

# CONGENITAL TRACHEO-OESOPHAGEAL FISTULA\*

A REVIEW ILLUSTRATED BY 34 CASES FROM THE CHILDREN'S HOSPITAL, JOHANNESBURG

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Congenital tracheo-oesophageal fistula, which is not uncommon, is inevitably fatal without treatment and the diagnosis cannot be delayed for any length of time after birth if success is to crown any surgical endeavour. The results, however, considering the magnitude of the anatomical problem, are encouraging.

## PATHOLOGY

In this condition, the proximal cervical segment of the oesophagus ends blindly in a thick-walled hypertrophied pouch, which varies in length from 3 to 4 cm. The upper end of the lower oesophageal segment is usually attenuated and opens by a small fistula 2-6 mm. in diameter into the trachea, less commonly into the carina, and rarely into the right main bronchus. There is always a gap between the distal end of the upper pouch and the proximal end of the distal oesophageal segment at its fistulous opening. Occasionally the two portions of the oesophagus are in close proximity, in others there is a gap of 1 or 2 cm., occasionally up to an inch, and in rare instances, when the fistula opens into the bronchus, a greater gap. Rarely, there is no fistula and the lower end of the oesophagus is atretic and not developed.

## Pathological Classification

According to Vogt's classification of atresia (Fig. 1) of the oesophagus there are three groups:<sup>1</sup> viz. (1) Complete atresia,

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(2) atresia with blind upper and lower pouches, and (3) atresia of the oesophagus with tracheo-oesophageal fistula (*a*) between trachea and upper segment, (*b*) between trachea and lower segment, or (*c*) between trachea and both segments. A 4th group, viz. tracheo-oesophageal fistula without atresia, should be added. Attempted closure of this isolated anomaly was first described by Imperatori<sup>2</sup> in 1939.

Of the published cases 90% fall into Vogt's group 3a. Of our 34 cases all but one, which fell into group 2, belonged to group 3a, though the gap between the segments varied considerably.

## Embryology

Essentially, according to Saunders,<sup>3</sup> the subdivision of the foregut into its tracheal and oesophageal components is due to ingrowth of two laterally placed septa into the developing lung bud. The failure of complete fusion results in tracheo-oesophageal fistula, whereas undue obliquity of union of these septa results in atresia.

## Associated Congenital Anomalies

In 13 of our operated cases associated congenital anomalies were noted; in 3 of them multiple anomalies were present. In only 2 cases were these anomalies incompatible with life. One mongol weighing 7 lb was not operated upon. These anomalies were as follows:

(a) *Skeletal Anomalies.* Gross spina bifida 1 case, cervical ribs 4 cases, accessory ribs 3 cases, absent rib 1 case, bifid vertebrae 2 cases, hemivertebrae 1 case (with 3 such vertebrae),

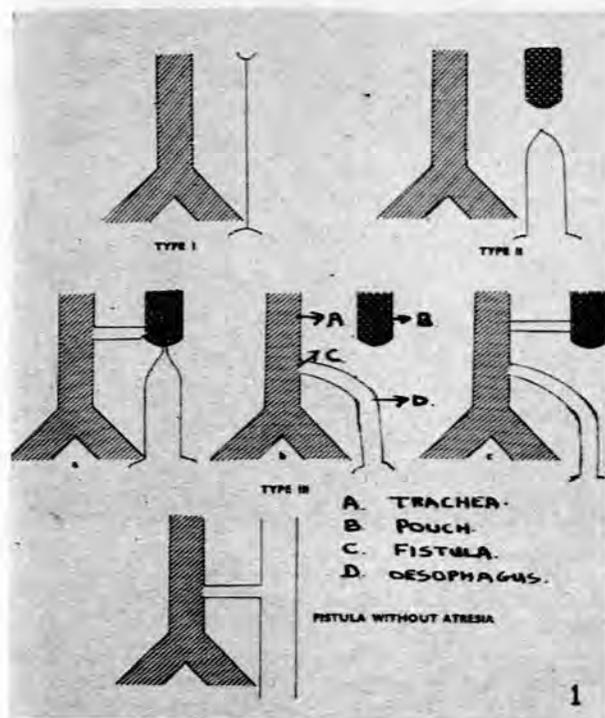


Fig. 1. Congenital tracheo-oesophageal fistula. Diagrams illustrating Vogt's classification.

fused vertebrae 1 case, and 1 case each of bifid right thumb, accessory digits, absent thumb metacarpal, and absent radius.

(b) *Cardio-vascular Anomalies.* One case each of ventricular septal defect, patent ductus, and right aortic arch. One case of tetralogy of Fallot successfully underwent surgery (Fig. 7).

(c) *Central-nervous-system Anomalies.* One case each of myelocoele, hydrocephalus, and mental retardation.

(d) *Gastro-intestinal Anomalies.* Two cases of imperforate anus and 1 case of perineal fistula.

(e) *Genito-Urinary Anomalies.* 1 case each of unilateral kidney, horse-shoe kidney, and hypospadias.

(f) *Cutaneous Anomalies.* One case with maldeveloped pinnae and imperforate external auditory meati.

#### HISTORY

Despite the fact that, according to Franklin,<sup>4</sup> the first case was observed by Durston as long ago as 1670 and that the condition was first described by Gibson<sup>5</sup> in 1697, the complete failure of attempts at surgery limited interest in this condition until 1939, when the first surgical success was reported.

Until 1936 all attempts to save these infants were limited to cervical oesophagostomy to allow the upper pouch to drain and gastrostomy to allow for feeding; and in the rare cases which survived an attempt was made to reconstitute an ante-thoracic oesophagus. No case survived these multiple procedures until 1939, when Ladd<sup>6</sup> reported his first survivor in an extensive experience. In Lanman's review<sup>7</sup> (1940) all the 30 cases terminated fatally. By 1940, however, Leven<sup>8</sup> had performed 28 of these multiple-stage operations and 9 patients had lived.

As long ago as 1913<sup>9</sup> Richter had suggested that the ideal

surgical treatment would be a direct operation aimed at ligating the fistula and anastomosing the oesophagus end-to-end by a transpleural approach. However, the first direct operation was performed extrapleurally in 1936 by Lanman.<sup>7</sup> Robert Shaw<sup>10</sup> reported the first case to survive the direct operation in 1939. This patient only lived for 12 days and autopsy demonstrated disruption of the anastomosis; Shaw stated that 'leakage from the anastomosis would inevitably occur'. In 1943 Haight and Townsley<sup>11</sup> reported the first survivor from this extrapleural operation, and ushered in a new era of hope in the treatment of these otherwise doomed infants. The first successful case outside the United States was operated upon by Franklin<sup>4</sup> of London in January 1947. In October 1947 Lee MacGregor<sup>12</sup> performed this extrapleural operation in South Africa under local analgesia but the child did not survive more than a few hours. Our first operation was in February 1949 and our first survivor was our third case, in May 1949. These cases were reported by F. W. Roberts.<sup>13</sup> Our first survivor is now 8 years old and has continued to thrive.

In October 1941 Franklin<sup>14</sup> had expressed the view that the extrapleural approach was too tedious, caused too much paradoxical post-operative chest movement, and was so frequently accompanied by minute tears of the pleura that he advised transpleural thoracotomy. He attempted this in 6 cases without success, and he employed the extrapleural approach successfully. Singleton and Knight<sup>15</sup> reported a 7-months survivor after a transpleural repair. Belsey,<sup>16</sup> of Bristol, employed this approach regularly; his first survivor was operated upon in December 1948. Since 1953 we have always used this approach.

#### INCIDENCE

Before the advent of surgery most infants died of this condition, under the diagnosis of neonatal atelectasis or bronchopneumonia. Haight<sup>17</sup> states that the anomaly occurs once in 2,196 births. Franklin,<sup>18</sup> from the Postgraduate School in London, gave the incidence as 1 in 2,635 deliveries, but Belsey<sup>19</sup> states that the condition is much commoner, occurring once in 800 births. As awareness of the condition amongst doctors spreads so the incidence rises. One of our cases was diagnosed by a midwife 250 miles from Johannesburg. She had attended a lecture at the Queen Victoria Hospital by Dr. Seymour Heymann, who had stressed the presenting symptoms to the class.

#### Sex Incidence

This anomaly is commoner in males. Of the 31 cases in which the sex was recorded, 21 were males, i.e. 68% (the sex was not recorded of 3 patients who died shortly after operation).

#### DIAGNOSIS

All neonates have an excess of oral mucus, but these infants with this condition characteristically froth excessively at the mouth; this recurs after aspiration. They have repeated attacks of cyanosis due to overflow and inhalation of saliva and mucus from the proximal pouch. They are avid feeders but immediately return everything unchanged, and the symptoms are aggravated when fluids are given. The abdomen rapidly distends as air passes through the fistula into the stomach *via* the lower oesophageal segment. After this initial phase secondary changes due to inhalation pneumonitis occur. The child is now more constantly cyanosed, with

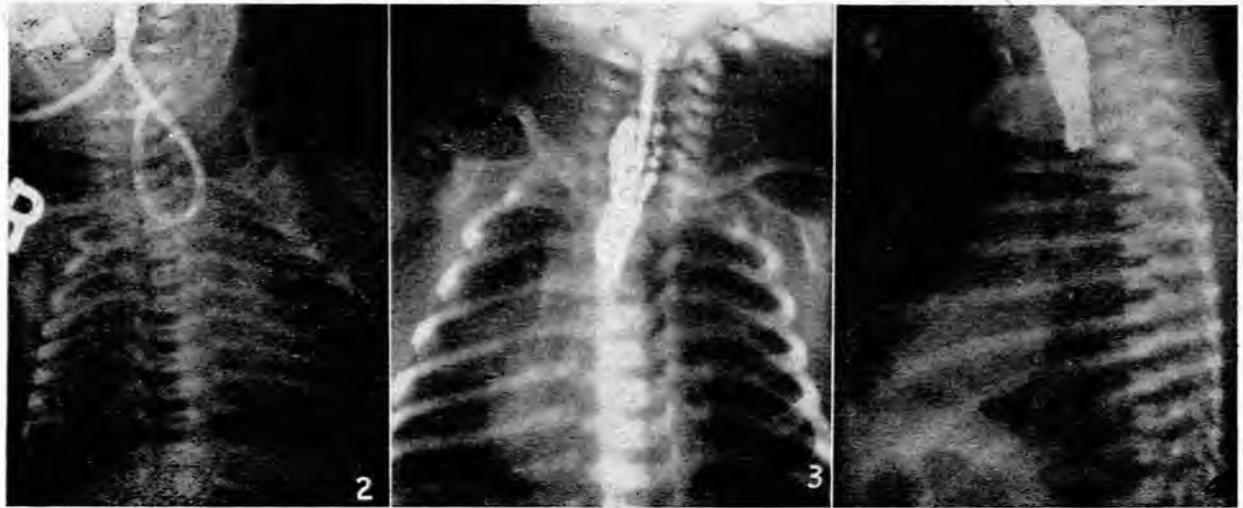


Fig. 2. Radiograph demonstrating coiling of catheter in upper blind pouch.

Fig. 3. Radiograph showing segmental atelectasis of right upper lobe and lipiodol outlining long upper blind pouch.

Fig. 4. Lateral radiograph showing upper pouch ending blindly. Also note air in abdomen indicating fistula between trachea and lower segment.

further exacerbations and progressive dyspnoea. Dehydration is soon followed by inanition and survival for longer than 5-7 days is exceptional—death resulting rather from pulmonary complications than from starvation.

The diagnosis is readily confirmed by the fact that a soft catheter cannot be passed down the oesophagus further than 10 cm. from the gum margin (Fig. 2).

Radiological investigation is useful, not only to confirm the diagnosis but to establish the type of anomaly. Straight films will demonstrate:

(a) Excess air in the stomach, indicating the presence of a fistula between the trachea and lower pouch. In such cases it is usually possible to effect an anastomosis.

(b) Absence of air in the stomach is commonly associated with an atretic lower segment and the usual anastomosis would be impossible. We have had one such case.

(c) The lateral film shows anterior displacement of the trachea with some compression of its posterior margin by the dilated hypertrophied pouch.

(d) The presence of inhalation atelectasis or pneumonitis—usually in the right upper lobe. This was present in at least 10 of our cases.

(e) The presence of rib and vertebral anomalies is noted. These were present in 8 of our cases.

A lipiodol swallow confirms the diagnosis. This is performed by passing a catheter into the proximal pouch, where it is obstructed at about 10 cm. (Fig. 2) and injecting 0.5 c.c. of lipiodol. Lipiodol outlines the pouch (Figs. 3 and 4) and the rare communication between it and the trachea will be demonstrated. It is essential to aspirate as much of the lipiodol as possible. In case 10 an excess of lipiodol overflowed into the lungs (Fig. 6).

The use of barium sulphate for this purpose cannot be condemned too strongly. The barium causes gross chemical bronchitis; 2 cases (operated on) died 6 and 8 hours after its

administration, and one other case had such gross bronchitis that surgery was impracticable.

#### PRE-OPERATIVE TREATMENT

The *pre-operative treatment* we adopt includes the following points:

1. *Postural drainage.* We used to adopt the Trendelenberg position to lessen spill-over of mucus from the blind upper pouch causing overflow inhalation pneumonitis; but more recently we have nursed the infants slightly head-up to prevent gastric secretions from passing up through the fistula and flooding the tracheo-bronchial tree.

2. Careful repeated intermittent aspiration of pharyngeal mucus and tracheo-bronchial secretions is employed.

3. An oxygen incubator is essential.

4. Steam is used in the incubator and, more recently, despite the suspected risks, *alevire* has been administered as a fine nebulized spray.

5. Antibiotic therapy is given to control pulmonary infection, viz, 50,000 units of penicillin G and 50 m.g. of streptomycin, both 6-hourly.



Fig. 5. Photograph showing incision used for the extra-pleural approach 10 days after surgery.

6. Ascorbic acid is given by injection to stimulate fibroblastic healing.

7. Vitamin K is administered, but not now in such large amounts as in the earlier cases.

8. Pre-operative intravenous hydration is rarely necessary in the first 4 days of life, but a cut-down in the left ankle is always set up immediately before operation for transfusion during surgery and for the assistance of the anaesthetist.

9. Bronchoscopy is performed on the table if there is lobar or pneumonic atelectasis or if there is an excess of tracheo-bronchial secretion.

Time of operation. We lost case 9, which was operated upon within 24 hours of birth. We feel that operation should not be undertaken till the second day of life, to ensure that postnatal lung expansion is complete. Although in our first 10 cases we operated within a few hours of diagnosis we feel that anaesthetist, theatre staff and surgeon are not able to give of their best in such prolonged and intricate surgery in the early hours; we have therefore prepared our more recent cases for surgery in the forenoon with, I think, improvement in our results.

#### Pre-operative Complications

1. Prematurity. There were no survivors in the 4 cases weighing less than 5 lb.

2. Atelectasis was present in 10 cases and was responsible for a number of post-operative chest complications.

3. Barium swallow was responsible for the death of 3 infants—2 of whom we unwisely operated upon in the hope of controlling the resultant chemical bronchitis.

4. Lipiodol pneumonitis occurred in one case. This should not happen if only 0.5 c.c. of lipiodol is used and if it is aspirated immediately after X-ray.

5. Ligation of the lower end of the oesophagus was performed in one case to prevent gastric reflux. A feeding gastrostomy was also performed. This infant was referred for surgery on the 27th day of life but unfortunately developed an empyema from leakage at the anastomosis.

6. Neonatal haemorrhage with a large ecchymosis of the abdominal wall was present in case 32. This infant did well after surgery, to die of intracranial haemorrhage on the third day.

#### THE OPERATION

##### Anaesthesia

There is no standard anaesthetic for these cases, but adequate ventilation and oxygenation, for which endotracheal intubation is essential, and controlled respiration at the time of anastomosis (because diaphragmatic movement tends to tear out the sutures) are essential for satisfactory transpleural surgery.

If pre-operative bronchoscopy is necessary I prefer doing this under anaesthesia. Diathermy coagulation of bleeding points saves a little time in the exposure but precludes the use of explosive anaesthetics, which may be an undesirable sacrifice for this economy. Once the pleura is entered mobilization is easier with controlled respiration, but it is not essential since the upper lobe is readily controlled with a small retractor. For adequate suturing of the anastomosis, however, absolute control of the respiration is, in my opinion, essential. Although relaxants are widely used to obtain this I personally prefer deep ether with rapid manual closed-circuit over-ventilation. Whatever technique is used, provided the infant

is pink and respirations controlled the result should be safe and satisfactory.

##### Technique of Repair

In our first 17 cases from 1948 to July 1953 we employed the right-sided extrapleural thoracic approach (Fig. 5). This approach is now only of historical interest in that it was used in the first successful case surviving the direct operation done by Haight<sup>11</sup> in 1943. For many years it proved a reasonable procedure, and some surgeons employed this approach as recently as September 1956.<sup>20</sup> Since July 1953, however, we have regularly employed a right-sided transpleural thoracotomy, either with resection of the 3rd or 4th rib or through the corresponding interspace. I believe this is quicker and gives better access to the anomaly and most authors agree with this opinion.<sup>21</sup>

On opening the pleura the lung is gently retracted downwards, the azygos vein divided, and the mediastinal pleura posterior to the trachea opened. Although the lower bulbous end of the upper blind pouch can, as a rule, readily be isolated and freed from the back of the trachea into the neck, it is preferable to isolate the upper fistulous end of the lower oesophagus first. Control of the fistula prevents further abdominal distension and makes ventilation easier. The oesophagus is isolated behind the right bronchus and as it is followed upwards it is carefully separated from the trachea. A small rubber catheter is passed around it for traction and the fistula isolated.

We have found great variation in the site of attachment of the fistula—once to the right main bronchus and twice at the level of the carina, but in the remainder always into the trachea but at a variable distance from the lower end of the pouch, several quite close to it—in a few 1 cm. distal but in most a little further down the trachea and in several 1 inch (2.5 cm.) beyond.

Once the fistula has been isolated an interrupted 00000 Deknatel minimal suture is passed through the upper border of its tracheal attachment. The fistulous track is now, divided, leaving a little cuff on the trachea as further interrupted sutures are placed and tied. The fistulous end of the oesophagus is now cut obliquely at a level where its lumen and musculature are adequate for anastomosis. A posterior layer of interrupted sutures are now passed from the pouch to the oesophagus (usually 6-8 sutures are possible) and after they have all been passed the tension is taken up on all of them and they are then tied individually. The pouch is now opened and a posterior interrupted mucosal layer is completed.

I now pass a fine catheter down the oesophagus and up through the pouch into the pharynx, from where it is partially withdrawn by the anaesthetist. The anterior mucosal layer of sutures is now inserted, with knots on the mucosa—the catheter prevents this layer from picking up the posterior mucosal layer. Finally the anterior muscular layer is placed; this is often difficult because the lower end is attenuated. The thicker pouch is then, as it were, rolled over on to the lower end and attached there. We have no hesitation, where necessary, in using only two layers of all-coats through-and-through sutures. The indwelling catheter is now withdrawn from the mouth. The anastomosis is anchored to the vertebral bodies to limit tension on the suture line. Penicillin (1 mega-unit) and streptomycin G1 are placed around the anastomosis and a catheter is passed through the 9th intercostal space for underwater drainage and the incision is completely sutured

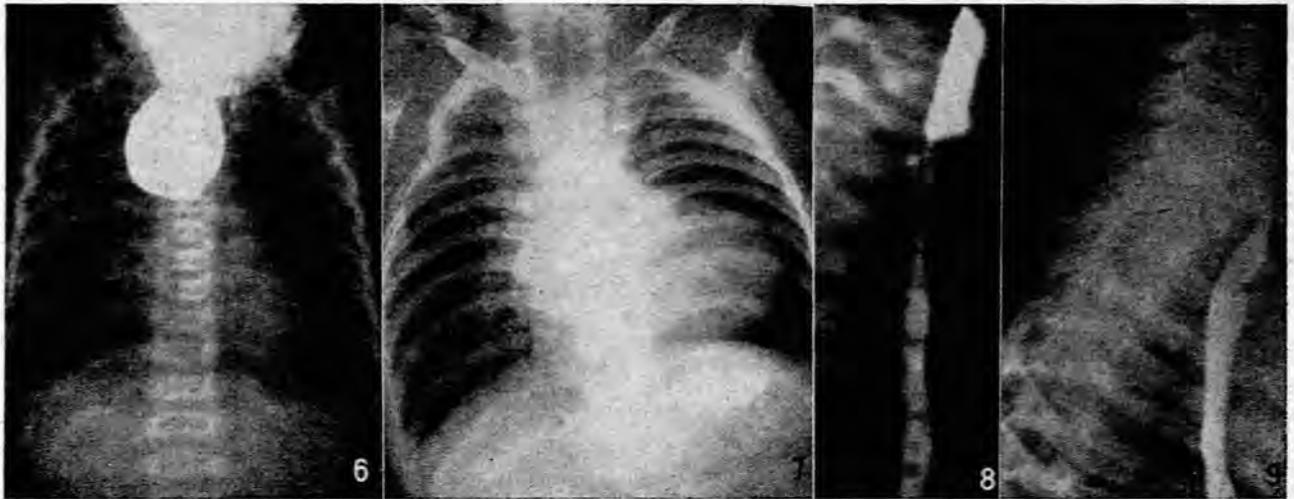


Fig. 6. Radiograph from an early case showing gross over-distension of pouch with lipiodol.

Fig. 7. Radiograph 10 days after successful surgery confirming patency of anastomosis in a case with the tetralogy of Fallot shown by the boot-shaped heart.

Fig. 8. Radiograph demonstrating local extravasation of lipiodol at site of anastomosis 5 days after surgery.

Fig. 9. Radiograph showing minimal narrowing at site of anastomosis two years after uneventful surgery in the first successful case.

in layers. If the lung has remained atelectatic, bronchoscopic toilet is again performed.

During the operation the blood loss is replaced and we have usually diluted 100 c.c. of blood with 100 c.c. of normal saline to ensure that there is no blockage of the polythene catheter during surgery. About 100-125 c.c. of this mixture is usually administered.

#### POST-OPERATIVE CARE

1. The infant is returned to the ward in an oxygen incubator which is humidified or into which alevaïre is introduced.

2. The pre-operative antibiotics are continued with for 7-10 days. Broad-spectrum antibiotics are prescribed when indicated.

3. Cautious pharyngeal suction through a soft rubber catheter is employed when necessary; it is essential in the first few hours, because oedema of the anastomosis does not allow the child to swallow its saliva.

4. The infant is turned from side to side every few hours to prevent hypostatic congestion and atelectasis. In 9 cases atelectasis was considered to be the cause of death.

5. Ascorbic acid and vitamin-B complex are given in the drip in the first few days and subsequently by injection or by mouth.

6. Fluid requirements are most carefully estimated and continued by intravenous drip. Great care is taken not to overload these infants and in the first few days we only use 5% dextrose. On the 3rd day 50 c.c. of Darrow's solution is given, and on the 4th day 50 c.c. of infants' plasma or serum. Sodium chloride has not been given in our cases in view of the saline-diluted blood transfusion which is given during the operation and continued very judiciously in the first few post-operative hours as indicated by the child's general condition and loss through the underwater catheter.

7. A bedside portable X-ray of the chest is taken on the 1st post-operative morning to assess any contralateral atelectasis and ipsilateral haemothorax or pneumothorax.

8. The intercostal catheter is usually removed on the 2nd post-operative day.

9. On the 4th post-operative morning if the anastomosis has been a satisfactory one, or on the 5th day if the anastomosis has been a little under tension, one drachm of sterile water is given orally drop by drop to lubricate the pharynx, as it were, and to stimulate swallowing. Half a drachm of lipiodol is then given orally and a portable X-ray is taken after 2 minutes and again after 5 minutes—the latter to disclose any delayed leak at the anastomosis and to confirm that the lipiodol has entered the stomach (Fig. 8). In 3 cases (11, 17 and 20) disruption of the anastomosis was evident, and resulted in death. In case 21 there was considerable delay at the cardio-oesophageal junction, and as no relief was obtained with Eumydrin a feeding gastrostomy was performed by Mr. W. Kark on the 8th day after surgery. This child subsequently did well and the gastrostomy was closed 3 months later.

10. Once the lipiodol swallow has shown that the anastomosis is patent and intact, a careful pipette feed of 1 drachm of sterile water with added streptomycin is given slowly each hour. The following day 2 drachms of dextrose water with streptomycin are given by mouth and if the infant has been able to cope with this quite satisfactorily  $\frac{1}{2}$  drachm of expressed milk is given and the amounts gradually increased until the infant is taking a normal diet for its age and weight. If larger or quicker feeding is embarked upon the child avidly takes the feed and, owing either to the trauma of the operation or possibly to some lack of adequate innervation of the pharyngo-oesophageal mechanism, choking and inhalation tend to ensue.

11. If convalescence is straightforward we discharge the patient as soon as possible to avoid the risk of long-continued hospitalization. Four have been discharged on the 10th day.

12. Gastrostomy has *not* been made a routine in our cases. Gross,<sup>22</sup> from a large experience in Boston, strongly recommends it to take the strain off the anastomosis. I feel it is an added surgical procedure not devoid of risk and that early

feeding is the best method of dilating the oesophagus. With this latter view Potts<sup>21</sup> agrees.

#### POST-OPERATIVE COMPLICATIONS AND MORTALITY

##### 1. Immediate

(a) Anaesthetic deaths, e.g. case 2, where spontaneous respirations did not return.

(b) Death from operative shock in the premature babes.

(c) Death due to inadvertent or erroneous pre-operative administration of barium, which caused uncontrollable bronchitis in 2 cases.

(d) Two cases died from associated anomalies, viz. ventricular septal defect and myelocele.

##### 2. Early

(a) Respiratory complications, which are inherent in the anomaly since the tracheobronchial tree is assaulted both by aspiration of infected material from the blind proximal pouch and by reflux of gastric secretions through the fistula:

(i) Atelectasis—segmental (unimportant), lobar (usually transient) or pneumonic (usually fatal).

(ii) Bronchitis and bronchopneumonia.

(iii) Pulmonary oedema from intravenous overload.

(iv) Anoxia—in the premature and in those operated upon too early.

Nine cases died from pulmonary complications; 2 cases recovered from mild transient atelectasis, and 2 suffered repetitive attacks of inhalation pneumonitis from incoordination of swallowing and 1 from cardiospasm.

(b) Due to disruption of the anastomosis:

(i) Empyema with oesophago-pleural fistula: This occurred in 3 of our cases.

(ii) Oesophago-cutaneous fistula.

(iii) Recurrent tracheo-oesophageal fistula.

These complications are usually fatal.

(c) Digestive disturbances: Diarrhoea when oral feeding is commenced is not unusual and is probably due to the irritant effect of food on the sterile empty bowel. In case 25 it proved intractable and the child died of a second attack of diarrhoea 2 months after operation.

(d) Oedema: due to protein lack or, more usually, excessive administration of salt.

##### 3. Delayed

(a) Lung abscess resulting from infection in an area of atelectasis or from inhalation of food in the early weeks of feeding before the swallowing mechanism has fully developed. This occurred in case 10, who has survived with a residual bronchiectatic abscess.

(b) Diarrhoea due to staphylococcal enteritis or moniliasis.

##### 4. Late

(a) Recurrence of tracheo-oesophageal fistula.

(b) Stricture at site of anastomosis (Fig. 9). This occurred in 5 cases (3, 4, 10, 13 and 20) and resulted in multiple minor incidents of aspiration episodes and in retention of oesophageal foreign bodies, to which these children appear prone. All the strictures have responded to dilatation.

(c) Bronchiectasis from obstructive inhalation pneumonitis developed in 2 cases.

#### RECOVERY RATE

The recovery rate of infants operated on for congenital tracheo-oesophageal fistula as reported by various authors, and in our own series, is indicated in the following table:

Author	Year	Patients	Recovery (No.)	Recovery %
Cameron Haight <sup>22</sup> ..	1952	131	57	43.4
R. E. Gross <sup>23</sup> ..	1953	233	109	46.8
C. Koop, <i>et al.</i> <sup>24</sup> ..	1954	74	34	45.9
H. W. Clatworthy <sup>25</sup> ..	1955	35	17	48.6
R. Nicks <sup>26</sup> ..	1955	14	5	36
R. Shaw <sup>27</sup> ..	1955	61*	25	41
		38†		
Willes Potts <i>et al.</i> <sup>21</sup>	1957	113	—	54
D. I. Adler and D. N. Fuller ..	1949-56	34	17	50

\* 1938-53. † 1948-53.

#### CONCLUSIONS AND SUMMARY

Congenital tracheo-oesophageal fistula with atresia is widely recognized clinically by excessive frothy oral mucus. Other congenital abnormalities are common but usually of a minor nature. Treatment cannot be delayed. Surgical repair offers a 50% survival rate and is possible only because of modern anaesthesia, fluid replacement and careful nursing.

The subject is reviewed, and the author's experience with 34 cases operated on at the Children's Hospital, Johannesburg, is reported.

The successes in this series have been attributable to the whole-hearted cooperation and supervision of the paediatricians at the Transvaal Memorial Hospital for Children, to whom I would like to express my deep appreciation viz. Drs. Seymour Heymann, S. N. Javett, A. E. Strawbaun and Jacques Theron. Sister Mackay has been in charge of these patients and to her and her staff I would like to pay tribute for their unflinching interest and meticulous care in their management. To successive housemen the survivors owe their existence. Of the 34 cases, 11 were operated upon by Mr. Dennis Fuller and one by Mr. G. Katz.

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