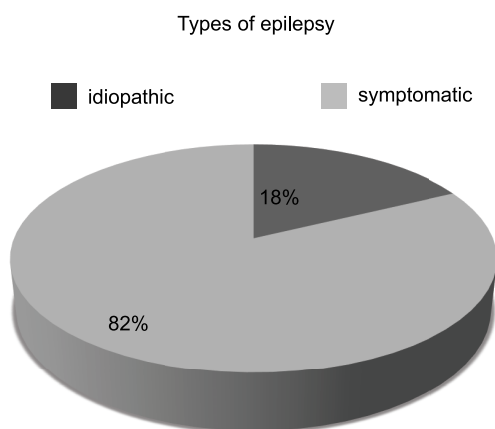
Follow up were reviewed retrospectively from master card of each patients records which is very systemically maintained in CDC. RNDA is used to determine functional status in the following domains gross motor, fine motor, vision, hearing, expressive language, cognition , behavior. The parameters of RNDA for each domain of the patients on discharge and follow up were recorded in the final sheet where impairment is graded 1=no, yes=2 and impairment or disability is graded as no=1 mild=2 moderate =3 and severe =4.

**Results**



**Fig 1** Distribution of sex among the study population

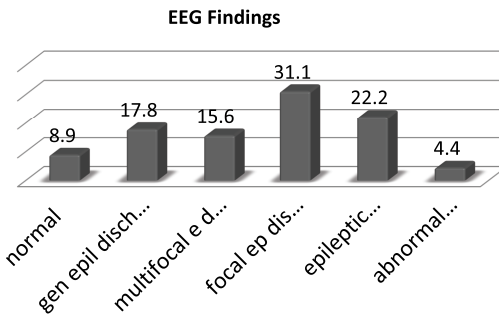
Figure shows distribution of sex among the study population 40% are female and 60% are male child



**Fig 2** Types of epilepsy

Etiologically 82% patients had symptomatic and 18% had idiopathic epilepsy.

**EEG Findings**



**Fig 3** EEG findings in 1<sup>st</sup> visit

In figure 3 EEG findings shows 8.9 % patients had normal EEG, 17.8% had generalized epileptiform discharge, 15.6 % had multifocal epileptiform discharge, and 31.1 % had focal epileptiform discharge, 22.2% epileptic encephalopathy, 4.4% abnormal background.

**Table I**

*Socio demographic characteristics of study population*

Domain		Total (45)
Age	< 12 months	15
	>12 months	30
Sex	Male	27
	female	18
Birth history	Eventful	21
	uneventful	24
Family history	Positive	6
	negative	39

Table I shows the age of the study population was less than 12 months 15 patients and more than 12 months 30 patients this age limitation and number of patients is very important for management purpose. Among the study population 27 patients male and 18 patients were female. Birth history was eventful in 21 patients and 24 patients are uneventful. Family history was positive in 6 patients and negative in 39 patients.

**Table II***Association and relation with seizure semiology (description of seizure)*

Domain		Without poor predictor	With poor predictor	Pvalue
Age of onset	<1 year	1(11.1%)	17(47.2%)	0.03
	>1year	8(88.9%)	19(52.8%)	
Duration of seizure	< 5 min	4(44.4%)	31(86.1%)	0.012
	>5 min	5(55.6%)	5(13.9%)	
Recurrence rate	High	5(55.6%)	33(91.7%)	0.017
	Low	4(44.4%)	3(8.3%)	
Immediate post ictal period (Drowsiness, unconscious, Abnormal behave Weakness of body)	Yes	1(11.1%)	17(47.2%)	0.03
	no	8(88.9%)	19(52.8%)	

In table II seizure semiology or description of seizures revealed patients < 1 year with poor predictors were 17 and > 1year 19 patents which are significant. Duration of seizure <5min with poor predictors 31 and >5 min in 5 patients in patients without poor predictors it is 4 & 5 which also shows significant

p value. High recurrence rate is in patients without poor predictors 5 and low in 4 patients. In patient with poor predictors recurrence rate is high in 33 patient and low in 3 patients. Immediate post ictal period indicates present in patients with poor predictors shows significant pvalue.

**Table III***Neurodevelopment assessment in patients with poor predictors shows different domains with epilepsy patients in 1<sup>st</sup> assessment*

	Normal	Mild	Moderate	Severe	Pvalue
Gross motor					
Without poor predictors	4(44.4%)	4(44.4%)	1(11.1%)	0(0%)	0.012
With poor predictors	2(5.6%)	15(41.7%)	17(47.2%)	2(5.6%)	
Fine motor					
Without poor predictors	3(33.3%)	6(66.7%)	0(0%)	0(0%)	0.0250
With poor predictors	2(5.6%)	18(50%)	14(38.9%)	2(5.6%)	
Vision					
Without poor predictors	4(22.2%)	5(55.6%)	2(22.2%)	0(0%)	0.98
With poor predictors	3(8.3%)	22(66.1%)	10(27.8%)	1(2.8%)	
Hearing					
Without poor predictors	4(44.4%)	4(44.4%)	1(11.1%)	0(0%)	0.65
With poor predictors	15(41.7%)	16(44.1%)	4(11.1%)	1(2.8%)	
Cognition					
Without poor predictors	3(33.3%)	5(55.6%)	1(11.1%)	0(0%)	0.97
With poor predictors	15(41.7%)	16(44.4%)	4(11.1%)	1(2.8%)	
Speech					
Without poor predictors	3(33.3%)	5(55.6%)	1(11.1%)	0(0%)	0.0280
With poor predictors	1(2.8%)	22(61.1%)	12(33.3%)	1(2.8%)	
Behavior					
Without poor predictors	3(33.3%)	5(55.6%)	1(11.1%)	0(0%)	0.0240
With poor predictors	1(2.8%)	20(55.6%)	14(38.9%)	1(2.8%)	
Seizure					0.05
Without poor predictors	1(11.1%)	7(77.8%)	1(11.1%)	0(0%)	
With poor predictors	1(2.8%)	13(36.1%)	12(33.3%)	10(27.8%)	

Pvalue&lt;.05 = Significant

**Table IV**  
*Neurodevelopmental assessment in different domains with epilepsy patients in final assessment*

Domain	Normal	Mild	Moderate	Pvalue
Gross motor	3(33.3%)	5(55.6%)	1(11.1%)	0.208
Without poor predictors	4(11.1%)	22(61.1%)	10(27.8%)	
With poor predictors				
fine motor				
Without poor predictors	3(33.3%)	6(66.7%)	0(0%)	.09
With poor predictors	4(11.1%)	22(61.1%)	10(27.8%)	
Vision				
Without poor predictors	2(22.2%)	5(55.6%)	2(22.2%)	0.65
With poor predictors	3(8.3%)	22(61.1%)	10(27.8%)	
Hearing				
Without poor predictors	3(33.3%)	6(66.7%)	0(0%)	0.385
With poor predictors	20(55.6%)	15(41.7%)	1(2.8%)	
Cognition				
Without poor predictors	3(33.3%)	6(66.7%)	0(0%)	.76
With poor predictors	12(33.3%)	22(61.1%)	2(5.6%)	
Speech				
Without poor predictors	3(33.3%)	6(66.7%)	0(0%)	0.97
With poor predictors	13(36.1%)	19(52.8%)	4(11.1%)	
Behavior				
Without poor predictors	3(33.3%)	6(66.7%)	0(0%)	0.474
With poor predictors	16(44.4%)	17(47.2%)	3(8.3%)	
Seizure				
Without poor predictors	3(33.3%)	6(66.7%)	0(0%)	0.176
With poor predictors	3(8.3%)	27(75%)	5(13.3%)	

Table III reflects the developmental domains in patients with or without poor predictors where findings in normal, mild, moderate and in severe form. In gross motor, fine motor, speech, behavior, and seizure shows significant p value in patients with poor predictors.

Table IV shows neurodevelopment status in different domains shows much improvement after treatment and therapeutic stimulation. There is no patient presented with severe impairment, moderate impairment in patients with poor predictors in different domains.

### Discussion

This study is conducted to ascertain the neurodevelopment outcome of patients with epilepsy by offering a hospital based multidisciplinary service. Epilepsy management is very much challenging for the patients of developing country like Bangladesh.<sup>5</sup>

In this study 40% child were female and male child were 60%, probably due to gender inequality of

seeking health care facilities. This has also similarities with other study.<sup>6, 11</sup>

EEG findings shows 8.9% patients had normal EEG, 17.8% had generalized epileptiform discharge, 15.6% had multifocal epileptiform discharge, and 31.1% had focal epileptiform discharge, 22.2% epileptic encephalopathy, 4.4% abnormal background. Similar findings has been found in other studies.<sup>10-12</sup> Etiologically 82% patients had symptomatic and 18% had idiopathic epilepsy.<sup>13</sup> Table I shows the age of the study population was less than 12 months 15 patients and more than 12 months 30 patients this age limitation and number of patients is very important for management purpose. Birth history was eventful in 21 patients and 24 patients are uneventful. Family history was positive in 6 patients and negative in 39 patients. Similar findings has been found in other study.<sup>13-14</sup>

In table II seizure semiology or description of seizures revealed patients < 1 year with poor predictors were 17 and > 1 year 19 patients which is

significant. Duration of seizure <5min with poor predictors 31 and >5 min in 5 patients in patients without poor predictors it is 4 & 5 which also shows significant p value. High recurrence rate is in patients without poor predictors 5 and low in 4 patients. In patient with poor predictors recurrence rate is high in 33 patient and low in 3 patients. Immediate post ictal period indicates present in patients with poor predictors shows significant p value .<sup>6,12-16</sup> Immediate post ictal period have to be considered as an important factor for management purpose. Table III reflects in patients with poor predictors significant P value in gross motor, fine motor, speech, behavior, and seizure in first follow up. These patients were treated and followed up regularly and last follow up showed significant improvement. No patient had severe impairment in last follow up and impairment grading also improved from severe to moderate, moderate to mild form. These findings should be kept under consideration for the management purpose.

### Conclusion

Early identification of predictor's poor seizure outcome influence the course of the seizures and neurodevelopmental outcomes which subsequently improved due to early stimulation and therapy.

### Limitation of study

Small sample size hospital based study, only referred patients were included. Retrospective information was analyzed in a tertiary care centre where families tend to bring only their severely impaired or disabled children, so it represents the severe end of the spectrum.

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## ORIGINAL ARTICLE

# Epidemiology and Outcome of Parapneumonic pleural effusion among the children admitted in Dhaka Shishu Hospital

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## Abstract

**Background:** Parapneumonic effusions (PPE) frequently occur as complications of pneumonia. Published data from developing countries is limited.

**Objective:** This study was conducted to describe the epidemiological characteristics, clinical findings and the treatment outcome of this disease.

**Methods:** A prospective study was done on 35 cases of parapneumonic pleural effusion from 2 months to 18 years of age who were admitted in Dhaka Shishu Hospital from May 2014 to September 2015. Diagnosis of pleural effusion was confirmed by chest radiography and aspiration of pleural fluid. All children were treated with parental antibiotics, chest tube or surgery according to British Thoracic Society guidelines.

**Results:** Positivity was higher in male children (77.42%). The most common age group affected was 1-5 years. Most common presenting complaints were fever (93.55%), breathlessness (87.10%) and cough (74.19%). All (100%) children had diminished chest movement, dull on percussion, diminished vocal resonance and diminished breath sound on the affected side. The majority of the pleural collections were on the right pleural space. About one-third of the parapneumonic effusions were empyemic. All children were treated with parental antibiotics and 10 patients required chest tube drainage. Remaining 11 patients underwent primary surgical management.

**Conclusion:** Parapneumonic pleural effusion was common in younger age. Antibiotic coverage and chest tube drainage was adequate in most cases. However, patients with extensive pleural involvement need surgery.

**Key words:** parapneumonic pleural effusion (PPE), empyema, vocal resonance, chest tube drainage

## Introduction

Pleural effusions (liquid in the pleural space), which occur less frequently in children than in adults, can be caused by a variety of infectious and noninfectious diseases. Pleural fluid accumulates in the pleural cavity whenever filtration exceeds the removal

mechanism and may be the result of increased filtration associated with impaired absorption or of normal filtration associated with inadequate removal.<sup>1</sup> Pleural effusions in children most commonly are infectious (50% to 70% parapneumonic effusion); congestive heart failure is a less frequent cause (5%

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to 15%), and malignancy is a rare cause.<sup>1</sup> Parapneumonic effusion is defined as fluid in the pleural space in association with pneumonia.<sup>2</sup> Nontuberculous bacterial pneumonia constitutes the most frequent origin of pleural effusion in children. Pneumonia can be complicated by an empyema, progressing from an exudative effusion, to a fibrinopurulent stage with loculations, and then organized with a thick fibrinous peel.<sup>3</sup> In various study it has been reported that, incidence of parapneumonic effusion ranges from 20% to 91% with an increase in morbidity and mortality.<sup>4</sup> Parapneumonic effusion is more common in boys than girls and more frequently encountered in infants and young children than in older children.<sup>5</sup> In a study it has been found that out of 176 children with pulmonary tuberculosis, from Spain, 39 (22%) patients had pleural effusion.<sup>6</sup> In a review of 74 patients with intrathoracic lymphomas, Chaginaud<sup>7</sup> found pleural effusion in 10 (71%) of 14 children with lymphoblastic lymphomas and in 7 (12%) of 60 children with non-Hodgkins lymphoma. This study was therefore conducted to describe the epidemiological characteristics, clinical findings and the treatment outcome of this disease.

### Materials and Methods

This prospective study was conducted in department of Pulmonology, Dhaka Shishu Hospital. Total 197 patients of either sex aged between 2 months and 18 years who had been diagnosed as pneumonia from May 2014 to September 2015 were selected for analysis. Among them 35 patients with pleural effusion confirmed by chest radiography and aspiration of pleural fluid were selected purposively. The exclusion criteria were very sick children, previously treated pleural effusion cases and any other chronic illness. At the time of admission, detailed history was taken and clinical examination was performed. Investigations like complete blood count, blood culture, Mantoux test, sputum for acid-fast bacilli (AFB) and chest X-ray and USG of chest were done. Pleural fluid was collected and subjected to cell count, protein, sugar, AFB and

Gram-stain, culture and Gene X-pert for AFB. Computed tomography (CT) scan chest was done whenever required. All information's were recorded in pre-tested semi-structured questionnaire. Ethical clearance was taken from institutional ethical committee. Treatment was done protocol was followed according to British Thoracic Society (BTS) guidelines. After diagnostic thoracocentesis and obtaining appropriate cultures, parenteral antibiotics were started and a chest tube was inserted in some cases. Antibiotics were changed if clinical response

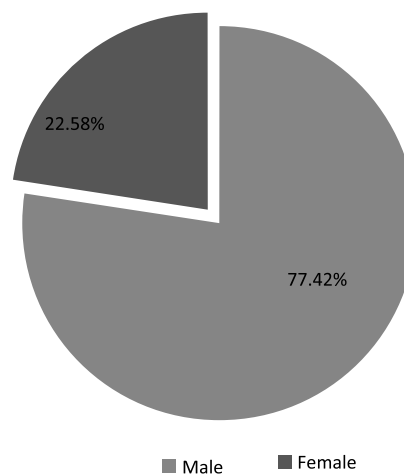
was inadequate. Repeat chest X-ray was done after treatment to see the signs of resolution. Loculated pus or marked thickenings of the pleura with encasement of the lung were better delineated on a CT scan as opposed to a chest radiograph. In these cases surgery were considered after failed tube drainage (TD). For surgery, patients were referred to National Institute of Chest Disease Hospital (NIDCH) and other hospitals also. Surgical decortications were done in most of the cases.

### Results

A total 35 patients were diagnosed as pleural effusion was selected in this study period. Among them 2 patients developed effusion due to tuberculosis and 2 patients due to lymphoma. These 4 patients were excluded from the study. Finally 31 patients were selected. Age distribution of cases, 6 (19.35%) were within 1 year, 15(48.39%) were between 1 to 5 years, 09(29.03%) were 6 to 10years, 1(3.23%) was above 10 years of age (Table-I).

Age (years)	Number	Percentage
< 1	06	19.35
1-5	15	48.39
6-10	09	29.03
> 10	01	3.23
Total	31	100

Positivity was higher in male children, 77.42% and 22.58% of female children was positive (Fig 1).



**Fig 1** Sex distribution of the studied children

Completely immunized were 31(100%). Most common presenting complaints were fever (93.55%), breathlessness (87.10%) and cough (74.19%), (Table II).

**Table II***Clinical presentation of studied children*

Presenting Features	Case	Percentage
Fever	29	93.55
Breathlessness	27	87.10
Cough	23	74.19

Regarding physical signs, all (100%) children had diminished chest movement, dull on percussion, decreased vocal resonance and diminished breath sound on the affected side. On the other hand 9 (25.7%) cases had mediastinal shifting (Table III).

**Table III***Physical signs of studied children*

Clinical signs	Number	Percentage
Diminished chest movement	31	100
Dullness on percussion	31	100
Diminished vocal resonance	31	100
Diminished breath sound	31	100
Mediastinal shifting	9	25.7%

The effusions were located on the right pleural space in 20 (64.52%) and on the left in 11 (35.48%) children (Table IV).

**Table IV***Site of pleural effusion among study population (n=31)*

Involvement	Number	Percentage
Right side	20	64.52
Left side	11	35.48

The effusions were purulent (empyema thoracis) in 10 (32.26%) and serosanguinous in 21 (67.74%) of the children. All of the patients were treated with

antibiotics. Among them 10 patients required chest tube drainage, 11 patients required primary decortications (Table V).

**Table V***Treatment of studied children*

Treatment	Number	Percentage
Antibiotic	31	100
Antibiotic+Chest drain	10	32.26
Antibiotic+Chest drain + Surgery	11	35.48

**Discussion**

This study reports similar epidemiological profile including age group, sex and clinical presentation of the disease as noted by other studies in India and Turkey.<sup>5,8-12</sup> Another study found that parapneumonic effusions are more common in boys than girls and most frequently encountered in infants and young children<sup>4</sup>. In this study fever, respiratory distress and cough were predominant presenting features. Restricted chest movement, dullness on percussion, diminished vocal resonance and diminished breath sound were common physical findings. Presenting features were similar to another study done in Ethiopia and Bangladesh.<sup>4,13</sup> It is noteworthy that the majority of the pleural collections were on the right pleural space. This is in agreement with reports by Gomez-Go et al<sup>14</sup> and Baranwal et al<sup>15</sup> among children in Nepal. The high incidence of right-sided pleural fluid collections may be related to the fact that the right main bronchus is shorter, wider and straighter in comparison to the left, facilitating the descent of the infection and/or causing foreign bodies to preferentially lodge there.<sup>15-18</sup> About one-third of the parapneumonic effusions found in this study were empyemic; this is similar to reports from other developing countries.<sup>9,14</sup> This may be related to the delayed presentation and the high pre-admission use of antibiotics which are often sub-therapeutic and substandard, or the use of unorthodox medicine, all of which allow the pathologic process to progress to the more advanced fibrinopurulent stage.<sup>15</sup> In this study, 18 children were presented with more than 10 days of illness found to have advanced stage of disease and among them 11 children were managed with surgical intervention, who showed loculated pus or marked

thickenings of the pleura with encasement of the lung on CT scan .Timely institution of proper management prevents the need

for any surgical intervention and avoids complications.<sup>19</sup> The optimal treatment of children with empyema remains controversial. The different modalities that are used when the child remains febrile despite adequate antibiotic treatment include chest drain.<sup>20</sup> The use of surgery for the treatment of childhood empyema has increased during the past few years. It can either be the primary procedure or implemented in cases of failure of treatment of chest drainage. <sup>21, 22</sup> In our study etiology was not identified and the number of sample size was small in relation to huge number of population. Long term outcome of the patients were not assessed.

### Conclusion

Parapneumonic pleural effusion was common in younger age. Antibiotic coverage and chest tube drainage was adequate in most cases. However, patients with extensive pleural involvement underwent surgery.

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## ORIGINAL ARTICLE

# Bubble Continuous Positive Airway Pressure (Bubble CPAP) in PTLBW Neonates with Respiratory Distress

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## Abstract

**Background:** Bubble continuous positive airway pressure is a well-established modality for providing ventilatory support to spontaneously breathing infants. It has been used in a variety of clinical situations.

**Objectives:** To evaluate the immediate outcome of PTLBW neonates with respiratory distress by using Bubble Continuous Positive Airway Pressure (BCPAP).

**Materials and Methods:** This Clinical Trial (Quasi-Experimental study) was conducted from 1<sup>st</sup> April 2013 to 30<sup>th</sup> September 2014 in the SCABU of Dhaka Shishu (Children) Hospital. Total 172 preterm babies with a gestational age <35 weeks, presented with clinical signs of respiratory distress were included in the study and among them 85 cases were taken as BCPAP group who were treated by bubble CPAP and 87 patients who could not be treated by BCPAP due to unavailability of BCPAP machine at that time were taken as control. Effects of BCPAP were assessed by comparing the clinical conditions, oxygen saturation and arterial blood gas status. Written informed consent from guardian was taken.

**Results:** Mean gestational age, weight and age at the time of admission were comparable in two groups (BCPAP vs Control: GA 31±1.61 vs 30±1.93 weeks, birth weight 1454±284 vs 1435±228gm, age 28±13 vs 31±19hrs). It was found that duration of O<sub>2</sub> need was significantly ( $p<0.05$ ) less in BCPAP group than the control group (5.61±1.61 vs 8.49±3.22 days), significantly ( $p<0.05$ ) less duration to improve respiratory distress (3.69±1.55 vs 7.67±2.76 days) and hospital stay (8.74±3.72 vs 12.67±11.75 days) was required in BCPAP group. Requirement of mechanical ventilation (IMV) was also less in BCPAP group (27.06% vs 51.72%,  $P<0.05$ ).

**Conclusion:** This study found that CPAP is an effective modality of treatment of PTLBW neonates with respiratory distress.

**Keywords:** Bubble CPAP, PTLBW, Respiratory distress

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## Introduction

Globally about 17% of all infants that are born alive each year have a low birth weight (LBW), defined as a body weight of less than 2,500 gm at birth (WHO, 1980, 1984). The rate of preterm birth for infants with less than 37 weeks gestational age has been steadily increasing since 1990 and is now at 12.5% or 1 of every 8 live births in the United States.<sup>1</sup> The proportion of low birth-weight infants, those who weight less than 2500 gm, has increased from 7.9% in 2003 to 8.1% in 2004.<sup>1</sup> Ninety percent (90%) of these infants are born in developing countries. Low birth weight is considered a major public health problem in populations where the prevalence is greater than 15% (ACC/SCN, 2000). Asia, and particularly South Asia, has a higher prevalence of LBW than any other continent in the world. In South Asia, the prevalence ranges from 15% to 30% (UNICEF & WHO, 2004). In Bangladesh, BBS/UNICEF country-wide survey in 2003-2004 revealed 36% of newborns are LBW.<sup>2</sup> The survey confirms that low birth weight is a major public health problem in Bangladesh. In developing countries, an increasing proportion (41%) of deaths of children less than 5 years of age occur in the neonatal period.<sup>3</sup> There are lot of complications of preterm low birth weight (PTLBW) among them respiratory distress is most common. Respiratory distress occurs in 0.96-12% of life births and is responsible for about 20% of neonatal mortality.<sup>4</sup> It is the most common presenting problem of newborns encountered within the first 48-72 hours of life<sup>4</sup> and remains the primary indication for admission to Neonatal Intensive Care Unit to combat respiratory failure.

For two decades, the standard treatment for very preterm infants was with assisted ventilation and surfactant. However, since ventilation may damage the lungs,<sup>5</sup> it has been hypothesized that the avoidance of ventilation might lead to less morbidity and mortality. Nasal continuous positive airway pressure is a well-established modality for providing ventilatory support to spontaneously breathing infants. It has been used in a variety of clinical situations.<sup>6</sup>

Nasal continuous positive airway pressure (NCPAP) is a modality that supports spontaneous ventilation by providing a positive airway pressure throughout the whole respiratory cycle.<sup>6</sup> CPAP is relatively inexpensive and can be easily taught, this could have the potential to be the optimal respiratory support

device to implement in developing countries.<sup>7, 8</sup> A wide variety of devices are used to deliver CPAP, including variable flow driver devices, single or binasal prongs where pressure is generated by a column of water (bubble CPAP) or a ventilator. Bubble CPAP (BCPAP) is appealing because of its simplicity and low cost. With this technique gas flows past the nasal device and the pressure is generated in the circuit by placing the distal limb of the CPAP circuit under a known depth of water that creates bubbles and pressure oscillations in the circuit.<sup>9</sup> Gas flow is increased until continuous bubbling is achieved. It has been suggested that use of BCPAP in the poorly compliant lung may promote lung volume recruitment and augment the efficiency of gas mixing.

CPAP delivered by underwater seal causes vibration of the chest due to gas flow under water; and these vibrations simulate waveforms produced by high frequency ventilation. Lee, et al.<sup>10</sup> demonstrated the superiority of bubble CPAP as compared to ventilator derived CPAP in premature infants. Bubble CPAP is also a less expensive method of respiratory support, most suitable to neonatal units with limited resources in developing countries.<sup>11</sup> Continuous positive airway pressure (CPAP), when applied to premature infants with Respiratory distress syndrome (RDS), re-expands collapsed alveoli, splints the airway, reduces work of breathing and improves the pattern and regularity of respiration.<sup>12</sup> Atelectrauma (repeated opening and collapse of the alveoli), biotrauma (intubation of the airway) and volutrauma (overstretching of the alveoli), the key determinants of ventilator induced lung injury are minimal or absent in gentler modes of ventilation such as nasal CPAP.<sup>13,14</sup> Bubble CPAP, when used appropriately, is more cost effective, less intensive, requires less training and has lower risk of complications. We conducted this clinical trial to evaluate the immediate outcome of PTLBW neonate with respiratory distress on Bubble CPAP.

## Materials and Methods

This Clinical Trial (Quasi-Experimental study) was conducted from 1<sup>st</sup> April 2013 to 30<sup>th</sup> September 2014 in the SCABU of Dhaka Shishu (Children) Hospital. Total 172 preterm low birth weight babies of less than 35 weeks gestation admitted within 72 hours of life with respiratory distress were included in the study. Among the 172 cases 85 were treated with

BCPAP and rest 87 patients who treated conventionally due to unavailability of BCPAP machine at that time were taken as control.

Patient with Gestational age <35 weeks admitted in the SCBU within 72 hours of life with respiratory distress were included. Babies requiring intubation at birth, severe perinatal asphyxia (APGAR <3 at 5 min) and those with major malformation were excluded. If patient parents refuse to continue treatment or patient was referred to other hospital due to unavailability of mechanical ventilation were also excluded from the study. Respiratory distress was documented by Fast breathing (R/R- >60/min) and any one of the followings: Low O<sub>2</sub> saturation (<87%), Retraction, Grunting, Nasal flaring and Severe chest indrawing. If the parents refused to give consent, were not included in the study. Eligible babies were started on Bubble CPAP with bi-nasal prongs (Fisher and Paykel Healthcare). PEEP was started at 5 cm of water and adjusted to minimize chest retractions. FiO<sub>2</sub> was adjusted to maintain SpO<sub>2</sub> between 87% and 95%. Flow was titrated to the minimum to produce continuous bubbling in the bubble chamber.

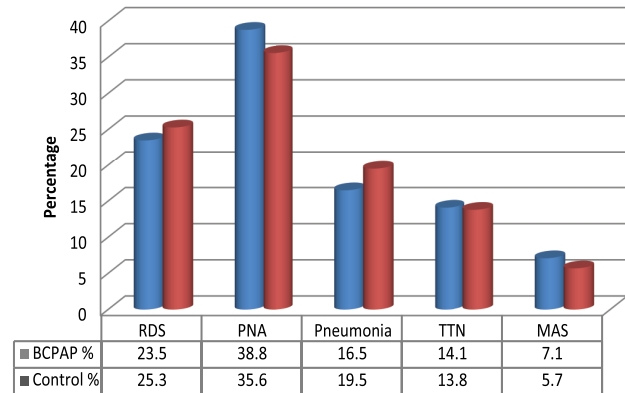
Bubble CPAP was considered to be successful if the respiratory distress improved and the baby could be successfully weaned off from CPAP. The criteria for weaning was absence of respiratory distress (minimal or no retractions and respiratory rate between 30 and 60 per minute) and SpO<sub>2</sub>>90% on FiO<sub>2</sub> <30% and PEEP <5 cm of water. Infants were diagnosed to have failed CPAP and were started on mechanical ventilation when they: (a) remained hypoxic, i.e. SpO<sub>2</sub><87% despite FiO<sub>2</sub>>70% and PEEP >7cm of water; (b) had severe retractions on PEEP >7cm of water; (c) had prolonged (>20 seconds) or recurrent apneas (>2 episodes within 24 hours associated with bradycardia) requiring bag and mask ventilation; and, (d) had severe metabolic acidosis or shock requiring inotropic support (dopamine and or dobutamine) >20µg/kg/min. Infants failing CPAP in the first 1 week of life were considered to be CPAP failures. Data collection of maternal variables included multiple births, pregnancy induced hypertension, preterm premature rupture of membrane, cesarean section and antenatal steroids. Gestational age was calculated based on mother's last menstrual period and or early pregnancy ultrasound scan or New Ballard score. Infant variables evaluated included birth weight, gestational age, Apgar score at 1minute, delivery room management (oxygen, bag and mask, intubation), X-ray chest, arterial blood gas and FiO<sub>2</sub> requirement. The other clinical data recorded are

patent ductus arteriosus (PDA) (clinical and Echo proven), pneumothorax, culture positive sepsis, pneumonia, necrotizing enterocolitis (NEC), chronic lung disease (oxygen requirement at 36 weeks PMA), germinal matrix – intraventricular hemorrhage (IVH), periventricular leucomalacia (PVL) (neurosonogram before day 7, at discharge and at 40 weeks PMA), retinopathy of prematurity (ROP) of any grade, duration of hospital stay among the survivors, and mortality. The study assessed the following outcomes: Duration of O<sub>2</sub> need, duration to improve respiratory distress, need for mechanical ventilation, duration of hospital stay, survive and discharge. Data were analyzed with the use of the Statistical Package for Social Science program (SPSS version 15.0). Data was compared between CPAP group with control group. The independent samples t-test was used for group comparisons of normally distributed variables. For comparisons of categorical data we used the Chi-square test. Statistical significance was determined at P value <0.05. The study was approved by the institute ethics committee and informed consent was obtained from either the father or a guardian.

## Results

Total 172 preterm low birth weight babies of less than 35 weeks gestation admitted within 72 hours of life with respiratory distress were included in the study. Among the 172 cases 85 were treated with BCPAP and rest 87 patients who treated conventionally due to unavailability of BCPAP machine at that time were taken as control. Mean gestational age, weight and age at the time of admission were comparable in two groups (BCPAP vs Control: GA 31±1.61 vs 30±1.93 weeks, birth weight 1454±284 vs 1435±228gm, time 28±13 vs 31±19hrs) (Table 1). Respiratory parameters like respiratory rate, chest indrawing, grunting and oxygen saturation were also comparable in both groups (R/R-70.87±6.05 vs 70.10±2.76, Gr-52 (61%) vs 51 (59%), C/I- 50(58%) vs 49(56%) and O<sub>2</sub> sat - 83.09±4.84 vs 82.37±6.62) (Table 1). Diagnosis of enrolled patients was RDS, PNA, TTN, Pneumonia and MAS and these were similar in both groups (Figure 1). There was significant (p<0.05) changes in arterial blood gas status in BCPAP group but not in control group (Table 2). There was significant (p<0.05) improvement of respiratory status i.e. decrease in respiratory rate (RR- 70.87±6.05 to 47.96±5.33) and increase oxygen saturation (SpO<sub>2</sub>- 83.09±4.84 to

94.24±4.97) at 48 hours after BCPAP intervention but no significant ( $p>0.05$ ) changes in control group (Table 3). It was found that duration of  $O_2$  need was significantly ( $p<0.05$ ) less in BCPAP group than the control group (5.61±1.61 vs 8.49±3.22 days), significantly ( $p<0.05$ ) less duration to improve respiratory distress (3.69±1.55 vs 7.67±2.76 days) and hospital stay (8.74±3.72 vs 12.67±11.75 days) was required in BCPAP group. Requirement of mechanical ventilation (IMV) was also less in BCPAP group (27.06% vs 51.72%,  $P<0.05$ ) (Table 4). It also showed that survived and discharged in BCPAP group was 65 (76.47%) and was statistically significant ( $P<0.05$ ) (Table 4).



**Fig 1** Distribution of diagnosis pattern among BCPAP and control groups of PTLBW neonates having respiratory distress (n=172)

**Table I**

*Baseline characteristics of neonates in both BCPAP and control groups (n=172)*

Baseline Characteristics	BCPAP Group (N=85) (mean±sd)	Control Group (N=87) (mean±sd)	P* value
Age on admission in Hours	28±13	31±19	0.24
Weight on admission in gm	1454±284	1435±228	0.62
Gestational age in wks	31±1.61	30±1.93	0.15
respiratory rate	70.87±6.05	70.10±2.76	0.28
Grunting [N (%)]	52 (61%)	51 (59%)	0.42**
Chest indrawing [N (%)]	50 (58%)	49 (56%)	0.76**
Oxygen saturation (%)	83.09±4.84	82.37±6.62	0.41
PH	7.20±0.05	7.18±0.07	0.08
PCO <sub>2</sub>	31.38±8.72	31.11±8.79	0.84
PO <sub>2</sub>	45.89±12.87	46.28±12.42	0.85
HCO <sub>3</sub>	15.14±4.44	15.03±4.43	0.86
BE	-10.49±4.27	-10.57±4.25	0.90

\* Independent 't' test, \*\* $\chi^2$  test

**Table II**

*Comparison of blood gas changes at different time point between both BCPAP and control groups (n=172)*

Blood Gas	Duration	BCPAP Group (N=85)(mean±sd)	Control Group (N=87)(mean±sd)	P* value
PH	Before intervention	7.20±0.05	7.18±0.07	0.08
	At 48 hrs of intervention	7.371±0.05	7.20±0.12	0.00
PCO <sub>2</sub>	Before intervention	31.38±8.72	31.11±8.79	0.84
	At 48 hrs of intervention	46.18±6.26	47.62±9.22	0.23
PO <sub>2</sub>	Before intervention	45.89±12.87	46.28±12.42	0.85
	At 48 hrs of intervention	86.06±38.24	46.86±7.47	0.00
HCO <sub>3</sub>	Before intervention	15.14±4.44	15.03±4.43	0.86
	At 48 hrs of intervention	18.64±3.75	15.50±3.39	0.00
BE	Before intervention	-10.49±4.27	-10.57±4.25	0.90
	At 48 hrs of intervention	-7.41±6.62	-12.12±6.82	0.00

\* Independent 't'-test

**Table III***Comparison of clinical changes at different time point between both BCPAP and control groups (n=172)*

Respiratory Charectaristics	Duration	BCPAP Group (N=85) (mean±sd)	ControlGroup (N=87) (mean±sd)	P* value
Respiratory rate(mean ± sd)	Before intervention	70.87±6.05	70.10±2.76	0.28
	At 48 hrs of intervention	47.96±5.33	69.09±1.80	0.00
Grunting[N (%)]	Before intervention	52 (61%)	51 (59%)	0.42**
	At 48 hrs of intervention	08(9%)	43 (49%)	0.00**
Chest indrawing[N (%)]	Before intervention	50 (58%)	49 (56%)	0.76**
	At 48 hrs of intervention	18(21%)	47 (54%)	0.00**
O2 saturation(mean ± sd)	Before intervention	83.09±4.84	82.37±6.62	0.41
	At 48 hrs of intervention	94.24±4.97	83.37±6.77	0.00

\* Independent 't' test, \*\* $\chi^2$  test**Table IV***Comparison of outcome in both BCPAP and control groups (n=172)*

Outcome	BCPAPGroup (N=85) (mean±sd)	Control Group (N=87) (mean±sd)	P* value
Duration of O <sub>2</sub> need in days	5.61±1.61	8.49±3.22	0.00
Duration to improve respiratory distress in Days	3.69±1.55	7.67±2.76	0.00
Hospital stay in days	8.74±3.72	12.67±11.75	0.00
Need for Mechanical ventilation [N (%)]	23 (27.06%)	45 (51.72%)	0.00**
Survived and discharged [N (%)]	65 (76.47%)	42 (48.28%)	0.00**

\* Independent 't'-test, \*\*  $\chi^2$  test**Discussion**

Role of CPAP in preterm and low birth weight infants is well documented.<sup>15, 16</sup> CPAP has been used primarily to treat surfactant deficiency in preterm infants for many years.<sup>17</sup> Particular interests in CPAP focuses on its potential role to reduce ventilator induced lung injury. One postulated mechanism is the avoidance of aggressive initiation of intermittent positive pressure ventilation with high tidal volumes and inadvertent hyperventilation or under ventilation that occurs in ventilated infants.<sup>18</sup> CPAP also protects the airway from mechanical injury and bacterial colonization related to the endotracheal tube. CPAP putatively increases both functional residual capacity and endogenous respiratory drive leading to decreased delivery room

intubations, reintubations and days on mechanical ventilation.<sup>18,19</sup> In addition; CPAP reduces the expression of superoxide and inflammatory mediators in tracheal aspirates which subsequently conserves surfactant.<sup>14</sup> More importantly the sustained CPAP pressure has been shown to stimulate the growth of the lung.<sup>20</sup>

This is one of the few prospective studies on the role of Bubble CPAP for preterm neonates with respiratory distress (gestation 28 to 34 weeks). In this study the mean age of starting CPAP was 28±13 hour as all the patients were out born, but study population was taken those who admitted within 72 hours of life. Mean gestational age and birth weight of the enrolled neonates were 31±1.61 weeks and 1454±284 gm in premature neonates who received

BCPAP. In a prospective study by Koti et al.<sup>21</sup> the median age of starting CPAP was 1.7 hours of life and all the patients were inborn. The mean gestation was 30.98±2 weeks and mean birth weight was 1387 ± 402 grams. The median duration of CPAP was 23.5 hours (range 2 -144 h). In a study by Courtney SE,<sup>22</sup> the birth weight was (mean ± sd) 1081± 316 g, gestational age 29 ± 2 weeks, age at study 13 ± 12 days and they did not shown the duration of CPAP.

In this study there was significant ( $p<0.05$ ) decrease in respiratory rate, increase in  $O_2$  saturation and also significant ( $p<0.05$ ) changes in arterial blood gas analysis after BCPAP intervention i.e, increased pH,  $PO_2$ ,  $HCO_3$  and BE but no changes observed in control group. There was significant ( $p<0.05$ ) change in duration of  $O_2$  need (5.61±1.61 vs. 8.49±3.22 days) and duration to improve respiratory distress (3.69±1.55 vs. 7.67±2.76 days). Mean duration of hospital stay was higher in neonates who were in control group (12.67±11.75 days) where as in BCPAP group it was 8.74±3.72 days. It also showed 65 (76.47%) of neonates who received bubble CPAP and 42 (48.28%) neonates in control group survived and the difference was found statistically significant ( $P<0.05$ ). In the study by Koti et al.<sup>21</sup> in infants surviving till discharge, the median duration of oxygen requirement was 102 (range 13-504 h) and median duration of hospital stay was 11 days (range 3-37 days). No baby had chronic lung disease. They did not show the blood gas analysis, respiratory rate and oxygen saturation changes in their study. In a study by De Klerk<sup>15</sup> et al, they had shown CPAP in preterm infants decrease the median duration of oxygen requirement (4 days) and median duration of hospital stay (6 days). They also found CPAP treatment decrease respiratory and some non-respiratory adverse outcomes.

In this study 27% of babies started on Bubble CPAP required ventilation whereas in control group it was 51%. No baby required oxygen for more than 28 days. Only one babies had pneumothorax but that baby was stabilized on Bubble CPAP and required neither ventilation nor chest tube drainage. In the study by Koti et al.<sup>21</sup> 25% of babies started on Bubble CPAP required ventilation. They found only two babies had pneumothorax but both these babies were stabilized on Bubble CPAP and required neither ventilation nor chest tube drainage. Lanieta et al,<sup>7</sup> had shown the introduction of BCPAP was associated with the

reduction in the need for mechanical ventilation and also decreased case fatality.

In a retrospective study by Ammari et al.<sup>23</sup>, the failure rate of Bubble CPAP was 24% in babies' d"1250g and 50% in babies d"750g. None of the babies with gestation >30 weeks failed CPAP. The main difference between our study and that by Ammari et al.<sup>23</sup> are, (a) ours is a unit which is doing Bubble CPAP for respiratory distress for 8 months before the onset of the study, (b) we used Fisher and Paykel nasal prongs<sup>23</sup> was used in this study while it was Hudson prongs in their study, (c) definition of CPAP failure included  $FiO_2 >70\%$  and PEEP >7cm for the first 7 days of life as against  $FiO_2 >60\%$  for the first 72 hours of life. No PEEP criteria were set in their study. These major differences might explain the differences in failure rates in the two studies. Since most events in the early neonatal period are reflections of the care and support in the first couple of days, we choose 7 days as the cut off for CPAP failures.

Establishing a NICU with mechanical ventilation would require high level of expertise and trained personnel, which is far from reality in many of the peripheral and district hospitals in developing countries. Lanieta et al.<sup>7</sup> have successfully demonstrated the usefulness of BCPAP in a developing country, and have also reported the cost effectiveness with use of Bubble CPAP.<sup>7</sup> Pieper et al.<sup>8</sup> have shown the importance of CPAP in the absence of neonatal intensive care and also the improved outcome in neonates treated with CPAP prior to transfer to a tertiary unit. Bubble CPAP, thus, may be considered as a primary mode of respiratory support in resource poor settings.

### Conclusion

It is concluded that Bubble CPAP is an effective way of improving oxygenation (e.g-  $O_2$  saturation and  $P^H$ ,  $PCO_2$ ,  $PO_2$  and BE) of preterm LBW babies with respiratory distress due to various causes. BCPAP use can reduce hospital stay and the need for mechanical ventilation.

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## ORIGINAL ARTICLE

# Single High Trans-Scrotal Incision Orchiopexy for Unilateral Palpable Undescended Testis: Experience in Dhaka Shishu Hospital

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## Abstract

**Background:** Palpable undescended testis (PUT) represents a common pediatric problem in many premature and some mature infants. There are several surgical techniques to correct PUT either through combined inguinal and scrotal incision or single transverse scrotal incision. This study assessed single high transverse scrotal incision for the management of PUT as regards to feasibility, postoperative success and final cosmetic result.

**Objective:** The aim of our study was to evaluate our modified extra dartos pouch technique in retaining testis in the scrotum.

**Methodology:** This prospective observational study was carried out in the division of paediatric surgery, Dhaka Shishu (Children) Hospital. A total of 90 male children with unilateral palpable undescended testes were included in the study from January 2014 to July 2016. They were randomly assigned for whom the extra dartos pouch technique was applied.

**Results:** The operation time was significantly shorter  $35.26 \pm 5.153$  minutes ( $p < 0.05$ ). Wound infection was found in 2 patients and inguinal congestion was found in 11 patients respectively. There was no statistical significant difference in developing wound infection and inguinal congestion. None of the patients developed scrotal hematoma, testicular atrophy and testicular re-ascent.

**Conclusion:** Single high trans-scrotal incision orchiopexy technique is a simple procedure. It is associated with one wound closure with less tissue dissection, good wound healing, cosmetic and a shorter operation time than the classical inguinal orchiopexy technique for unilateral palpable undescended testis.

**Key Words:** Undescended Testis, Trans-scrotal Incision, Orchiopexy.

## Introduction

Failure of testicular descent is one of the most common disorders in childhood.<sup>1</sup> Cryptorchidism word derived from Greek kryptos meaning "hidden" and orchis meaning "testis" refers to absence of a

testis from the scrotum.<sup>2</sup> A cryptorchid testis may halt along the normal pathway of descend (undescended testis), may deviate from the normal pathway of descend (ectopic testis), or may die or never develop (absent testis).<sup>3,4</sup> A testis that cannot

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be manipulated to the bottom of the scrotum without undue tension on the spermatic cord is called undescended testis.<sup>5</sup>

Undescended testis is a very common anomaly of the male infant<sup>4,6</sup>, affecting 1% boys at the age of 1 year old<sup>3,4,7</sup>, 3% of full-term male infants and 33% in premature babies at birth<sup>3,8</sup>. About 80% of undescended testes are palpable and 20% are non-palpable.<sup>3,4,8,9</sup> Unilateral undescended testis is four times more common than bilateral.<sup>10</sup> Patients with undescended testes should be treated because of increased risk of infertility, testicular cancer, torsion and/or accompanying inguinal hernia, as well as because of psychological stigmata.<sup>11</sup>

Treatment options of undescended testis are hormone therapy and surgery.<sup>9</sup> There are multiple surgical options. The basic principles in the surgical management were established in the last century, involving concurrent inguinal and scrotal incision. The classic approach in managing undescended testis involves both inguinal and scrotal incisions to relocate the testis in the scrotal position.<sup>5,8</sup> Though it has some alarming morbidity like wound infection, re-ascent and testicular atrophy, it provides adequate visualization and mobilization of the testis to be relocated in descended scrotal position. But now single high trans-scrotal incision for mobilization of palpable undescended testis is performed to decrease the potential morbidity of the traditional approach.<sup>8</sup> The present study prospectively reviewed the outcome of patients treated using a single high scrotal incision.

### Material and methods

The prospective observational study was carried out in Division of Pediatric Surgery of Dhaka Shishu (Children) Hospital, Dhaka from January 2014 to July 2016. The ethical permission was taken from Ethical Review Committee of Bangladesh Institute of Child Health (BICH). The protocol was commence the orchiopexy procedure with a high scrotal incision any patient from 6 months to 12 years unilateral palpable undescended testis admitted at Dhaka Shishu (Children) Hospital, regardless of the testicular anatomical position after taking the informed written consent from legal guardian (Fig 1). The possibility of conversion to conventional orchiopexy was explained to the parents. In all, 84 high scrotal orchiopexy (HSO) were carried out in 90 patients (Table I).

**Table I**

*The number and position of the undescended testes*

Number of patient	Position		
	SIP	IN	NS
90	36(40%)	30(33.3%)	24(26.7%)
Number (%)			

SIP, Superficial inguinal pouch. IN, Inguinal canal. NS, Neck of scrotum.

The first surgical step of the single scrotal incision orchiopexy was made a transverse skin incision that was commonly made along the high scrotal skin fold. On opening the deep fascia proximally, an avascular plane is entered. The testis or its gubernacular attachment was usually identified (Fig 2) The testis was carefully mobilized, delivered through the wound and the spermatic cord freed, using blunt and sharp dissection. The patent processus vaginalis was meticulously isolated, followed by its transfixation, ligation and division as high as possible. At this stage, most testes had a sufficiently long spermatic cord to reach a satisfactory scrotal position. Some testes may require further mobilization of the spermatic cord to reach the scrotum, achieved by blunt sharp retroperitoneal dissection through the opening of the external inguinal ring, after its retraction. The sub dartos pouch was made adequately. The testis was then relocated into the sub dartos pouch, and three fixing sutures were made between the testicular tunica albuginea and inner scrotal wall upper pole, lower pole and laterally to prevent ascent.

The scrotal skin was closed with interrupted absorbable sutures.<sup>8,12</sup> The operation time was recorded by wall clock.



**Fig 1** Right sided undescended testis before operation



**Fig 2** Right sided undescended testis during operation

All cases were followed upto 6 month after surgery(Fig 3). The outcomes of surgical approaches were operation time in minutes, post operative complication- wound infection, scrotal haematoma, inguinal/scrotal congestion, testicular re-ascent and testicular atrophy. The data collected from the respondents were analyzed. After completion of data collection, to maintain consistency, the data were checked and edited manually and verified before tabulation. Data were coded, entered and analyzed in a computer. The statistical analysis was conducted by using SPSS (statistical package for social science) version 20 statistical software. The findings of the study were presented by frequency, percentage. Means and standard deviations for continuous variables and frequency distributions for categorical variables were used to describe the characteristics of the total sample. Associations of continuous data were assessed using student t- test. Associations of categorical data were assessed using Chi-square test. For both test,  $p < 0.05$  was considered significant.

### Results

Eighty four testes (93.33%) were palaced into the scrotum satisfactorily using a single high scrotal incision. Six testes (6.7%) were required a second groin incision with extensive retroperitoneal dissection to achieve a good scrotal position. The mean age of children were  $45.54 \pm 32.57$  months . Mean operation time was  $35.26 \pm 5.153$  min where P value  $< 0.05$  (done by student t-test).



**Fig 3** Right sided undescended testis after operation

**Table I**

*Age of the participants*

Age(in months)	Trans-scrotal group (n=84)	Inguinal group (n=6)	P value
MeanSD	45.54±32.57	44.49±33.97	0.895
Range	11-132	6-120	
Median	38.00	36.00	

The p value is 0.895(obtained by t-test).

**Table II**

*Operation time of participants*

Operation time (In minutes)	Trans-scrotal group (n=84)	Inguinal group (n=6)	P value
MeanSD	35.26±5.153	52.46±3.860	0.0001

The p value is 0.0001(done by student t-test)

**Table III**

*Post operative complications.*

Complications	No.	(%)
Wound infection	2	(2.2%)
Inguinal/scrotal congestion	11	(12.2%)
Hematoma	0	(0%)
Testicular re-ascent	0	(0%)
Testicular atrophy	0	(0%)

### Discussion

In 1989, Bianchi and Squire reported their experience with scrotal orchiopexy claimed that their

technique was suitable for any palpable testes, and even recommended the approach for hernia repair.<sup>1</sup> However, the basic principles of the method used are similar and thus the present study is an independent review of a technique originated by other surgeons. Cryptorchidism is a very common anomaly of the male genitalia.<sup>3</sup> The undescended testis deserves treatment early in life to prevent potential adverse effect on fertility, as infertility was found with unilateral cryptorchid boy up to 10.5% of the patients<sup>13</sup> and to allow early detection of testicular malignancy, as 5 to 10 fold increase in malignancy in men with previous history of cryptorchidism compared to those descended testes.<sup>14,15</sup> Despite the recommendations for the treatment of the cryptorchidism before 1 year of age, many of the patients fail to do that, due to the socio-economic characteristics of the public health system, the lack of parental information and difficult access to tertiary health care.<sup>16</sup>

Ninety patients with palpable unilateral undescended testes were admitted in Dhaka Shishu (Children) Hospital, during the study period. There is an overall consensus that early orchiopexy is beneficial for future fertility and may be protective against the increased risk for testicular malignancy in cryptorchid patients.<sup>8</sup> In this study, the age of the participants was from 6 months to 132 months (11 years). Moreover as the study place was a hospital which allows patients up to the age of 12 years, participants up to age of 11 years were included so that follow up can be done for next 6 months. The John Radcliffe Hospital Cryptorchidism Study Group found that spontaneous descent occurred in the first three months and beyond that time it was rare. So the recommended age of orchiopexy is 6 months.<sup>17</sup>

The age was similar to a previous study, that showed the mean age were 40.54±10.3 months.<sup>12</sup> In another study, it was shown that the mean age were 3.44±2.2 years<sup>18</sup> which was similar to our study.

There was six conversion (6.7%), from single scrotal incision to inguinal incision orchiopexy in this study. In a study by Ramzan et al<sup>18</sup> 10 out of 134 (7.4%) patients, scrotal orchiopexy required conversion into two incision inguinal approach due to tearing of hernial sac and inadequate mobilization. Another study conducted by Eltayeb et al<sup>8</sup> showed that, in 3 out of 35 (8.5%) cases scrotal orchiopexy required conversion into two incision inguinal approach to gain more cord length.

The main advantage of single high trans-scrotal orchiopexy technique was less operation time. The mean difference of time was highly statistically significant ( $p < 0.001$ ). In another study, mean operation time in scrotal incision orchiopexy group was 40.5±25.9 minutes ( $p < 0.001$ )<sup>12</sup> which resembles the present study. Similarly, another study showed, mean operating time in scrotal incision orchiopexy was 28±10 minutes ( $p < 0.0001$ ).<sup>18</sup> The present study result also matches the previous study where the researcher also found significant statistical difference regarding operating time ( $p = 0.001$ ).<sup>8</sup>

The post operative wound infection is one of the common complications after orchiopexy.<sup>4</sup> In the present study, only 2 (2.2%) participants developed post operative wound infection. There were purulent discharge at the wound site in those patients. Though there was difference of post operative wound infection. This observation was similar to the study by Ramzan et al<sup>18</sup>, where, wound infection was found in 2 (1.4%).

While dealing with inguinal/scrotal congestion, it was found that there is no statistical significant regarding inguinal/scrotal congestion as the  $p > 0.05$  which was obtained from Pearson's Chi-square test. Patients, who developed inguinal / scrotal congestion, were treated by conservative care.

In this study, after operation, none of the patient developed scrotal haematoma, testicular reascent and testicular atrophy in any group. This resembles the result of the study conducted in Korea by Na et al<sup>12</sup>. In that study, none of the patient developed testicular reascent and testicular atrophy. Only one patient developed scrotal haematoma in scrotal incision orchiopexy.<sup>12</sup> Another study conducted by Eltayeb et al<sup>8</sup> showed 2 cases out of 35 (5.7%) developed hematoma which resolved spontaneously. In the same study 1 case in had developed atrophy and 3 cases had re-ascent of the testicle which were scheduled for redo-surgery.

### Conclusion

Single high trans-scrotal incision orchiopexy technique is a simple procedure. It is associated with one wound closure with less tissue dissection, good wound healing, cosmetic and a shorter operation time than the classical inguinal orchiopexy technique for unilateral palpable undescended testis.

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## ORIGINAL ARTICLE

# A Comparative study between Swenson abdominoperineal pull through and Transanal, full thickness, Swenson like approach with or without laparotomy for Hirschsprung disease

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## Abstract

**Background:** Hirschsprung's disease (HD) present with functional intestinal obstruction at the level of aganglionosis due to developmental disturbances in the enteric nervous system. Different operative techniques with individual advantages and disadvantages are existing to treat HD.

**Objective:** The aim of the study was to evaluate the better outcome between Transanal full thickness swenson like pull through with or without laparotomy and Swenson Abdominoperineal pull through.

**Methods:** A prospective comparative study had been carried out in the division of pediatric surgery, Dhaka shishu (Children) Hospital during the period of January 2014 to January 2016. Total 56 patients with HD patients were included in the study irrespective of colostomy except major co-morbidities and total colonic aganglionosis. Patients were allocated in both groups by random sampling. Twenty eight patients were allocated in Group-A and operated by Swenson abdominoperineal pull through and 28 patients were allocated in group-B and operated by Transanal, full thickness, swenson like procedure with or without laparotomy.

**Results:** Among the study population 32(57.14%) were male and 24(42.86%) were female with age range was 1 day- 96 months. Four patients developed urinary incontinence, 3 patients developed cuff abscess, 1 patients developed pelvic abscess, 4 patients developed fecal incontinence, 3 patients developed postoperative intestinal obstruction and in group-B no patient developed urinary and fecal incontinence but 4 patients developed cuff and anastomotic abscess and 1 patient developed enterocolitis. The mean Operation time  $114.14 \pm 13.02$  in group-A and  $80.46 \pm 29.03$  minutes in group-B. Mean hospital stay in group-A was  $6.18 \pm 1.83$  days and  $5.05 \pm .43$  days in group-B.

**Conclusion:** The newer modification, Transanal, fullthickness Swenson like procedure can be better technique in the management of HD.

**Key words:** Hirschsprung, Transanal Full Thickness, Swenson

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## Introduction

Hirschsprung's disease is a developmental disorder of the intrinsic component of the enteric nervous system that is characterized by the absence of ganglion cells in the myenteric and submucosal plexuses of the distal intestine.<sup>1,2,3</sup> Because these cells are responsible for normal peristalsis, patients with Hirschsprung's disease present with functional intestinal obstruction at the level of aganglionosis.<sup>1</sup>

The absence of ganglion cells or aganglionosis starts at the distal bowel beginning at the internal anal sphincter and extending proximally for various length, in 5% to 10% of cases can involve the entire colon or even a significant amount of the small intestine.<sup>4</sup> The incidence of Hirschsprung's disease is approximately 1 in 5000 live-born infants.<sup>1</sup>

The initial attempt to cure the disease was a diverting colostomy by various authors. The procedure relieved the symptoms but recurred after closure of colostomy. This, now we can explain by persistence of disease in the distal colon. Swenson's goal was removal of the entire aganglionic colon, with an end-to-end anastomosis above the anal sphincter.<sup>1</sup> Swenson pull through was the historic breakthrough in the understanding of pathogenesis and treatment of HD. The procedure, soon became popular as Swenson's procedure, brought a realistic hope that children with HD can be cured.<sup>5</sup>

Swenson is the least often chosen technique as surgeons all over the world was complaining about fecal, urinary and sexual dysfunction in children under went Swenson pull through.<sup>6</sup> yet, it is hard to surpass or even match the results that had been obtained by Swenson over many dedicated years. He was certainly a blessing to patients with congenital aganglionosis coli.<sup>7</sup>

As there are some complications like fecal incontinence, urinary incontinence, calf abscess and sexual dysfunction with Swenson pull through; some other procedures were described by several authors.<sup>8-16</sup> So search for newer ideas never ends. In this continuity Levitt MA et al.<sup>5</sup> introduced a newer idea. They thought that if they carried out the pelvic dissection very close to the wall of the rectum as they do in PSARP then morbidity might be minimized. Upon their thought they started Swenson like transanal pull through for Hirschsprung's patients.<sup>4</sup> They carried out the operation through transanal route if the patient had

Short segment HD, and abdominal and perineal route for patients with long segment disease.

We also believe that if the pelvic dissection can be carried out through transanal route rather than abdominal route in case of classic Swenson pull through then morbidity can be minimized, provided that transanal pelvic dissection done very close to the rectal wall. On this background we wanted to compare the outcome of abdominoperineal Swenson pull through with the most recent procedure transanal full thickness Swenson like pull through.

## Methods

The randomised controlled study was conducted in the department of paediatric surgery, Dhaka Shishu(Children) Hospital during the period of January 2014 to July 2016. Patients with Hirschsprung's disease diagnosed by history, radiology and histopathology irrespective of colostomy and level of aganglionosis, attending surgery outpatients department were the study subjects. First 56 patients admitted through SOPD with Hirschsprung's disease were included in the study after fulfillment of inclusion criteria and allocated in group-A and Group-B by random sampling. 28 patients in group-A were operated by classic Swenson procedure (abdominoperineal) and Group – B patients were operated by transanal, fullthickness, Swenson like approach with or without aid of laparotomy. Necessary informations and written informed consents were taken from the guardians for this study. After getting written consent from parents in a preformed questionnaire, clinical examination, evaluating preoperative management, operative findings, post operative management and follow up at 2nd, 4th, 8th and 12th weeks following operations and collected data were arranged in systematic manner and data was analysed by SPSS 19.0 software system and significant tests were performed by chi-square for qualitative data and un-paired t test for quantitative data.

## Operative Technique

All the patients were operated under general anaesthesia. All 28 patients of Group A underwent classic Swenson or abdominoperineal pull through where as all 28 patients of Group B underwent transanal Swenson like pull through. Mechanical bowel preparations along with rectal irrigations are recommended. Prior to operation a total body preparation from the nipples to the toes and urinary catheterization is performed.

### Swenson procedure

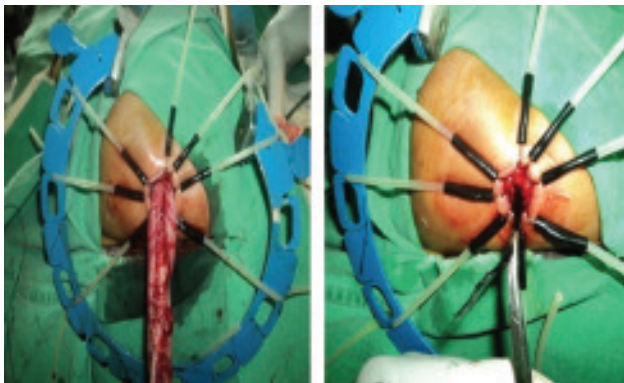
The operation was originally done through a laparotomy, with the anastomosis being performed from a perineal approach after eversion of the aganglionic rectum. coloanal anastomosis was done 1cm proximal to dentate line. Dissection continued up to the tip of the coccyx. Then the dissected aganglionic gut was pulled through the anus in everted fashion. Last of all the healthy ganglionic gut was pulled through the aganglionic gut keeping a seromuscular cuff measuring 1 cm anteriorly and 0.5 cm posteriorly (Fig 1).

### Transanal, fullthickness, Swensonlike procedure

The procedure was done through transanally. After preserving the dentate line within the hook of lone star retractor, full thickness dissection was carried out proximal to traction sutures placed 1 cm proximal to dentate line. Then dessection carried out on rectal wall to avoid injury to neuro vascular structures, finally aganglionic segment was removed and anastomosis was done by 2 layers 4-0 polygaktactin 910 suture material(Levitt MA et al.<sup>6</sup> (Fig 2).



**Fig 1** Steps of classic abdominoperineal Swenson Pull through. (A) Identification of normal colon with mobilization. (B) Coloanal anastomosis.

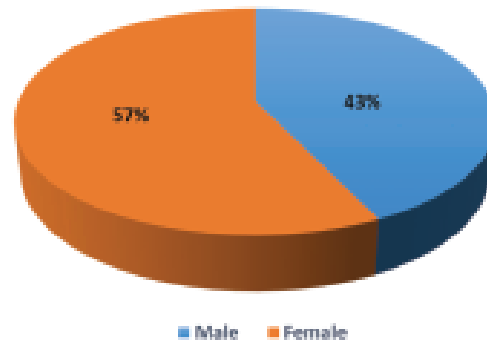


**Fig. 2** Steps of transanal full thickness Swenson like pull through. (A) Replace hook 1cm proximal to Dentate line. (B) Pull through of aganglionic narrow colon.

### Results

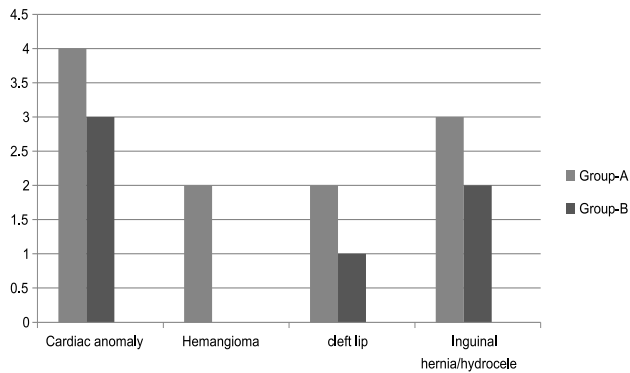
After employing exclusion criteria 56 patients were evaluated, with age at the time of operation ranging from 1 day to 96 months. Mean age of group-A were 21.16±11.57 months and in group-B mean age were 14.71± 19.68 months. Among all study population there were 32 male(57.14%) and 24 (42.86%) were female, a male:Female ratio of 1.3:1.(Fig I).

Features	Group-A (Classic Swenson)	Group-B (Transanal, Swenson like)
<b>Patients(n)</b>	28	28
Age(month)		
Mean ± SD	21.16±11.57	14.71±19.68
Range	6.5 – 49	1 - 96
Sex		
Male	20 (37.71%)	12 (21.43%)
Female	08 (14.29%)	16 (28.57%)



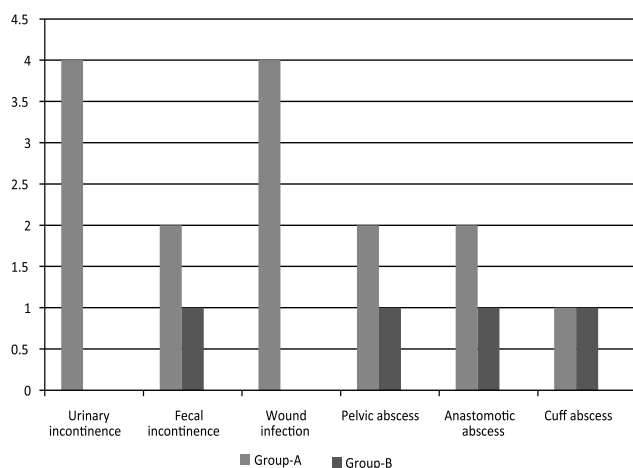
**Fig 3** Sex distribution among study groups

Associated anomalies in the series included 4 patients in Group-A & 3 patients in Group-B with minor cardiac anomaly, 2 patient in Group-A & 1 patient in Group-B with cleft lip. Pre operative evaluation with contrast enema revealed a transition zone between rectosigmoid to mid transverse colon. The average length of resected specimen was 22.7±11.9cm. in group-B and 3 patient in Group-A & 2 patients in Group-B had Inguinoscrotal swelling 23.17±17.67cm in group-A.



**Fig 4** Associated congenital anomalies among study populations

Postoperative complications were evaluated. No patients in group-B needed perioperative blood transfusion but 2 patients in Group-A needed post operative blood transfusion. Four Four patients in group-A and no patients in group-B developed urinary incontinence (p value is<0.05). Two patients in group-A and 1 patient in group-B developed fecal incontinence(p value>0.05). Four patients in group-A and no patient in Group-B developed wound infectio.(p <0.05). Five patients in group-A and 3 patients in group-B developed anastomotic/cuff/pelvic abscess(p>0.05). Five patients in group-A and 1 patient in group-B developed piost operative intestinal obstruction(p<0.05)



**Fig 4** Post operative complications

The mean operation time from incision to dressing was  $68.46 \pm 29.03$  minutes in group-B and  $114.14 \pm 13.02$  in group-A which was statistically significant(p value<0.05). Mean hospital stay from

operation to discharge was  $6.18 \pm 1.83$  days in group-A and in group-B it was  $5.01 \pm 0.43$  days(p value<0.05).

Variables	Group-A	Group-B	P Value
<b>Operation Time</b>			
Range	90-136	55-110	
Mean±SD	114.14±13.02	80.46±29.03	<0.05
<b>Hospital Stay</b>			
Range	5-9	4-6	
Mean±SD	6.18±1.86	5.05±0.43	<0.05

### Discussion:

This randomized control trial was conducted in the Department of Paediatric Surgery, Dhaka Shishu (Children) Hospital, Dhaka during the period of July 2014 to January 2016.

56 patients with Hirschsprung's disease having different length of colonic aganglionosis; with or without diverting colostomy were included by randomized sampling to form study sample volume. These 56 patients were grouped into: Group A , included 28 patients who were subjected to perform classic Swenson procedure and in Group B, included 28 patients who were operated by Trans anal full thickness Swenson like pull through with or without laparotomy.

In this study, surgical complications, outcome, operative cost and post operative hospital stay between the two groups were compared. In our study age range of the patients were 1 months to 96 months with the mean age 17.93 months. Though the maximum age limit in inclusion criteria was 18 years but we did not get any patient beyond 96 months or 8 years. The mean ages of Group - A and Group B were  $21.16 \pm 11.57$  months and  $14.71 \pm 19.68$  months. There was no significant difference (P value >.05) between the mean ages. In group A the mean age is higher than group B. It might be due to intentional delay by the surgeons who perform Swenson pull through waits for definitive pull through operation until the baby gains 10 Lb weight. In group B patients who need laparotomy the mean age was  $31.41 \pm 26.56$  months. Whereas patients in group B who underwent only transanal approach,

mean age  $8.025 \pm 11.23$  months. In the study of Levit MA et al.<sup>6</sup> mean age of the Patients were  $18.8 \pm 26$  months, which is very close to the mean age of our study.

Regarding sex distribution, in group A that is in classic Swenson group 20 (37.71%) patients were male and 08 (14.29%) patients were female. On the other hand in group B 12 (21.43%) patients were male and 16 (28.57%) patients were female. Among the whole study group 32 (57.14%) were male patients and 24 (42.86%) were female patients. Male: Female ratio was 1.33:1. Our result is very similar with other studies like Zhang SC et al.<sup>17</sup> and Levit MA et al.<sup>6</sup>

After confirmation of the diagnosis by means of rectal biopsy, contrast enema and preoperative evaluation, definitive resection pull through was performed in all 56 patients. 28 patients in Group A were operated by abdominoperineal classic Swenson pull through and in 28 patients of Group B Trans anal full thickness Swenson like pull through with or without laparotomy was applied. All the patients were followed up daily after pull through till discharge from the hospital and after discharge at 2nd, 4th, 8th and 12th weeks following operations.

In this study 4 (14.28%) patients in group A developed urinary incontinence where as no patients in group B developed urinary incontinence. Though incontinence can not be judged beyond 3 years of age, in our study patients having urinary dribbling even after last follow up after 3 months was noted as incontinence.<sup>18</sup> In this context there is significant difference between 2 groups. Probably the massive pelvic dissection in Swenson abdominoperineal pull through leads to injury to the nerve plexus around the bladder and causes incontinence.<sup>19</sup> In fact 9 patients in group A developed urinary dribbling post operatively, but in subsequent follow up on 8<sup>th</sup> and 12<sup>th</sup> week, we found that incontinence resolved in 5 patients and persisted in 4 patients. This 5 patients probably had neurogenic component which lead to overflow incontinence. Regarding group B, we noticed 4 patients had urinary dribbling after catheter removal. But in subsequent follow up on 8<sup>th</sup> week urinary dribbling of 3 patients resolved and 1 resolved on 12<sup>th</sup> week. In the study of Zhang SC et al.<sup>17</sup> showed 3.1% patients developed urinary incontinence, but he judged it after a long follow up more than 3 years. So we have to follow up more to conclude finally about urinary incontinence.

Regarding Pelvic or anastomotic or cuff abscess, 5 patients in group A and 3 patients in group-B developed pelvic/anastomotic/cuff abscess. We performed 2 stage operation by exteriorising the labelling colostomy for that 1 patients of group A who developed calf abscess. On the other hand 1 patients of group B who developed pelvic abscess also underwent 2 stage operation.

Like urinary incontinence fecal incontinence should be judge after 3 years of age.<sup>20</sup> moreover patients with stoma was ensured to have stoma closure 8 to 12 weeks of pullthrough, and patients having perianal fecal soiling during 12<sup>th</sup> weeks followup was labelled as fecal incontinence. Regarding this point of view 2 patients in group A developed fecal incontinence where as 1 patients in group B developed fecal incontinence. Zhang SC et al.<sup>17</sup> showed 9.1% patients in their study developed fecal incontinence. In our study it is 7.14%. Both the result is very much similar.

Postoperative wound infection denotes infection of abdominal wound in case of abdominoperineal Swenson pull through and patients of Swenson like transanal pull through where laparotomy was done. Upon this condition 4 patients that is 14.14% patients in group A developed post operative wound infection. On the other hand no patients in group B developed wound infection among 8 patients who needed laparotomy. Statistically there is difference between 2 groups. Wound infection was treated with wound swab C/S, regular dressing and culture sensitive antibiotics. Probably the higher operation time and fecal spillage during operation of group A patients influence more infection rate.

Obstructive symptoms occur in approximately 10% of children after surgical correction.<sup>21</sup> In the study there are more post operative obstructive symptoms in Classic swenson procedure because of more intraperitoneal handling or persistent symptom due to allied disorder like IND, hypoperistalsis, desmosis coli or Internal sphincteric achalasia.<sup>22-24</sup> In the study of TariqGM, Brereton RJ, Wright VM et al<sup>21</sup> the obstructive symptoms were 10% which is more than our study.

Regarding operation time, the mean operation time in group A was  $114.14 \pm 13.02$  minutes and in group B it was  $80.46 \pm 29.03$  minutes. Statistically there is significant gap between 2 groups. In group B patients who need laparotomy, mean operation time was

110.75± 8.10 minutes which is very near to abdominoperineal classic swenson operation. But if we concentrate with patients who only underwent transanal swenson like approach; the mean operation time was 68.35±25.60. Prolonged operation time of group A might lead to high wound infection rate among group A patients. In the study of Leily M et al.<sup>25</sup> the mean operation time was 134.3±51.4 minutes. Our mean operation time of abdominoperineal Swenson pull through was lower than the operation time taken by Leily et al<sup>25</sup>. Both the results are not very much dissimilar.

Unlike developed country, operation cost is important for a developing country like us. In our study operation cost includes cost of suture materials, all silicon folleys catheter, Urobag, Diclofenac sodium suppositories, Inj.Pathedine and surgica gloves. For some poor patients in our study place there was free bed and medicine supply from hospital. But the above mentioned things were mandatory for every patient to purchase. For that reason we included the above mentioned things for operative cost. In group A the mean operation cost was BD tk 3515.18±133.39 and in group B, it was BD tk 2564.46 ± 79.78. As in group A more suture materials needed to close the abdominal wound which leads to high operation cost.

Post operative hospital stay was ranges from 5 days to 7 days with the mean±SD was 6.18±1.82 days where as in group B range of post operative hospital stay was 4 to 6 days and mean was 5.04±.43 days. In group B 1 patient was discharged on request on 4<sup>th</sup> POD, that makes the range 4 to 6 days. After removal of the catheter on 5<sup>th</sup> POD, patients who developed retention, burning micturation or fever was kept admitted for more days. Moreover more wound infection among group A patients prolonged mean post operative hospital stay in group A patients.

### Conclusion

Transanal, Full thickness, Swenson like pullthrough with or without laparotomy can be a better approach to treat Hirschsprung's disease as this technique takes less operation time, less hospital stay and have less post operative complications.

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## REVIEW ARTICLE

# A Review of Complementary Feeding Practices in 6-23 Months' Children: A Worldwide Challenge

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### Background and objective

The first five years of life demands adequate nourishment to support a child's growth and development and is a pillar to the child's future. In Bangladesh, the condition of adherence to the Infant and Young Child Feeding (IYCF)\* guidelines has remained much below optimal, as evidenced from multiple national level reports. According to the Bangladesh Demographic Health Survey (BDHS)-2014, average 23% of the young children are appropriately fed during the complementary feeding period.

In a developing country such as Bangladesh, multiple micronutrient deficiencies have a widespread prevalence and often co-exist in the same individual. These include vital nutrients such as iron, iodine, folic acid, calcium, vitamin D, vitamin A and zinc. Chronic deficiencies of vital micronutrients may result in irreversible damage to child's growth and

development. Iron deficiency remains the most prevalent micronutrient deficiency with almost 50% of under-5 children reportedly suffering from anaemia.

Thus, it is necessary to translate knowledge into action through finding a structured solution towards effective implementation of nutritional interventions and evaluate the need for comprehensive approach to help bridging gaps of nutrition among infants and young children in Bangladesh.

Adherence to complementary feeding practices in Bangladesh

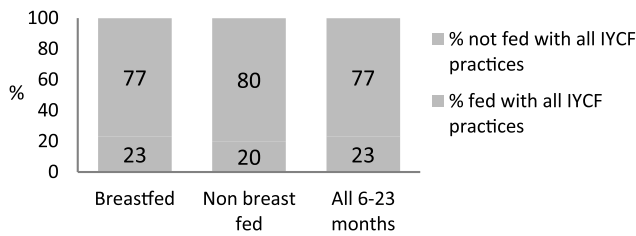
The Bangladesh Demographic and Health Survey (BDHS) 2014 reported that only 23% of all infants between 6-23 months of age are fed according to the IYCF guidelines (Figure 1). The four parameters of complementary food according to the IYCF guidelines include timely, adequate, properly fed and safe.

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\*IYCF Guidelines recommend exclusive breastfeeding for first 6 months of life; and complementary feeding with continued breastfeeding thereafter.

Contrary to the recommendations, the BDHS 2011 reported that complementary food is not initiated in almost one-third of the infants in Bangladesh.<sup>1</sup>






Source: BDHS 2014

**Fig 1** Non-adherence to feeding guidelines in Bangladesh<sup>12</sup>

**Gaps in the home-based meals in Bangladesh**

The IYCF guidelines include the ‘minimum acceptable diet’ as an important parameter to support optimal practices during the complementary feeding period. They include the minimum dietary diversity of at least 3 food groups and minimum meal frequency of at least twice a day for 6-8 month olds and at least thrice a day for 9-23 month old breastfed infants (Figure 2).

	Breastfed children	Non-breastfed children
<b>Milk</b> 	Given breast milk	Given milk, Infant formula or milk products such as chosse and yoghurt
<b>Frequency</b> 	Fed at least twice a day age 6-8 months and three times a day at age 9-23 months	Fed at least three times a day age 6-8 months and four times a day at age 9-23 months
<b>Diversity</b> 	Fed at least three different food groups	Fed at least four different food groups

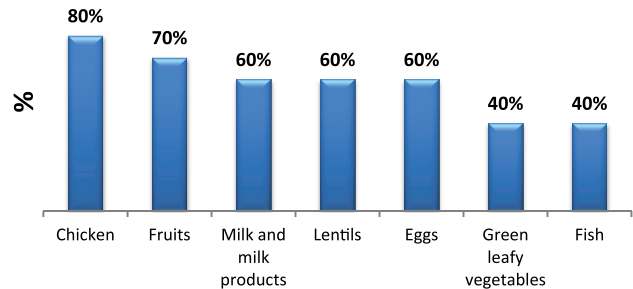
Source: A life free from hunger: Savethechildren, 2012

**Fig 2** Criteria for a minimum acceptable diet for a child aged 6–23 months<sup>2</sup>

In Bangladesh, the most common complementary foods include milk, rice gruel, suji, sugar water, sago and barley, which have a poor nutrient and calorie delivery. Statistics suggest that these common home based complementary foods are unable to fulfill the nutritional needs among infants in Bangladesh.<sup>3,4,5</sup>

According to the dietary diversity survey conducted by Rah et al. among 165,111 children, a vast number of young children aged 6 to 23 months consume less

than the recommended amounts of nutrient-rich foods as part of their complementary feeding (Figure 4). The study brought to surface that 1 in 5 children do not meet their daily dietary diversity score.<sup>5</sup>



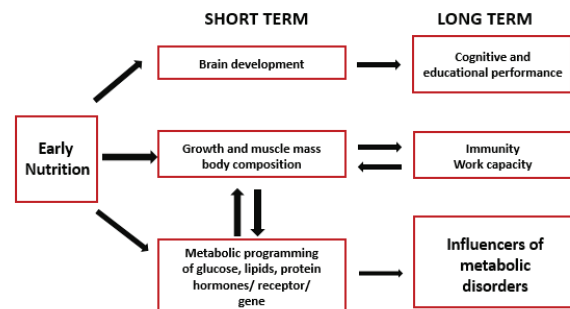
Source: Rah et al, 2010

**Fig 4** Percentage Pof infants who do not consume nutrient-rich foods (6- to 23-month-olds)<sup>5</sup>

**Burden of Hidden hunger among infants and young children in Bangladesh**

Hidden hunger is defined as a form of undernutrition that occurs when intake and absorption of vitamins and minerals are low to sustain good health and development.

With the limited quantity of food that an infant consumes, it becomes prudent to provide the right nutrition with every meal. Micronutrients are imperative to support key aspects of growth and development, beneficial for life (Figure 5).



Source: UNICEF

**Fig 5** The short term and long term effects of early nutrition<sup>6</sup>

**Prevalence of nutritional anaemia**

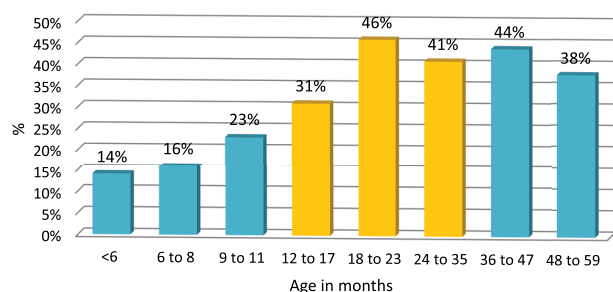
Inadequacy of multiple micronutrients directly contributes to the onset of anaemia; among which deficiency of iron plays the fundamental role.<sup>7</sup> According to BDHS2011,51% of infants (6-23 months)

in Bangladesh reportedly suffer from anemia. 21% were reported to be moderately anaemic and 1% severely anaemic. Among all age groups, anaemia is reported to be most prevalent among 9 to 23 month old infants during the complementary feeding period across socioeconomic status.<sup>1</sup>

Various factors may contribute to this dire problem of iron deficiency and anaemia in the country. Limited dietary diversity, low bioavailability, presence of phytates, limited caregiver knowledge, unhealthy environment, inadequate health care services and maternal anaemia are some of the key factors responsible for iron deficiency among infants in Bangladesh.<sup>8,9,10</sup>

Undernutrition during infancy in Bangladesh and its impact

Chronic insufficiency of key micronutrients may lead to faltered linear growth or stunting<sup>11</sup>. Globally, Bangladesh remains among the top 10 countries with a high stunting prevalence, amounting to 6 million children.<sup>11</sup> Statistics suggest the highest prevalence of stunting during the complementary feeding period, establishing its close relationship with compromised nutrition during this critical period of growth (Figure 6).<sup>11, 12</sup>



Source: BDHS 2014

**Fig 6** Prevalence of stunting in under-5 children in Bangladesh<sup>12</sup>

Stunting can also compromise cognitive development, IQ scores and a lifetime of learning.<sup>13,14</sup> Furthermore, it is associated with loss of nearly a year of school and 16% increased risk of failing at least 1 grade in school and is also known to diminish IQ by as much as fifteen points.<sup>15,16,17</sup> Stunting is also associated with increased risk of dropping out of schools by 3 folds.<sup>13</sup> Furthermore, a stunted child is more susceptible to repeated illness and in the long term has been associated with a 4-fold increased risk of mortality.<sup>11</sup>

Challenges with effective complementary feeding practices

### State of nutritional interventions

Various government and non-government organizations are actively working towards promotion of IYCF practices in Bangladesh. The Directorate General of Health Services (DGHS) and Directorate General of Family Planning (DGFP) implemented by The Ministry of Health and Family Welfare (MOHFW) are among the cardinal government organizations which promote IYCF through their national and regional arms. The other government organizations include Institute of Public Health Nutrition (IPHN), Bangladesh Breastfeeding Foundation (BBF), Bangladesh Pediatric Association (BPA), Community-based Kangaroo Mother Care (CKMC), etc.<sup>18,19</sup> In addition, a number of non-government organizations -such as Alive and Thrive, and GAIN – also promote optimal feeding practices.<sup>19</sup>

In order to eradicate malnutrition and promote optimal IYCF practices, these programs require 90% coverage. However, in Bangladesh, the coverage of essential nutritional interventions remains limited. Other than vitamin A supplementation, which has a comparatively higher coverage, other vital interventions such as age-appropriate complementary feeding and consumption of iron rich foods have reported a low coverage amounting to as low as 50%.<sup>20,21</sup>

Improving complementary feeding practices in Bangladesh: Recommendation from the Micronutrient Health Panel

The rampant prevalence of micronutrient deficiencies clearly suggests that home based diet is unable to provide optimal nutrition to infants. The panel unanimously resonated an ultimate need to bridge gaps in infant's nutrition to support overall growth and development, imperative for his/her healthy future.

Complementary food fortification as a sustainable solution to bridge gaps in nutrition Supplementation is a known solution to complete nutritional needs. However, the panel strongly agreed that supplementation is a problem-solution approach when clinically obvious/ diagnosed deficiency exists.

In addition, Bangladesh being among the countries with a considerably high carrier rate of thalassemia amounting to 7% of its population and 7000 babies born each year with thalassemia, a supplementation approach in these infants can lead to significant iron

overload resulting in tissue damage and serious complications.<sup>22,23</sup>

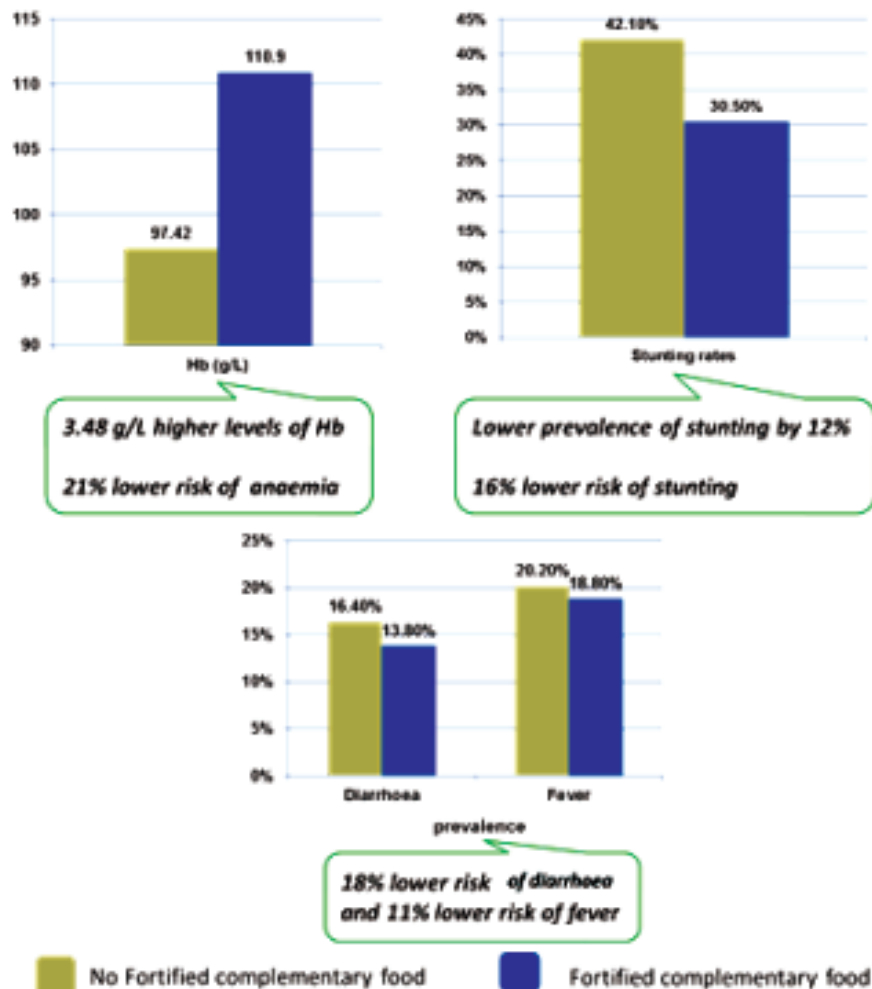
A study conducted by Sazawalet al.<sup>24</sup> compared the outcomes of fortified complementary feeding, home fortification with sprinkles—a micronutrient rich mix—and control in 292 young children aged 6 to 24 months. The adherence of sprinkles was approximately 63% and that of fortified complementary foods was around 81%. The study also showed significantly better outcomes with regard to growth markers such as weight-for-age and length-for-age compared with the control group. In addition, more infants displayed an acceptance towards ready-to-eat fortified foods compared to sprinkles. Therefore, ready to eat fortified complementary foods were concluded as a superior preference in infant nutrition.

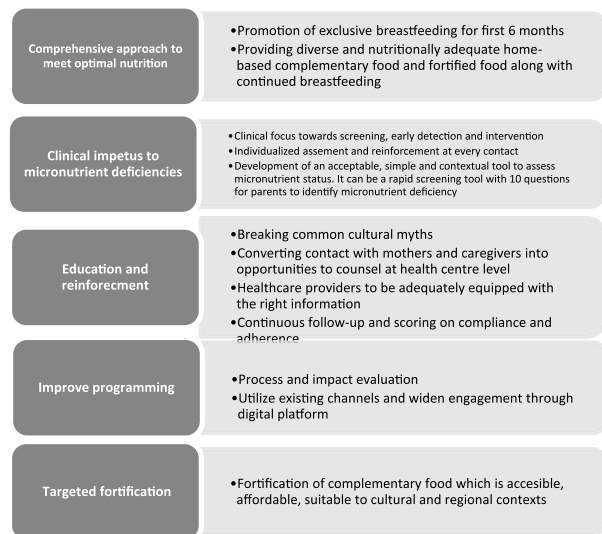
### Clinical evidence

A study conducted in India compared the intake of fortified complementary foods (n=10,338) versus non

fortified complementary food (n=12,812) among 6 to 23 months old children. The study evaluated 8 parameters namely, haemoglobin levels, anaemia, moderate anaemia, stunting, underweight, diarrhoea, fever and cough. 6 out of 8 parameters showed positive outcomes with fortified complementary food. The findings included: 3.48 g/L higher levels of Hb, 21% lower risk of anaemia, 12% lower prevalence of stunting, 16% lower risk of stunting and 18% lower risk of diarrhoea and 11% lower risk of fever in the group on fortified complementary food (figure 7).<sup>25</sup>

Thus it may be concluded that fortified complementary food could be an effective alternative towards bridging gaps in infants' nutrition. Of the major micronutrients, iron deserves specialized attention when considering targeted fortification. Combining various micronutrients which aid iron absorption could be beneficial to help complete infant's every day nutritional needs.





**Figure 8: Summary to the panel consensus**

**Fig 7** Positive health outcomes with fortified complementary food<sup>25</sup>

### Summary and conclusion

Micronutrient deficiencies require a clinical focus towards screening and early intervention to support overall growth and development of infants. The panel concluded that various approaches to micronutrient deficiencies have their own applications and benefits. Among all the possible approaches, prevention and education is the first order need. Supplementation can be beneficial where there is a presence of clinical deficiency, as a problem solution approach. Customized fortification with key micronutrients can help infants to meet their RDA levels and also prevent micronutrient deficiencies in the long term. Of all the nutrients, iron deserves cardinal focus, since the widest gaps exist for this particular micronutrient. Customized fortification based on regional and cultural context for infants can be a vital arm in providing targeted nutrition to infants. Customized fortification with right mix of micronutrients in the desirable levels can be a part of everyday nutrition as it has no known adverse effects and not just a need based approach. Therefore, a cost-effective, accessible, geographically specific, targeted fortification can be a sustainable solution to help meet the nutritional needs of infants, essential to support their growth and development and thus their healthy future.

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  25. Data on file.

## CASE REPORT

# Congenital Adrenal Hyperplasia (Salt Wasting)

Fahmida Zabeen<sup>1</sup>, M. Quamrul Hassan<sup>2</sup>, Nurun Naher<sup>3</sup>

### Introduction

The term congenital adrenal hyperplasia (CAH) involves a group of autosomal recessive disorders, which involves a deficiency of an enzyme involved in the synthesis of cortisol, aldosterone, or both.<sup>1</sup> Clinical symptoms of the different forms of CAH result from deficiency or excess production of particular hormones.<sup>2</sup> More than 90% cases of Congenital Adrenal Hyperplasia (CAH) are caused by a defect in the enzyme 21-hydroxylase.<sup>3</sup> CAH in most contexts refers to 21-hydroxylase deficiency. Congenital adrenal hyperplasia occurs among people of all races with an overall incidence of 1:15,000 live births for the classic form of 21-OH deficiency.<sup>4</sup> The two most serious neonatal consequences of 21 OH deficiencies occur when there is minimal measurable hydroxylase activity from prenatal period: life-threatening salt-wasting crises in the first month of life for XX and XY infants alike and severe virilization of female infants.<sup>1</sup>

As ill as these infants can be, they respond rapidly to treatment with hydrocortisone and intravenous saline and dextrose quickly restores blood volume, blood pressure, and body sodium content, and reverses the hyperkalaemia. With appropriate treatment, most infants are out of danger within 24 hours. Management of infants and children with CAH is complex and warrants long term care in a pediatric endocrine clinic. Management of adrenal crisis is an endocrine emergency. Classic congenital adrenal hyperplasia (CAH) with 21-hydroxylase deficiency is a rare autosomal recessive hereditary disorder of adrenal steroid biosynthesis. Samples for newborn screening should be collected on the 3rd day of life. In extremely high 17-OHP levels, the newborns should immediately be presented to the pediatric endocrinologist since salt-losing crisis may occur early. The clinical course in patients with classic CAH and 21-hydroxylase defect depends on early diagnosis of the disease. The maintenance therapy of CAH with glucocorticoid and mineralocorticoid should be adequate to replace deficient steroid and minimize adrenal sex hormone thus preventing virilization. Patients with CAH should be regularly followed up from childhood to adulthood by multidisciplinary teams who have knowledge of CAH. It is appropriate to offer genetic

counseling (including discussion of potential risks to offspring and reproductive options) to young adults who are affected, are carriers, or are at risk of being carriers.

### Case Report

A 36 days old Bangladeshi male baby was born of a non-consanguineous parents presented at Apollo hospital emergency room with the complaints of seizures for 2 days, repeated vomiting for 1 week, reluctant to feed and hyperpigmentation over face and trunk since 2 weeks of his age. There was no H/O fever. He was born at term normally with normal birth weight. He was exclusively breastfed. His only elder sib (brother) was normal.



**Fig 1** At 40 days of age 1<sup>st</sup> admission (4<sup>th</sup> day of admission)

On examination, he was moderately dehydrated, lethargic and was in shock, having systolic blood pressure 60 mm of Hg. His anthropometry was at the 3<sup>rd</sup> percentile for weight for age (weight: 3.4 Kg) and grossly wasted having weight for length <3<sup>rd</sup> percentile. No dysmorphic features were noted. Genital examination revealed hyperpigmented male genitalia with both testes descended. Phallus was normal in length and caliber with the urethral meatus at the tip. Physical exam was remarkable for marked hyperpigmentation over trunk and face.

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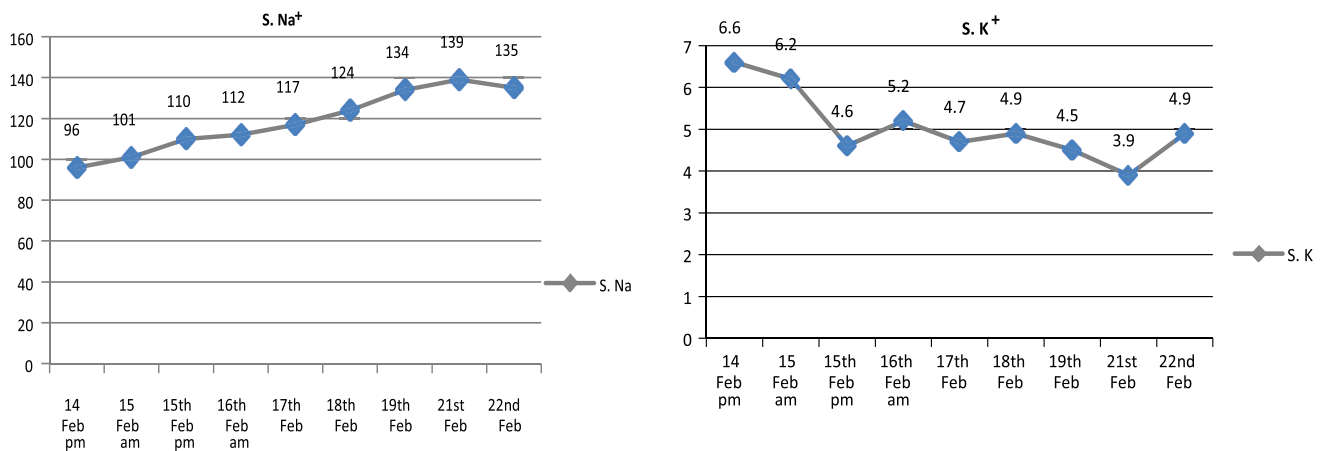
He was suspected as a case of CAH (salt wasting variety) in view of seizure, failure to thrive, dehydration, hypotension and hyperpigmentation.

His serum Electrolytes at emergency showed severe Hyponatraemia (Serum  $\text{Na}^+$  96 mmol/L), Hyperkalaemia (Serum  $\text{K}^+$  6.6 mmol/L) & Hypochloreaemia (Serum  $\text{Cl}^-$  62 mmol/L). He had natriuresis (Urinary spot  $\text{Na}^+$  52 mmol/L). His renal profile (Serum Creatinine 0.55 mg/dl), LFT and Blood sugar were normal. His septic screening was negative. There was no acidosis (Serum  $\text{HCO}_2^-$  21 mmol/L). USG of whole abdomen was normal. In view of suspicion of CAH of 21-hydroxylase deficiency, serum 17-OH-Progesterone level was done. It was elevated >38  $\mu\text{g}/\text{dl}$  (normal 5-25  $\mu\text{g}/\text{dl}$ ) and serum Cortisol was low at 4.6  $\mu\text{g}/\text{dl}$  (5-25  $\mu\text{g}/\text{dl}$ ).

The hypovolaemia with shock was managed with bolus normal saline infusion, and Hyponatraemia was treated with 3% Sodium Chloride, started I/V glucocorticoid (Hydrocortisone) and oral

Fludrocortisone. On 3<sup>rd</sup> day of admission Hydrocortisone was switched to oral Hydrocortisone 1 mg every 8 hourly. After 8 days of therapy, his Serum Electrolytes were normal and child remained seizure free all through and had no vomiting. He had started putting on weight chart on day 6 onwards.

After diagnosis, we followed up the child every monthly and closely monitored growth parameters, biochemical investigations and hormone assays. The baby got admitted twice before his 1<sup>st</sup> birthday, once at the end of December 2015 for Rota viral Diarrhoea and mild Bronchopneumonia and second at the end of January 2016 for bilateral Pneumonia. During each admission, he required stress dose Inj. Hydrocortisone and episode of hypokalaemia during second admission addressed carefully. For this hypokalaemia, recently increased dose of Fludrocortisone was modified and continued previous dose, reduced Hydrocortisone dose, continued oral supplemental Potassium and restarted oral 3% NaCl discharge. He was advised for follow up within 2 weeks of discharge with repeat investigations.



**Fig 2** Biochemical Profile (S. Electrolytes) during hospital stay



**Fig 3,4,5,6** F/U at 1 month 25 days, F/U at 4 months, 2<sup>nd</sup> admission at 11 months, 3<sup>rd</sup> admission at 1 year

**Table I***Anthropometry during subsequent follow-up during infancy*

F/up date	Age (days)At F/UP	Wt.(Kg)	Length (cm)	BP (mm of Hg)
1 <sup>st</sup> 26.02.15	1 m 18 d	3.65	55	93/72
2 <sup>nd</sup> 05.03.15	1 m 25 d	3.98	55.5	82/52
3 <sup>rd</sup> 25.03.15	2 m 15 d	5.22	58	85/54
4 <sup>th</sup> 13.04.15	3 m	6.52	60.5	80/55
5 <sup>th</sup> 12.05.15	4 m	7.5	65	85/56
6 <sup>th</sup> 07.06.15	5 m	8.1	65.5	88/54
7 <sup>th</sup> 08.07.15	6 m	8.35	66	87/58
8 <sup>th</sup> 15.07.15	7 m	8.53	67	85/55
9 <sup>th</sup> 20.08.15	8 m	8.66	68	88/60
10 <sup>th</sup> 17.09.15	9 m	9.1	70	85/55
11 <sup>th</sup> 21.10.15	10 m	9.48	73	85/56
12 <sup>th</sup> 23.01.16	1 yr 14 d	10.2	76	88/60

**Table II***Biochemical Profile and hormone assay during follow-up in Infancy*

DATE	S. Na	S. K	S. Cl	S. Cortisol, ACTH, Aldosterone	Treatment
1 <sup>st</sup> 26.02.15	139	5	102		Tab. Fludrocortisone 0.1 mg: Half tab. once daily Tab. Hydrocortisone 10 mg: 1/10 <sup>th</sup> Tab. 8 hrly 3% NaCl: 2.5 ml. with each feed,
2 <sup>nd</sup> 05.03.15 3 <sup>rd</sup> 25.03.15 4 <sup>th</sup> 13.04.15 5 <sup>th</sup> 12.05.15	133 to 139	5.2 to 5.6	99 to 102	S. Cortisol : normal 6.5 to 16.2	Tab. Fludrocortisone 0.1 mg: Half +o+1/4 <sup>th</sup> Tab. Hydrocortisone 10 mg: 1/10 <sup>th</sup> Tab. 6 hrly 3% NaCl: 3 ml. with each feed
6 <sup>th</sup> 07.06.15	141	4.4	103	ACTH-5pg/ml (8.3 -57.8) S. Cortisol 309 (101 -609)	Tab. Fludrocortisone 0.1 mg: Half +o+1/4 <sup>th</sup> Tab. Hydrocortisone 10 mg: 1/10 <sup>th</sup> Tab. 8 hrly 3% NaCl: 3 ml with each feed
7 <sup>th</sup> 08.07.15	141	2.5	102	ACTH- 10 pg/ml (8.3 -57.8) S. Cortisol 283.7 (101 -609)	Tab. Fludrocortisone 0.1 mg: Half tab once daily Tab. Hydrocortisone 10 mg: 1/10 <sup>th</sup> Tab. 8 hrly 3% NaCl: 3 ml with each feed
8 <sup>th</sup> 15.07.15 9 <sup>th</sup> 20.08.15	140 to 142	3 to 5	102 to 106	ACTH- 25.9 pg/ml (8.3 -57.8) S. Aldosterone low 12 (20-180), S. Cortisol basal 66.7 (101-690)	As above
10 <sup>th</sup> 17.09.15 11 <sup>th</sup> 21.10.15	134 to 138	4.7	99	ACTH- 56.4 pg/ml (8.3 -57.8) S. Aldosterone normal 31.22 (20-180), S. Cortisol basal 51.9 (101-690)	As above
2 <sup>nd</sup> Admission 24 <sup>th</sup> to 28 <sup>th</sup> Dec, 2015	148	5	115	Rota viral diarrhoea Mild Bronchopneumonia	Required stress dose Inj. Hydrocortisone CRP -Negative Sepsis screen - Negative
12 <sup>th</sup> 23.01.16	135	4.7	107	ACTH - 15.1 pg/ml (8.3 -57.8) S. Aldosterone 39 (20-180), S. Cortisol 8 am 236 (101-690)	Tab. Fludrocortisone 0.1 mg: 2/3 <sup>rd</sup> once daily Tab. Hydrocortisone 10 mg: 1/12 <sup>th</sup> Tab. 8 hrly Added Extra salt with food
3 <sup>rd</sup> Admission 27 <sup>th</sup> Jan to 2 <sup>nd</sup> Feb, 16	135 136	2.8 3.9	104 101	Bilateral Pneumonia	Required stress dose Inj. Hydrocortisone, Potassium supplements, reduced Tab. Fludrocortisone and restarted 3% NaCl orally on discharge CRP -Positive Sepsis screen - Negative

## Discussion

CAH has a wide spectrum of clinical severity depending upon the enzyme deficiency and the residual enzyme activity. The incidence of classical CAH is 1:10,000-1:20,000.<sup>5</sup> Affected males who are not detected in a newborn screening program are at high risk for a salt-wasting adrenal crisis because the excessive amounts of adrenal testosterone produce little effect on the genitalia of male infants; so their normal male genitalia do not alert medical professionals to their condition. They are often discharged from the hospital after birth without diagnosis and experience a salt-wasting crisis at home.<sup>6</sup> Our case was symptomatic since 2 weeks age, brought at emergency after 4 weeks age with features of hypotension, shock and generalized hyperpigmentation. His biochemistry showing severe hyponatraemia, hyperkalaemia and raised serum 17-OH-Projesterone level.

In classic 21-hydroxylase deficiency, laboratory studies will show very high concentrations of 17-hydroxyprogesterone at 3 days age in a full-term infant and hypoglycemia (due to hypocortisolism), hyponatremia (due to hypoaldosteronism) and hyperkalemia (due to hypoaldosteronism). Hyperpigmentation is due to co-secretion with melanocyte-stimulating hormone (MSH).

The lack of aldosterone results in a high rate of sodium loss (may exceed 50 mEq/L), which was also seen in our reported case being 52 mEq/L. With this rate of salt loss, the infant cannot maintain blood volume and hyponatraemic dehydration begins to develop by the end of the first week of life. Basic chemistries will reveal hyponatremia, with a serum Na<sup>+</sup> typically between 105 and 125 mEq/L. Hyperkalemia in these infants can be extreme-levels of K<sup>+</sup> above 10 mEq/L are not unusual as can the degree of metabolic acidosis. This reported case had S. Sodium 96 mEq/L and S. Potassium of 6.6 mEq/L. Ability to maintain circulation is further limited by the effect of cortisol deficiency. The early symptoms are spitting and poor weight gain, but most infants with severe CAH develop vomiting, severe dehydration, and circulatory collapse (shock) by the second or third week of life.

The lack of steroid product impairs the negative feedback control of adrenocorticotropin (ACTH) secretion from the pituitary, leading to chronic stimulation of the adrenal cortex by ACTH, resulting in adrenal hyperplasia.

Appropriate glucocorticoid and mineralocorticoid replacement during infancy in SW-21OHD children is critical for maintaining a positive sodium balance that enables adequate body growth and brain development. Studies evaluating the late effect of neonatal sodium deficiency in neurological performance, such as motor function, intelligence (IQ), memory, language and behavior, have shown poor neurodevelopment outcomes in the second decade of life.<sup>7</sup>

Therefore, the goal of therapy in SW-21OHD patients is to normalize the androgen levels and total-body sodium depletion to allow normal growth and development and avoid volume overload.<sup>[8]</sup> In our case study, the required glucocorticoid dose was stable during the first year of life. In contrast, the need for Fludrocortisone was higher during the first six months and continually decreased during later infancy follow-up; at one year of age, the mean dose of Fludrocortisone was 25 % lower than the initial dose, which suggested an improvement in the kidney regulation of salt and water balance.

The limited sodium content in breast milk most likely also corresponds with greater 9- $\alpha$ -fludrocortisone requirements in the first semester of life. The sodium composition of breast milk ranges from 3-18 mEq/L, and the milk volume production is 800 mL/day, which provides sufficient sodium to supply a healthy full-term newborn (i.e., 1-2 mEq/kg/day) but not enough to replace the sodium deficit that arises in salt wasting newborns (i.e. 4-5 mEq/kg/day).<sup>9-11</sup>

The range of mineralocorticoid doses was wider in the first semester of life, most likely because of the greater inter-individual variability of mineralocorticoid sensitivity and/ or greater difficulty in setting up an initial mineralocorticoid dose. Therefore, individual drug adjustments during this period should be assessed routinely.

The frequency of evaluation should vary depending on individual needs.<sup>12</sup> The goal of glucocorticoid replacement therapy is to replace deficient steroids, minimize adrenal sex hormone and glucocorticoid excess, prevent virilization, optimize growth, and promote fertility.<sup>13</sup> Hydrocortisone in tablet form is the treatment of choice in growing children. Treatment for CAH principally involves glucocorticoid replacement therapy, usually in the form of hydrocortisone (10-15 mg/m<sup>2</sup>/24 hours) given orally in two or three daily divided doses.<sup>14</sup>

Glucocorticoid therapy for children involves balancing suppression of adrenal androgen secretion against iatrogenic Cushing's syndrome in order to maintain a normal linear growth rate and normal bone maturation. Over treatment with glucocorticosteroids can result in Cushingoid features and should be avoided. It often occurs when serum concentration of 17-OHP is reduced to the physiologic range for age. An acceptable range for serum concentration of 17-OHP in the treated individual is higher (100-1,000 ng/dL) than normal, provided androgens are maintained in an appropriate range for gender and pubertal status. Efficacy of mineralocorticoid replacement therapy is monitored by measurement of blood pressure & early morning plasma renin activity or direct renin assay in a controlled position (usually upright).

We followed the child monthly and closely assessed growth and development and did regular check of Serum electrolytes, ACTH, Aldosterone and Cortisol but not reviewed serum 17-OHP and plasma renin assay. During periods of stress (e.g., surgery, febrile illness, shock, major trauma), all individuals with classic 21-OHD CAH require increased amounts of glucocorticoids. Typically, two to three times the normal dose is administered orally or by intramuscular injection when oral intake is not tolerated. Our baby received stress dose Hydrocortisone during the two admissions on later infancy and he responded well.

### Conclusion

Classic congenital adrenal hyperplasia (CAH) with 21-hydroxylase deficiency is a rare autosomal recessive hereditary disorder of adrenal steroid biosynthesis and management of adrenal crisis is an endocrine emergency. Early diagnosis and appropriate management and timely follow up from childhood to adulthood by multidisciplinary teams who have knowledge of CAH is essential for maintaining normal life.

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## CASE REPORT

# Colpocephaly: A Rare Brain Malformation

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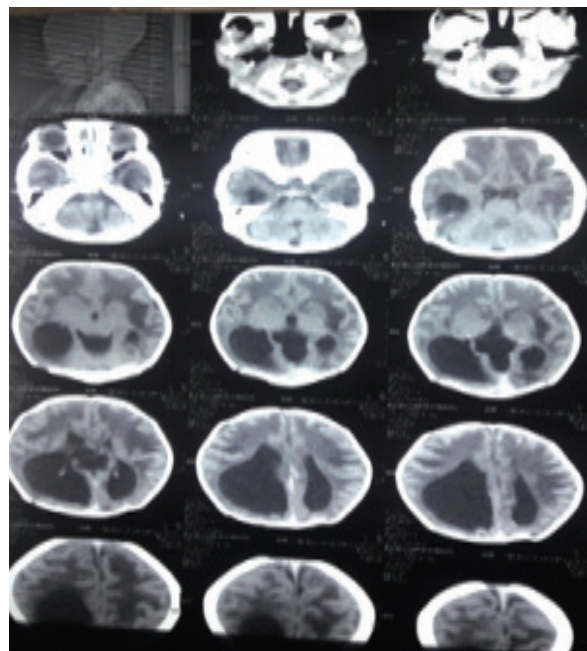
### Introduction

Colpocephaly is a very rare congenital anomalies of the brain<sup>1,2</sup> It is usually diagnosed in prenatal period, or early childhood as the patient usually suffer from mental retardations, seizures, and delayed motor milestone with presence of other congenital anomalies like meningocele, corpus callosal agnesis.<sup>1,2,3</sup> Colpocephaly is a term used to describe a congenital abnormal enlargement of the occipital horns of the lateral ventricles associated with normal frontal horns. It was first described these malformations as vesiculocephaly.<sup>4</sup> Later it was termed as colpocephaly, from the Greek kolpos, meaning hollow<sup>5</sup>. The aetiologies of colpocephaly were diverse. Some authors suggested that the term colpocephaly should be used as a descriptive term, not a specific congenital malformation of the central nervous system. Colpocephaly is a disorder of multiple and diverse aetiologies, including (1) chromosomal anomalies such as trisomy-8 mosaicism and trisomy-9 mosaicism; (2) intra-uterine infection such as toxoplasmosis; (3) perinatal anoxic-ischemic encephalopathy; and (4) maternal drug ingestion during early pregnancy, such as corticosteroids, salbutamol, and theophylline.<sup>4,5</sup> We present the following case in a patient who is the first issue of a consanguineous parents with colpocephaly, corpus callosum agnesis with seizures, vision, cognitive and motor impairment.

### Case Report

Tahmid a four months old boy 1<sup>st</sup> issue of a consanguineous parents was admitted in Dhaka Shishu(Children) Hospital with the complaints of generalized clonic seizures, which occurred several times in a day. He had a history of perinatal asphyxia

with Hypoxic Ischaemic Encephalopathy (HIE grade-II). His antenatal period was uneventful. He was conscious, mildly pale, having microcephaly(OFC 37.5cm >-3SD), he was severely stunted and moderately wasted with motor, vision, hearing and cognitive impairment. He was provisionally diagnosed as neurometabolic disorder but radio imaging (CT scan of brain) showed both lateral & 3<sup>rd</sup> ventricles are moderately dilated, colpocephaly with corpus callosal agnesis. TORCH screening was done as the child had growth retardation with developmental delay to exclude congenital infection which shows cytomegalovirus infection.



**Fig 1** CT scan of brain shows lateral & 3<sup>rd</sup> ventricles are moderately dilated, colpocephaly with corpus callosal agnesis

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**Fig 2** *Tahmid at the age of 4 months*

Tahmid was initially treated with phenobarbitone for control of seizures. Cytomegalovirus infection was treated with gancyclovir for 21 days. At a follow-up visit after 2 weeks his head circumference was 38 cm and his vision and hearing was improved. As there was no definite treatment for colpocephaly he was advised to receive monthly follow up for physical, visual stimulation, speech and cognitive therapies.

### Discussion

Colpocephaly is a rare congenital brain anomalies with familial occurrence has been noted in three reports. A genetic origin with an autosomal or X-linked recessive inheritance was suggested.<sup>6</sup>In our case it is genetic origin. Early reports suggested that the ventricular enlargement in colpocephaly was caused by a white matter development arrest, occurring between the middle of the second month to the fifth month of foetal life.<sup>6,7</sup> Later publications also favoured the idea that colpocephaly may indeed results from an anatomic mal-formation due to white matter developmental arrest. An embryological mechanism was proposed as follows: the lateral ventricles arise as large cavities of the telencephalic

vesicle. The normal developmental decrease in size of the ventricles occurs only after the formation of the foramen of Magendie, which decompresses the ventricular cavities. The occipital horns are further attenuated and shaped by the increase in size, volume, and myelinisation of the fibres in the ventricular walls as well as by association fibres of the corpus callosum and forceps, tapetum internal parieto-occipital fissure, and the calcarine fissure. It was suggested that yet-unknown causes may interfere with this delicate developmental process and the end result of such developmental arrest may be expressed as disproportionately enlarge occipital horns, which render the radiological appearance of colpocephaly.<sup>6</sup> Colpocephaly has been found in association with several other central nervous system malformations: agenesis of corpus callosum, neuronal migration disorders (lissencephaly, pachygyria), schizencephaly, macrogyria, and enlargement of cisterna magna, cerebella atrophy, optic nerve hypoplasia, chorioretinal coloboma, microcephaly, meningomyelocele, and hydro-cephalus.<sup>2,7</sup> Agenesis of corpus callosum is the most frequently associated malformation; our patient supports this finding. The types of brain malformations associated with colpocephaly can be related to insults occurring any time between 1 and 4 months of gestation. Other associated anomalies included micro-gnathia, hypoplastic nails, simian creases, Pierre-Robin syndrome, and neurofibromatosis.<sup>7</sup> Another possible explanation for the development of disproportionately enlarged occipital horns of the lateral ventricles is per ventricular leukomalacia due to destruction of the white matter of the occipital lobe.<sup>7</sup> Colpocephaly typically has been reported in association with various degrees of mental retardation, seizures, and motor and visual abnormality.<sup>8</sup> In this case the child also has the similar presentation. Colpocephaly is usually non-fatal. There has been relatively little research conducted to improve treatments for colpocephaly, and there is no known definitive treatment of colpocephaly yet. Specific treatment depends on associated symptoms and the degree of dysfunction. Anticonvulsant medications can be given to prevent seizure complications, and physical therapy is used to prevent contractures (shrinkage or shortening of muscles) in patients that have limited mobility. Patients can also undergo surgeries for stiff joints to improve motor function. The prognosis for individuals with colpocephaly depends

on the severity of the associated conditions and the degree of abnormal brain development.<sup>9</sup>

### Conclusion

Although colpocephaly is a rare congenital brain malformation of unknown aetiology, if any patient present with repeated seizures with global developmental delay, colpocephaly should be kept in mind as a differential diagnosis.

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## ABSTRACT FROM CURRENT LITERATURE

### Adolescent Growth in Overweight and Non-Overweight Children in Japan: a Multilevel Analysis

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*Paediatric and Perinatal Epidemiology*, 2014, **28**, 263–269

**Background:** A trend towards earlier pubertal growth has been identified along with an increase in childhood obesity rates. The study aimed to identify the differences in growth patterns during adolescence between overweight/obese and non-overweight children in Japan.

**Methods:** The participants were children from a prospective cohort study called Project Koshu, who were born between 1991 and 1998, in Japan. They were classified as overweight/obese or non-overweight according to their body mass index (BMI) in the first grade of elementary school (6<sup>th</sup> years of age) and were followed until graduation from junior high school (14<sup>th</sup> years of age). Anthropometric data were collected at an annual medical check-up in their school. Height gain trajectories were constructed by BMI categories using multilevel analyses. This analysis was stratified by gender.

**Results:** Overall, 111/850 (13.1%) girls and 109/911(12%) boys were defined as overweight/obese at baseline. Approximately 80% of the children were followed until the third grades of junior high school. Overweight/obese girls gained more height in the first half period, reached their peak height gain about a year earlier than non-overweight girls, and experienced an earlier decline in height gain. Similarly, overweight/obese boys gained more height than non-overweight boys initially. Additionally, non-overweight boys maintained a higher rate of height gain from the age at peak height gain, although the age at peak height gain did not differ between the two groups.

**Conclusions:** The overweight/obese children grew faster than the non-overweight children in the early pubertal stages, and the non-overweight children caught up and exceeded in height gain at a later stage.

### Anti-S. typhi Vi IgG levels in children with and without typhoid vaccinations

Sriandayani, Tonny H. Rampengan, Hesti Lestari, Novie Rampengan

*Paediatr Indones*, Vol. 54, No. 5, September 2014

**Background** Typhoid fever is endemic to Indonesia, with an annual incidence of 13/10,000 people. Vaccination has been shown to be an effective method to prevent typhoid fever. Of several vaccine types, the polysaccharide Vi vaccine is the most commonly used typhoid vaccine in developing countries. Results of previous studies remain inconclusive on the necessity of revaccination every 3 years.

**Objective** To compare the mean serum antibody titers of anti-S, typhi Vi IgG and the proportion of children with protective antibody levels between children with and without typhoid Vi vaccination.

**Methods** We conducted a cross-sectional study at Tuminting District, Manado from June to September 2012. Data was analyzed using independent T-test and Fisher's test. Serum anti-S. typhi Vi IgG levels were measured by enzyme-linked immunosorbent assay (ELISA) method.

**Results** Seventy-six subjects were divided into two groups: 38 children who had received the typhoid Vi vaccination more than 3 years prior to this study and 38 children who never had typhoid vaccinations as a control group. No statistically significant difference in age and gender was found between the two groups. The mean serum anti-Vi IgG level was 0.55 ug/mL (SD 0.58; 95%CI 0.36 to 0.74) in the vaccinated group, significantly higher than that of the control group [0.31 ug/mL (SD 0.42); 95%CI 0.17 to 0.44; P=0.038]. The proportion of children with protective anti-Vi antibody level was higher in the vaccinated group (23.7%) than in the control group (10.5%), however, this difference was not statistically significant (P=0.128).

**Conclusion** The mean serum anti-S, typhi Vi IgG antibody level in children who had been vaccinated more than 3 years prior to the study is higher than in children who had never received typhoid vaccinations. Nevertheless, the mean antibody titers are generally non-protective in both groups. Also, the proportion of children with protective antibody levels is not significantly different between the two groups.

### Probiotic *Weissella paramesenteroides* on enteropathogenic *E. coli*-induced diarrhea

Aslinar, Yusri Dianne Jurnal, Endang Purwati RN, Yorva Sayoeti

*Paediatr Indones.* 2014;54: 1-8.

**Background** Enteropathogenic *Escherichia coli* (EPEC) is a causative agent of intestinal inflammation and microfloral imbalance, leading to diarrhea. The presence of tumor necrosis factor- $\alpha$  (TNF- $\alpha$ ) in the feces is an indicator of inflammation in the intestinal mucosa. *Dadih*, (local made of fermented buffalo milk), contains probiotics and is widely consumed by the people in West Sumatera, Indonesia. *Weissella paramesenteroides*, a probiotic lactic acid bacteria (LAB), has been isolated from *dadih* and is believed to be useful for improving intestinal microflora balance and inhibiting the activity of harmful microbes.

**Objective** To determine the efficacy of *W paramesenteroides* administration in various doses and durations on bowel frequency, stool's TNF- $\alpha$  levels, and intestinal microflora balance on mice with EPEC-induced diarrhea.

**Method** This randomized experimental animal study examined two factors relating to the effects of *W paramesenteroides* on EPEC-induced diarrhea, namely doses of probiotics (factor A), and durations of observation (factor B). The subjects consisted of 100 male white mice (*Mus musculus*) aged 8 weeks, with weights of 25-30 grams. The outcomes measured were bowel frequency, stool's TNF- $\alpha$  levels, and the balance of intestinal microflora on mice with EPEC-induced diarrhea. Subjects were divided into 5 groups: the negative control group (received neither EPEC nor probiotic), positive control group (received only EPEC), and three experimental groups (received EPEC and different doses of *W paramesenteroides*). Probiotics were given twice at the 12-hours and 24-hours for the experimental groups, while the durations of observation consisted of baseline, 12 hours, 24 hours, and 36 hours.

**Results** After 36 hours, subjects with EPEC-induced diarrhea who received *W paramesenteroides* administration in doses of  $2 \times 10^8$  (A3), were found to have the largest decline of mean defecation (a 4.4-fold decline) and the largest decline of stool's mean TNF- $\alpha$  levels (48.3 pg/mL), compared to the positive control group, and other experimental groups who received higher doses of probiotics.

The highest increase of mean LAB (up to  $57.50 \times 10^7$  cfu/g), the lowest mean of aerobic bacteria ( $2.5 \times 10^7$  cfu/g), and *E. coli* ( $1.5 \times 10^7$  cfu/g) were also found in A3 group.

**Conclusion** Administration of *W para-mesenteroides* at the dose of  $2 \times 10^8$  has beneficial effects on reducing bowel frequency, decreasing stool's TNF- $\alpha$  levels, and improving the balance of intestinal microflora in mice EPEC-induced diarrhea.

### Vitamin E effect on osmotic fragility in p thalassemiamaajor

Agus Fitrianto, Moedrik Tamam, Nyoman Sud Widyastiti

*Paediatr Indones.* 2014;54:280-3.

**Background** Blood transfusion remains the main therapy for anemia in P thalassemia major patients. However, frequent transfusions can cause oxidative stress in response to iron overload. Vitamin E is considered to be the best lipid-soluble exogenous antioxidant in humans. It can protect phospholipid membrane from peroxidation. Erythrocyte osmotic fragility is a useful test to assess for the improvement of red blood cells in thalassemia patients after vitamin E supplementation.

**Objective** To investigate the effect of vitamin E for improving erythrocyte osmotic fragility in P-thalassemia major and for decreasing the need for frequent transfusions.

**Methods** This was a double blind placebo controlled randomized clinical trial on children aged 2-14 years with thalassemia major who received frequent blood transfusions. Fifty subjects were divided into 2 groups: group I with vitamin E supplementation and group II with placebo, as a control group, for a period of 1 month. Pre- and post-treatment data on erythrocyte osmotic fragility and hemoglobin level were analyzed with non-paired T-test.

**Results** Improved erythrocyte osmotic fragility was found: in group I, pre-treatment 31.59 (SD 6.342)% to post-treatment 38.08 (SD 7.165)%, compared to the control group pre-treatment 34.40 (SD 6.985)% to post-treatment 29.26 (SD 9.011)% (P=0.0001). Comparison of the mean delta Hb level in group I was 0.94 (SD 0.605) gr% and that of group II was - 0.23 (SD 1.199) gr% (P= 0.0001).

**Conclusion** Vitamin E supplementation improves erythrocyte fragility and Hb level in P-thalassemia major pediatric patients.

## DSH NEWS



Inauguration of Cardiac ward of Dhaka Shishu Hospital by Honorable Health and family welfare minister Mohammad Nasim along with Honorable Chairman of the management board, Dhaka Shishu(Children) Hospital(DSH), National Professor Dr. Shahela Khatun, Honourable Director of DSH Professor Manzoor Hussain, Honourable Director General of Health Services, Professor Dr. Kazi Din Mohammad Nurul Hoque and Honourable Academic Director of BICH Professor MAK Azad Chowdhury, to improve the paediatric cardiac care in our country



Visitors from WHO, Visited Dhaka Shishu (children) Hospital

## BICH NEWS

BICH is the academic wing of Dhaka Shishu Hospital. It was established in 30<sup>th</sup> January, 1983. It is affiliated with Dhaka University, Bangabandhu Sheikh Mujib Medical University (BSMMU) and Bangladesh College of Physicians and Surgeons (BCPS). It has been conducting different courses e.g. DCH, FCPS, MD Paediatrics, MS Paediatric surgery & B.Sc in Health technology. It also conduct different sub-specialty courses e.g. FCPS Neonatology, FCPS Haemato-oncology, FCPS Nephrology, MD Neonatology, MD Haemato-oncology and MD Nephrology. It conducts 3 months certificate course in Paediatrics and 15 days Intensive course for MCPS. It organizes IMCI training and Palli Shishu Rural Health Training. Apart from this, the Institute also run its regular academic activities. It has established Basic Science Department since 2006.

Diploma course of paediatric nursing has started from 1st January 2012 and Diploma in paediatric physiotherapy under process.

### **Library facilities**

The library of BICH has a rich collection of updated medical texts and reference books and reputed Medical Journals of home and abroad. BICH has introduced Broad Band facilities which are open to all students, teachers/ consultants of hospital for 24 hours. Facilities of library are also improved by HINARI. Students can download 2230 Medical Journals & more than 50 Paediatric Journal.

### **Present News**

A newly formed classroom in BICH has been named as Prof. Sultan Ahmed Chowdhury as a tribute to First Honorary Director of Dhaka Shishu Hospital.

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## Postgraduate courses/training in paediatrics and child health

1. FCPS in paediatrics : Twice in a year, in the months of January and July.
2. Recognized center by BCPS for training in FCPS (Paeditric surgery) .
3. Recognized centre for course and training in different subspeciality as: Neonatology, pediatric Nephrology, paediatric haematology and Onchology, paediatric pulmonology and paediatric Neuroscience.
3. MD/MS in paediatrics : Part I: In the month of January every year; 2nd and 3rd parts twice every year.
4. DCH course : Once in a year in the month of July.
5. Three months certificate course : The institute every year runs 3 months certificate course on paediatrics for general practitioners & other post graduate candidates e.g. MCPS.  
(1st August – 31st October)
6. Training programme on IMCI (Integrated management of childhood illness), Essential Newborn Care for doctors and nurses. KMC (Kangaroo Mother Care) traing, ETAT (Emmergency Triage, Assessment and Treatment) training.

**Contact Person** : Prof MAK Azad Chowdhury  
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# Students Qualified from Bangladesh Institute of Child Health

## Undergoing Courses of BICH

Institution	Courses
Bangabandhu Sheikh Mujib Medical University	MD(Paediatric)
	MD (Neonatology)
	MD (Nephrology)
	DCH
	MS (Paediatric Surgery)
Bangladesh Collage of Physicians and Surgeons (BCPS)	FCPS (Paediatric)
	FCPS (Neonatology)
	FCPS (Haemato-Oncology)
Dhaka University	BSc (Health Technology)

## Students Qualified From BICH Till January 2015

Name of Courses	Number
DCH	327
MD (Paediatrics)	95
MS (Paediatrics)	86
FCPS (Paediatrics)	22
MD (Neonatology)	12
MD (Paediatric Nephrology)	4
<b>Total</b>	<b>546</b>

## Foreign Students Qualified From Bich Till January 2013

Country of Origin	Course	Number
Nepal	DCH	23
	MS(Paed. Surgery)	2
	MD(Paed)	1
India	MD(Paed)	1
Iran	DCH	1
Iraq	DCH	1
Somalia	DCH	1
Sudan	DCH	1
<b>Total</b>		<b>31</b>

## Present Students January 2015

Name of Courses	Number of Students	
DCH	14	
MD (Paediatrics)	Part-I	13
MD (Paediatrics)	Part-II	8
MD (Paediatrics)	Final Part	2
MS (Paediatrics)	Part-I	6
MS (Paediatrics)	Part-II	2
MS (Paediatrics)	Final Part	6
FCPS (Paediatrics)	Part-II	2
FCPS	(Paediatric Nephrology)	1
<b>Total</b>		<b>54</b>

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## **Seminar/Symposium & CME/CPD programs held at BICH (January to June, 2015)**

<b>Date</b>	<b>Topic</b>	<b>Presenter</b>
25 Jan 2015	Nocturnal Enuresis	Department of general paediatrics
22 Feb. 2015	An Update of Aplastic Anaemia:	Department of Haematology and Oncology
26 April, 2015	An Approach to Short Stature:	Department of Endocrinology and Metabolic disease
07 May 2015	World Thalassaemia Day “enhancing partnership towards patient-centred health systems: good health adds life to years”.	Department of Haematology and Oncology
14, June 2015	Portal Hypertension:	Department of Paediatric Gastroenterology, Hepatology and Nutrition