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CONGENITAL OESOPHAGEAL ATRESIA AND TRACHEO-OESOPHAGEAL FISTULA*

A REVIEW OF 36 PATIENTS

BY

K. D. ROBERTS

From the Children's Hospital, Birmingham

"Atresia of the oesophagus, with or without tracheo-oesophageal fistula, has been a baffling problem for the surgeon. If there is any surgeon who has attempted to save the lives of patients suffering from these malformations who has not had many disappointments and numerous trials and tribulations, I have not heard of him."—W. E. LADD (1944).

In this paper the clinical features, complications of operation, and results of treatment are considered in a personal series of 36 babies suffering from congenital abnormalities of the oesophagus. The patients were treated in the Children's Hospital, Birmingham, during the period June, 1953, to June, 1957. During this time, 44 such babies were admitted and, as an indication of the relative occurrence of these abnormalities, it may be stated that the average annual admissions for congenital hypertrophic pyloric stenosis were 85.

The classification of oesophageal abnormalities used is that described by Gross in 1953 (Fig. 1). Thirty-three babies belonged to Group C and one each to Groups A, D, and E.

In 1955, Roberts, Carré, and Inglis noted that Group C could be divided into two subgroups (Fig. 2), namely, Group C(i) in which the fistula enters the bifurcation of the trachea or the bronchus and there is a considerable length of atresia; these cases are unfavourable for primary anastomosis (18 patients); Group C(ii) in which the fistula enters the trachea high up and the upper oesophageal segment overlaps the fistula or the gap between the segments is not great (14 patients).

In one baby in the series, there were two fistulae between the lower oesophagus and the trachea, one being at the thoracic inlet and the other at the bifurcation of the trachea. No similar case has been described in the literature and this one has provisionally been classified in both categories as C(i)–(ii).

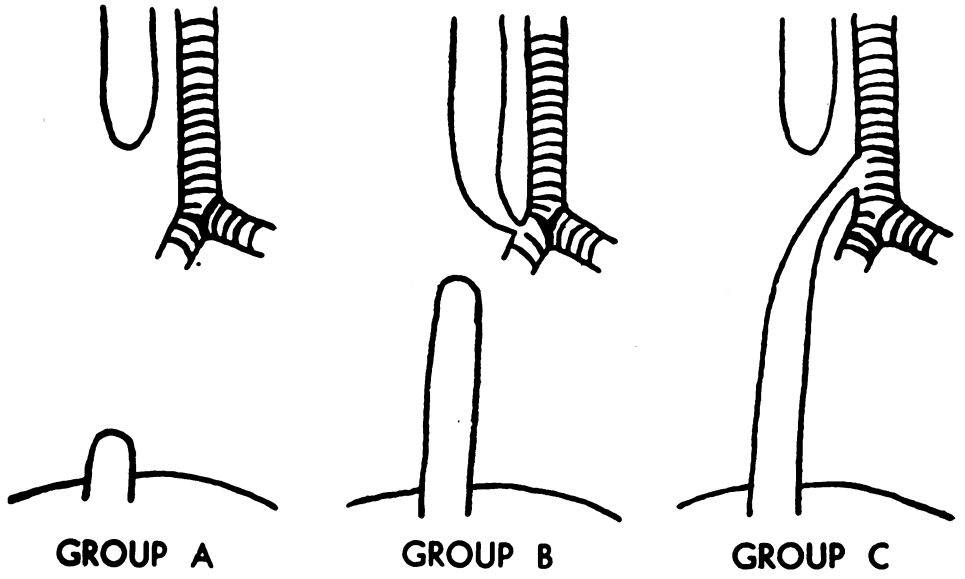
* Based on a paper read before the Society of Thoracic Surgeons of Great Britain and Ireland on November 22, 1957.

A feature of the condition which has been neglected is the association of atresia with maternal hydramnios. It has long been believed that atresias of the alimentary tract may cause hydramnios, as the foetus is unable to swallow and absorb amniotic fluid, and the incidence is particularly marked in the case of oesophageal atresia (Ballantyne, 1904; Scheurer, 1928; Brigham, 1929; Meyer, 1929). In the present series of 36 babies, 35 had oesophageal atresia and hydramnios had been present in 13; the incidence may well be higher than this, as details of the pregnancy have not always been available.

CLINICAL SYMPTOMS AND SIGNS

The essential feature of total oesophageal obstruction in the newborn is the inability of the baby to swallow its saliva, so that a characteristic fine frothy mucus is continually produced in the mouth, unlike the rather coarse bubbles which are frequently found for a time after birth (Belsey and Donnison, 1950). The froth may be bile-stained owing to regurgitation of alimentary contents through a tracheo-oesophageal fistula into the trachea. Lecutier (1955) has given an interesting account of "paradoxical haematemesis" in a baby with a Group C abnormality, and one of the babies in the present series was said to have "vomited" blood shortly after delivery.

Signs of respiratory obstruction with stridor may be due to a fold of mucous membrane in the trachea at the site of the fistula (Franklin and Graham, 1953). Stridor was noted before operation in only one baby in the series and recurred after operation, but no tracheal fold was found at post-mortem examination to account for it. If feeds are attempted, "spill-over" from the obstructed oesophagus into the larynx causes cough and cyanosis. Where oesophageal atresia is suspected no feeds should be given until full investigation of the oesophagus has shown that



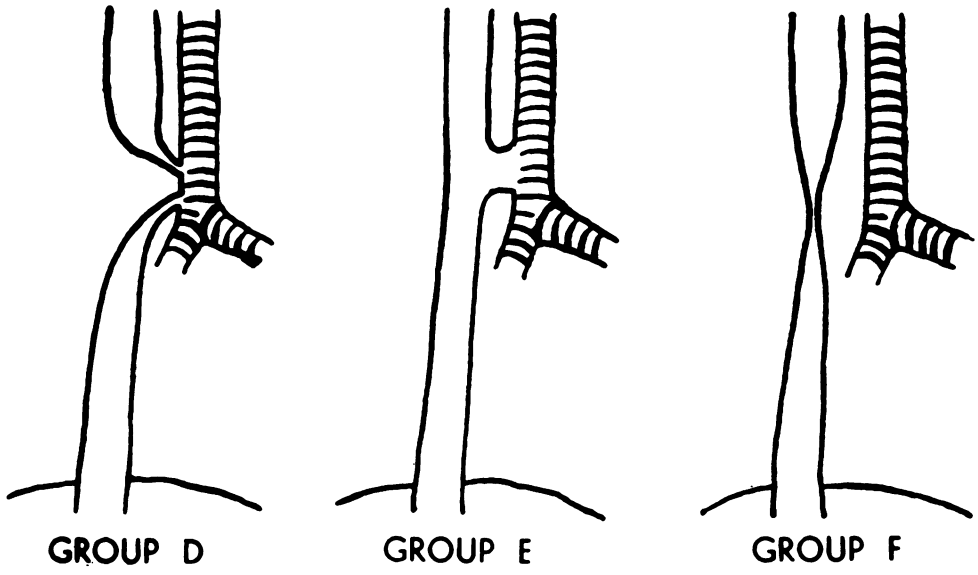


FIG. 1

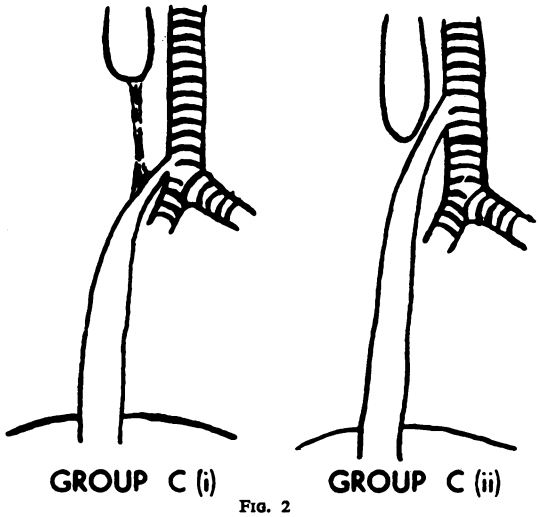


FIG. 2

FIG. 1.—The varieties of oesophageal abnormality. (Modified after Gross.)

FIG. 2.—The subdivisions of Group C.

this can be done. Owing to the inevitable aspiration of some oral secretions most infants have a "wet" chest on admission, while atelectasis (usually of the right upper lobe) will be indicated by diminished air entry.

Occasionally, air may be forced into the stomach when the infant cries so that the abdomen is distended and tympanitic.

The clinical features vary according to the type of abnormality and are summarized in Table I.

TABLE I
CLINICAL FEATURES IN VARIOUS TYPES OF OESOPHAGEAL ABNORMALITY

Clinical Feature	Group A	Group B	Group C	Group D	Group E	Group F
Excess oral mucus	Always	Perhaps	Always	Perhaps	No	Perhaps
Cough and cyanosis with feeds	,,	Always	,,	Always	Perhaps	,,
"Wet" bronchial tree	Usually	May be severe	Usually	May be severe	,,	,,
Abdominal distension	Never	Never	Frequent	Frequent	Frequent	No

In Groups B and D, if the fistula from the upper oesophagus is large the infant is likely to have a severe aspiration pneumonia on admission. If the communication between the oesophagus and trachea is small in Group E cases there may be only slight difficulty with feeds, cough and cyanosis only occurring in certain positions, and the condition may not be incompatible with survival. Thus two reported patients were aged 6 years (Imperatori, 1939) and 4 years (Haight, 1948) at the time of operation.

Of the present cases, 31 of Group C showed excessive oral mucus as did also the babies in Group A and Group D. The infant's bronchial tree in the last case was not excessively "wet," as the fistula between the upper oesophagus and the trachea was only small and was, in fact, not recognized before or during operation but at post-mortem examination. The only patient in Group E had no oral frothy mucus, but in this patient the fistula was large enough to cause coughing and cyanosis at each of three attempted feeds.

When oesophageal atresia is suspected, an attempt should always be made to demonstrate this by introducing an oesophageal catheter which can be passed no further than 10 to 12 cm. from the alveolar margin. This was done in 26 of Group C cases and showed obstruction to be present; it was also a positive finding in both cases of Groups A and D. It is important to use a rigid enough tube, as this may otherwise coil up in the upper oesophageal pouch and appear to have reached the stomach; this happened in two babies and tube feeds were given, resulting in overspill into the larynx; passing a larger tube confirmed the presence of oesophageal obstruction.

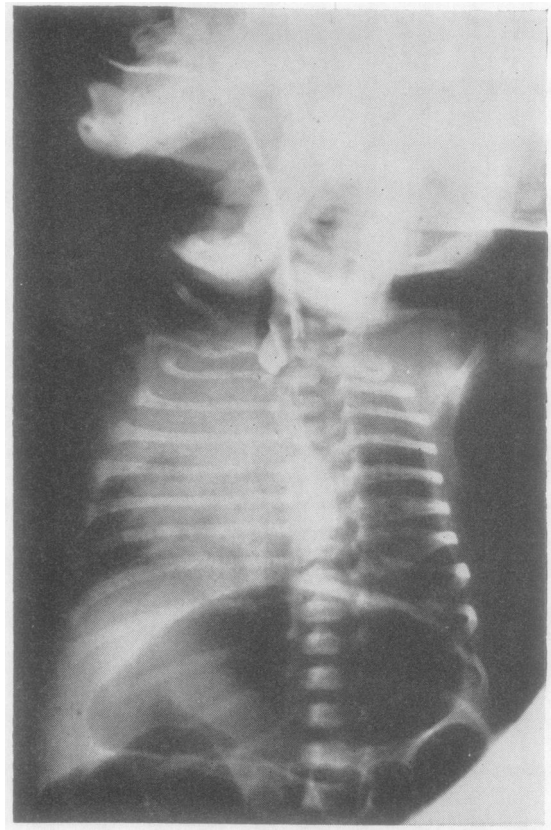
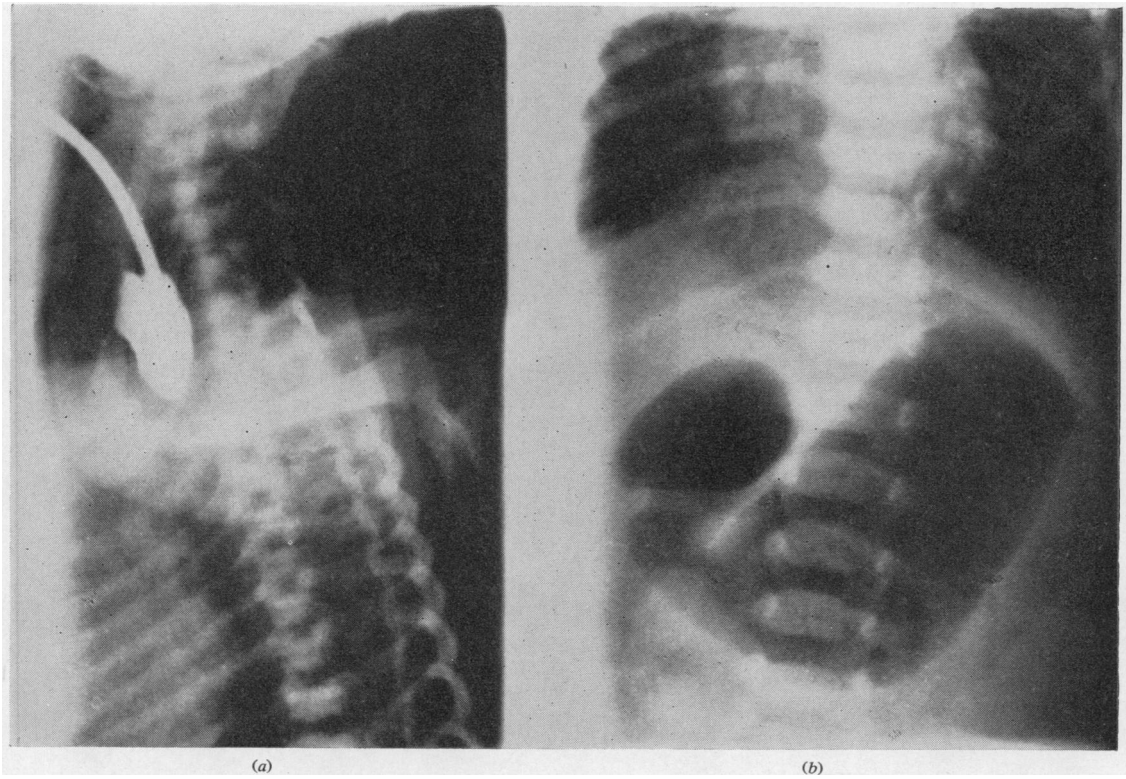


FIG. 3.—Group C abnormality: note the opacity of the right upper lobe and the distended stomach.

RADIOLOGICAL INVESTIGATION

Radiological examination of a baby suspected of having an oesophageal abnormality should begin with fluoroscopy in antero-posterior and lateral views in order to evaluate the state of the lungs and note the appearance of the stomach and small bowel. It is not possible to distinguish radiologically between atelectasis and aspiration pneumonia in these babies and it may only be possible to state that there is an abnormal increase in density (Fig. 3) in one or more lobes (Holt, Haight, and Hodges, 1946).

Where gas is seen in the stomach and intestines a fistula between the lower oesophagus and trachea must be present, but the absence of air does not exclude the presence of a small fistula; in the



(a) (b)

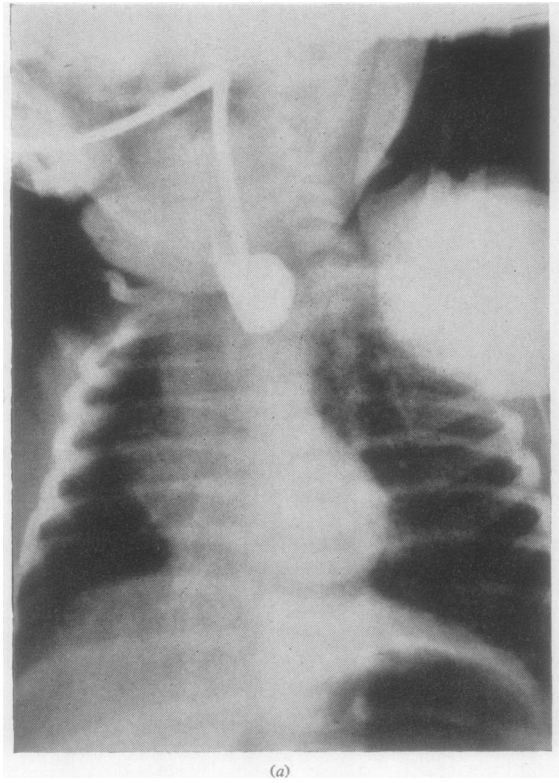
FIG. 4.—(a) Oesophageal atresia, with tracheo-oesophageal fistula and duodenal atresia. (b) The stomach and first part of the duodenum are distended with air, but there is no gas in the small or large intestine.

present cases the fistula was always large enough to transmit air to the gastro-intestinal tract. A duodenal atresia may be revealed by gaseous distension of the stomach and duodenum and no gas shadows in the remainder of the intestine (Fig. 4).

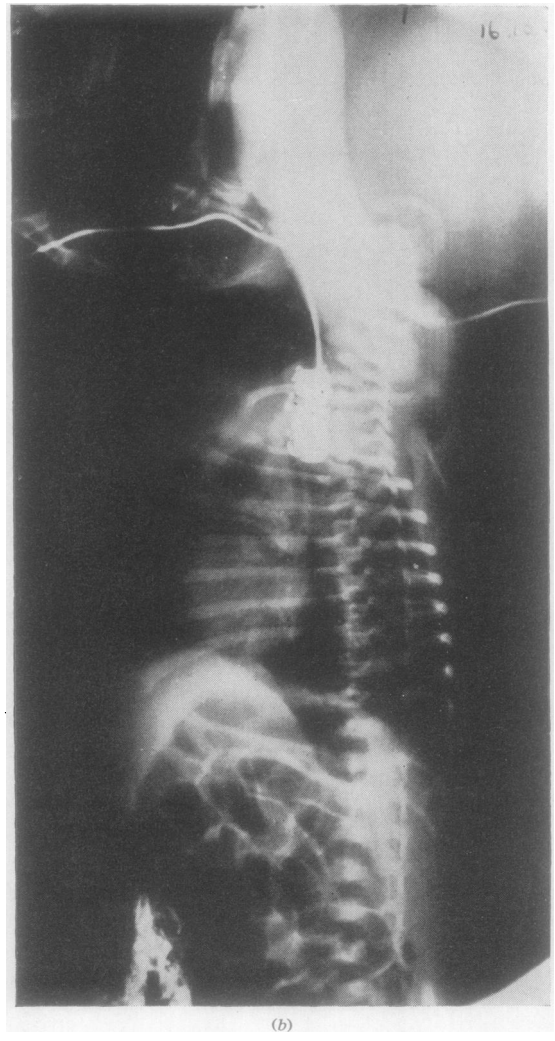
A "pencil-like" airway may be seen extending from the region of the trachea to the stomach, indicating the lower oesophageal segment. In the lateral view, the dilated, hypertrophied upper oesophageal pouch may produce a characteristic anterior displacement and narrowing of the trachea (Selander, 1941). Screening may also reveal other developmental defects such as congenital cardiac abnormalities.

After fluoroscopic examinations a contrast medium should be introduced into the upper oesophagus; barium sulphate suspension must not be used owing to its irritant effect should any enter the bronchial tree. Iodized oil ("lipiodol") should be put in through an oesophageal catheter (Roberts and others, 1955), and the technique described by Astley (1956) is satisfactory. This consists of introducing a soft rubber catheter into the oesophagus under radiological control, the catheter having first been filled with iodized oil. When the catheter is just in the oesophagus a few drops of oil are injected; these outline the rounded termination of the oesophageal pouch at the level of the second to fourth thoracic vertebrae (Fig. 5). This blind pouch shows up-and-down excursion with respiratory movement. Too much oil increases the risk of spill-over into the larynx and trachea, and is a serious error of technique. Radiographs are taken and the catheter is withdrawn after aspirating all the oil from the oesophageal pouch. In Groups B, D, and E oil may pass directly into the bronchial tree if the fistula is large enough. An important indication of the presence of the fistula to the upper oesophagus in Groups B and D is the absence of the typical dilatation of the upper oesophageal pouch seen in Group C. This was not appreciated in the only baby in Group D in the series, and the presence of the upper fistula was not diagnosed radiologically or found at operation, being only discovered at necropsy; the upper fistula was small and did not appear to transmit iodized oil to the trachea at fluoroscopy, but oil spilled over into the larynx,

FIG. 5.—(*a*) Antero-posterior view of oesophageal atresia and tracheo-oesophageal fistula, Group C; note the typical dilatation of the upper oesophageal segment and the gas in the stomach. (*b*) Lateral view of Group C abnormality; again note the distended oesophageal pouch and gas in the intestines.



(*a*)



(*b*)

an error of technique (Fig. 6). A similar case was reported by Krediet (1955), in which the upper fistula did not fill with iodized oil and was not discovered until after death. Krediet felt that there were no routine means by which this rare abnormality could be diagnosed where radiological examination provided no clue.

A Group E abnormality may be difficult to demonstrate. Astley (1956) recommended that the infant should be examined in a variety of postures, particularly prone and with varying degrees of obliquity. Ferguson (1951) has stated that even if preliminary examination with iodized oil has excluded oesophageal atresia, barium sulphate should be avoided, and has pointed out that recurring or persistent pneumonitis may be due to a small tracheo-oesophageal fistula. Ferguson suggested passing two thin latex rubber balloons into the oesophagus and inflating them to occlude the lumen, one balloon being in the lower oesophagus, the other in the upper. A catheter is let down to the segment of oesophagus between the balloons and air is injected to distend it. A small amount of iodized oil is injected and the increased oesophageal pressure forces the oil into the trachea (Fig. 7).

In the only Group E abnormality treated at the Children's Hospital barium sulphate suspension had been given at another hospital; this revealed the fistula, and fortunately the contamination of the bronchial tree did not have serious effects (Fig. 8).

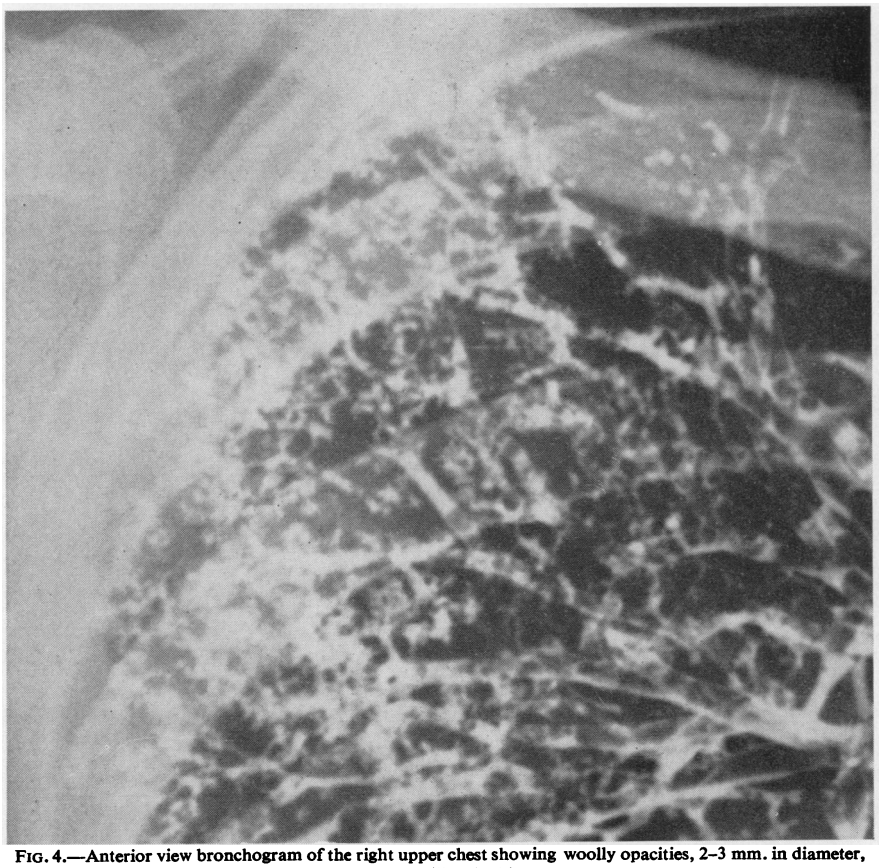


FIG. 4.—Anterior view bronchogram of the right upper chest showing woolly opacities, 2–3 mm. in diameter, towards the axilla. (×1.)

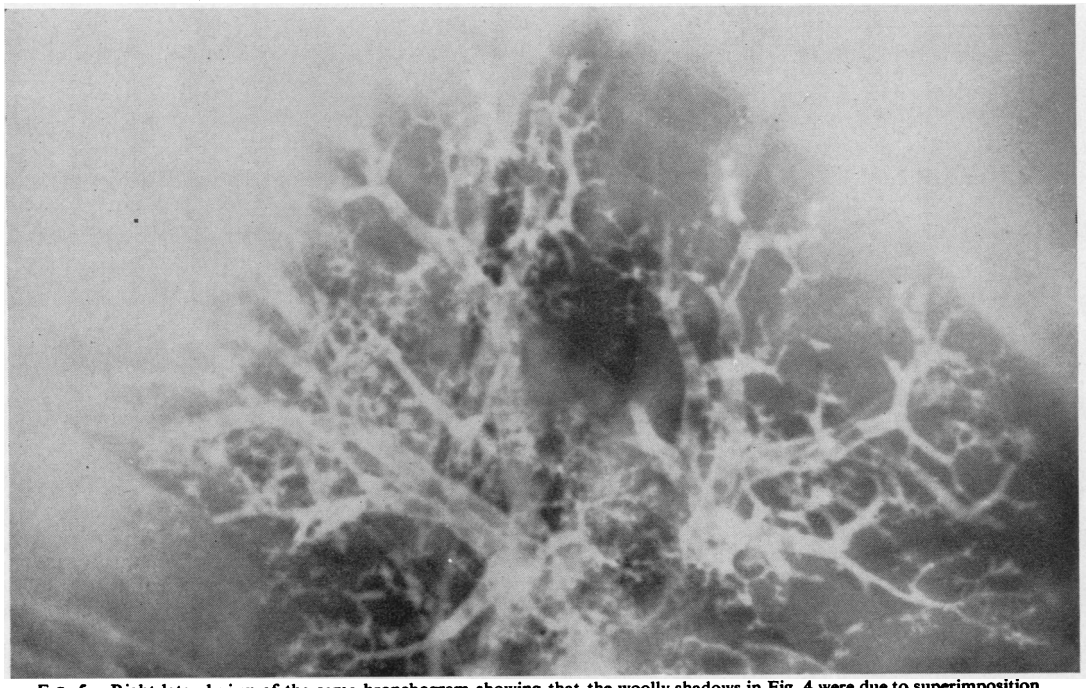


FIG. 5.—Right lateral view of the same bronchogram showing that the woolly shadows in Fig. 4 were due to superimposition of line shadows. (×1.)

Other Abnormalities

Congenital abnormalities are often multiple, and other defects were present in seven of the 36 babies. These are summarized in Table II.

Table II
DEFECTS IN ADDITION TO OESOPHAGEAL ABNORMALITY

Case No.	Other Defects
16	Persistent ductus arteriosus
20	Incomplete rotation of the gut; agenesis of right kidney; pelvic left kidney
22	Absent thumbs; absent left radius
28	Right arching aorta
29	Imperforate anus; rectovesical fistula; agenesis of right kidney
30	Ventricular septal defect
33	Mongolism; suprabiliary duodenal atresia; Meckel's diverticulum; incomplete rotation of the gut

Treatment

It is not the purpose of this paper to discuss details of operative treatment and management. Whenever possible, a primary oesophageal anastomosis was carried out, but where tension was too great or other factors contraindicated an anastomosis, the first stage of a staged procedure was done, that is, closure of the fistula, left cervical oesophagostomy, and Stamm gastrostomy. The mortality in the 36 patients is given in Table III.

On considering the type of operation performed initially in the 33 Group C babies, it is apparent that one may anticipate a high survival from primary anastomosis in patients of Group C(ii) in contradistinction to those in Group C(i) where anastomosis is technically more difficult (Table IV).

Table III
MORTALITY IN 36 CASES OF OESOPHAGEAL ABNORMALITY

Group	Total No. of Patients	Primary Anastomosis	First Stage	Second Stage	Died
A	1	—	1	—	1
C	33	25	—	—	10
			8	—	3
				5	3
D	1	1	—	—	1
E	1	1	—	—	0

Table IV
MORTALITY AFTER VARIOUS PROCEDURES IN DIFFERENT TYPES OF GROUP C ABNORMALITY (33 CASES)

Operation	Group	No. of Cases	Deaths
Primary anastomosis ..	C(i)	11	7
,, ,, ..	C(ii)	13+(1)*	3
,, ,, ..	C(i)–(ii)	1	0
,, ,, ..	C(i)	7	3
,, ,, ..	C(ii)	(1)*	0

* This baby first had a primary anastomosis, but later a first-stage operation had to be done owing to severe stricture formation and recurrence of tracheo-oesophageal fistula.

Five babies underwent a second-stage reconstruction operation, two attempts being made in one of them. The survival in these babies is shown in Table V.

Table V
MORTALITY IN FIVE BABIES UNDERGOING RECONSTRUCTIVE PROCEDURES

Case	Operation	Result
26	Oesophago-gastrostomy ..	Lived*
27	,, ,, ...	Lived
31	(1) Colon replacement	
	(2) Oesophago-gastrostomy ..	Died†
32	Colon replacement	Died
33	,, ,, ...	,,

* Died at 3½ years of age as a result of a road accident.
† At the first reconstructive procedure the interposed colon became black due to traction on the vascular pedicle and was therefore excised.

Complications

Pre-operative evidence of aspiration of mucus into the bronchial tree was proved radiologically in 16 patients, the commonest site being the right upper lobe. In the babies surviving operation the affected lobe eventually re-expanded. New areas of lung opacity developing usually indicated further spill into the bronchial tree due to incoordination of swallowing, stricture formation, or recurrence of the tracheo-oesophageal fistula. Other threats to survival were a variety of conditions causing intrapleural tension.

Post-operative Oesophageal Stricture.—Some narrowing of the oesophageal anastomosis is common after a primary anastomosis for atresia. Significant stenosis leads to difficulties in feeding and, more important, to the overspill of oral secretions into the bronchial tree. Strictures may be divided into two groups: early, developing within six weeks of the anastomosis, and late, occurring more than six months after operation or persisting from the early stage (Figs. 9 and 10).

In nine babies early strictures were found and in three late, one of these babies also having had an early stricture but apparently being well for 17 months after discharge from operation (Table VI). An early stricture is caused by necrosis due to tension at the suture line. Thus any attempt to excise an early stricture and re-anastomose the oesophagus is likely to fail, as the oesophagus has not had time to grow and adequate length will not be available. A plastic operation was performed in one baby, but it continued to aspirate mucus into the bronchial tree and died. Re-anastomosis was done for a stricture and a recurrence of a

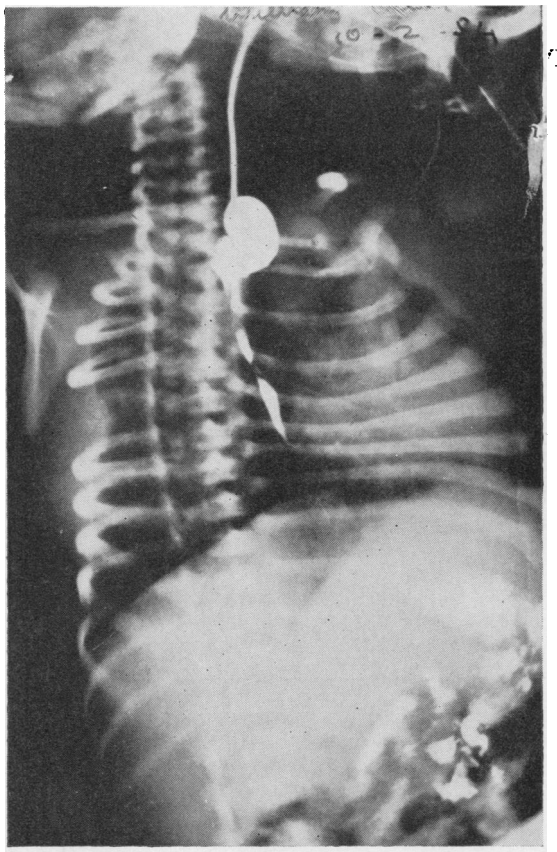


FIG. 9 (*a*)

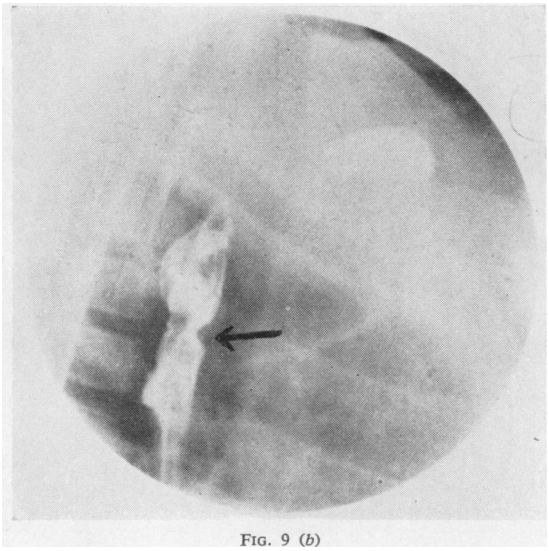


FIG. 9 (*b*)

FIG. 9.—(*a*) Severe stricture at the site of anastomosis 10 days post-operatively; oral feeds were supplemented by gastrostomy feeds. (*b*) The same patient aged 2½ years.

FIG. 10.—Oesophageal stricture 20 months after primary anastomosis.

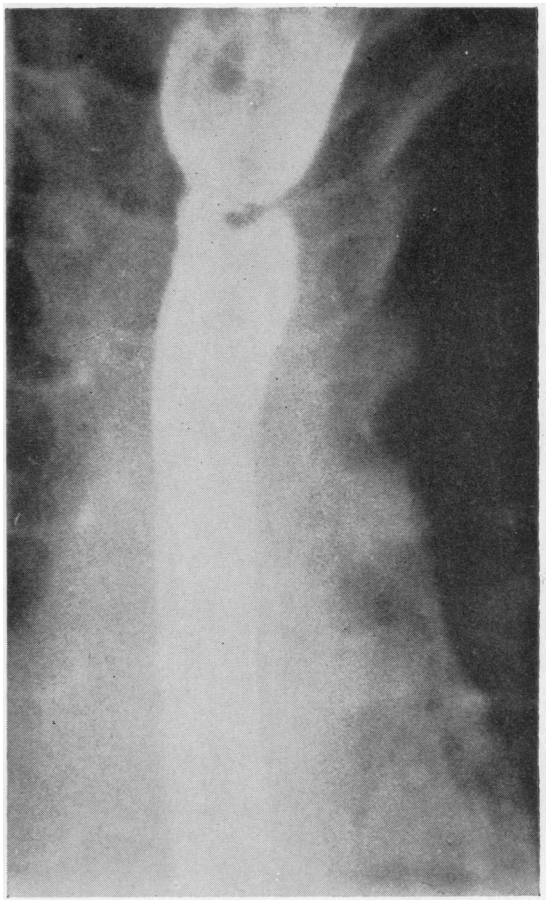


FIG. 10

TABLE VI

EFFECT OF OESOPHAGEAL STRICTURE AND ITS TREATMENT ON SURVIVAL

Case No.	Type of Stricture	Treatment	Result
4	Early	Prolonged gastrostomy feeds	Survived
9	,,	None needed during first admission	Survived but stricture persisted. See below
10	,,	Prolonged gastrostomy feeds	Survived; later had recurrence of tracheo-oesophageal fistula
11	,,	,, ,,	,,
21	,,	,, ,,	Died; lung abscess due to penetration of stomach, diaphragm, and left lower lobe by gastrostomy tube
22	,,	(1) Gastrostomy (2) Plastic operation	Died
23	Early + recurrence of tracheo-oesophageal fistula	Re-anastomosis and closure of fistula	,,
31	,, ,,	(1) Dilatations (2) Conversion to first-stage operation	Survived
36	Early	Prolonged gastrostomy feeds	,,
6	Late	Dilatations	,,
7	,,	,,	,,
9	,,	,,	,,

tracheo-oesophageal fistula in one baby; a tracheostomy was necessary as further overspill of mucus was not prevented and eventually the anastomosis broke down to cause a right pyopneumothorax from which the baby died. Oesophageal dilatation in the acute stage is also unlikely to succeed and failed in the only baby in which it was attempted.

From the experience of the above nine patients, the management of an early stricture has been developed. If overspill of mucus is not excessive, and a naso-gastric polythene tube can be passed through the stricture, this provides a satisfactory method of giving feeds, while the tube acts as an indwelling bougie; when an iodized oil swallow with the tube *in situ* shows an adequate lumen oral feeds can be resumed, supplemented at first by tube feeds. A difficulty with a naso-gastric tube may be atonicity of the upper dilated oesophageal segment so that saliva cannot be swallowed past the tube through the stricture and spills over; a gastrostomy is then necessary for feeding. An unusual cause of death in one patient was perforation of the stomach, diaphragm, and left lower lobe by the gastrostomy tube, ultimately causing a lung abscess. When the stricture fails to respond to conservative measures (in three of the eight cases in the present series) a procedure likely to prevent death from aspiration pneumonia is a thoracotomy to divide the oesophagus, the lower segment being closed and the upper end exteriorized as a cervical oesophagostomy: a gastrostomy is done for feeding.

Late strictures have occurred in three patients and have responded to oesophageal dilatation. The difficulties with the late cases are nutritional, as the infant is unable to swallow solid foods. Mucus overspill is not a feature, and corrective operations do not seem to be necessary.

RECURRENCE OF THE TRACHEO-OESOPHAGEAL FISTULA.—As with oesophageal stricture, two groups of recurrent fistula have been found (Table VII). Early fistulae are those developing within

TABLE VII

EFFECT OF RECURRENCE OF TRACHEO-OESOPHAGEAL FISTULA AND ITS TREATMENT ON SURVIVAL

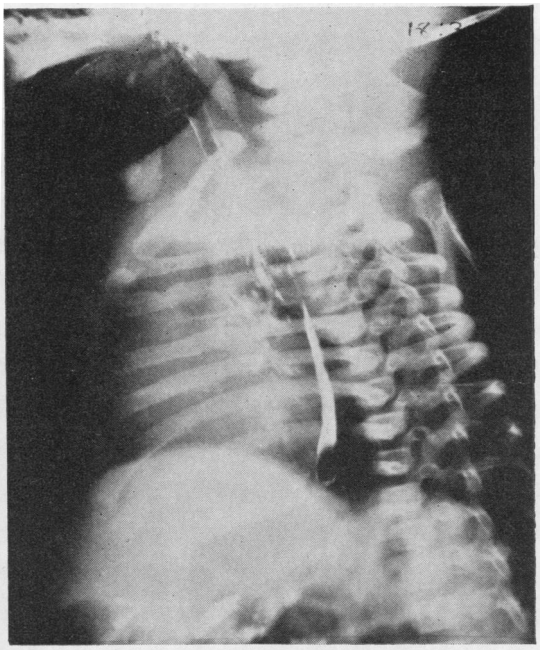
Case No.	Type of Recurrence of Tracheo-oesophageal Fistula	Treatment	Survival
23	Early	Operative closure	Died
31	,,	Conversion to first-stage operation	Survived
3	Late	Operative closure	,,
10	,,	,, ,,	,,
11	,,	,, ,,	,,

six weeks of the primary operation, and late recurrences are those found after three months from operation.

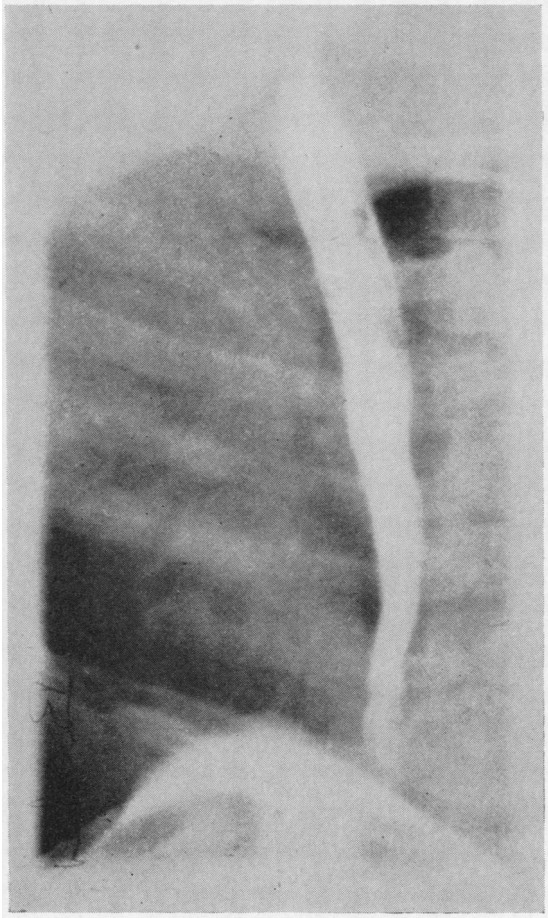
Two early recurrences were diagnosed. In one the addition of a severe oesophageal stricture influenced the attempt to excise the stricture, close the fistula, and re-anastomose the oesophagus, but leakage from the anastomosis caused the baby's death. The other early recurrence was associated with a stricture, but, in the light of the previous case, at re-operation the oesophagus was divided, the lower end closed and the upper end exteriorized. The fistula closed and a feeding gastrostomy was done; the baby survived but died after a reconstruction operation.

Three late recurrences were found. In one baby it was successfully closed at the age of 7 months (Fig. 11). The other two were successfully operated on at the ages of 1 year and 7 months respectively, and in both there had been an early oesophageal stricture.

The coexistence of a severe oesophageal stricture with recurrence of a tracheo-oesophageal fistula in the two early cases, and the fact that in two of the late recurrences an early stricture had been present, suggests to us that the cause of recurrence is necrosis at the oesophageal suture line. Certainly in one early recurrence at operation the oesophagus communicated with the

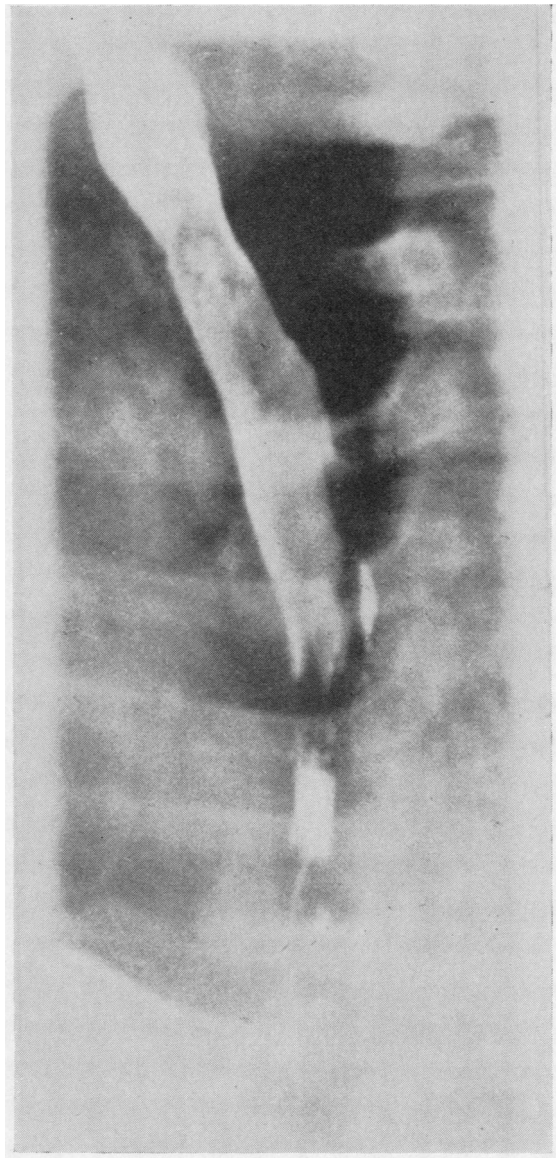


(a)



(c)

FIG. 11.—(a) Recurrence of tracheo-oesophageal fistula three months post-operatively. (b) Recurrence of tracheo-oesophageal fistula seven months post-operatively; note the "lipiodol" lying outside the oesophagus. (c) Normal oesophagus following operative closure of the fistula.



(b)

trachea via a small peri-oesophageal abscess, and in the other there was recent acute mediastinitis. However, it is possible that in one of the late cases the fistula may have recurred owing to a portion of fistula being left attached to the trachea at the primary anastomosis as the appearance at the second operation suggested; infection in a "tracheal diverticulum" might lead to abscess formation in the mediastinum (comparable to a bronchial stump abscess) and the abscess could later rupture into the oesophagus.

From the above cases the management of a recurrent fistula can be described. First, an early fistula, being associated with recent infection, should not be treated by early operative closure, as this is likely to break down. Whenever possible the baby should be tided over with naso-gastric tube or gastrostomy feeds until the acute mediastinal infection has subsided. Should operation be necessary, for example, if a coexistent stricture leads to considerable mucus overspill into the bronchial tree, then the correct procedure appears to be an exteriorization as described for early stricture.

The second point is that a recurrent fistula is unlikely to close spontaneously. Such manœuvres as cauterization of the fistula with silver nitrate through a bronchoscope (attempted in one baby) are of doubtful value, as sufficient cauterization to destroy the fistula would also cause breakdown and mediastinal abscess. In one baby the fistula apparently closed after a gastrostomy, but at a later examination it was still open and operative closure was necessary.

Cough with feeds, perhaps associated with cyanosis, may be due to oesophageal stricture or a recurrence of tracheo-oesophageal fistula. The latter may be difficult to demonstrate radiologically, although the persistence of a radiological picture of bilateral pneumonitis, or of a recurrent pneumonitis, is suggestive. Since the fistula takes an oblique course upwards from the oesophagus to the trachea, radiological screening in a variety of positions (particularly prone) after an iodized oil swallow is necessary. Ferguson's method (1951) may be used. Should repeated examinations with iodized oil be negative, but the clinical picture and radiological evidence of pneumonitis still point to the presence of a fistula, bronchoscopy may be helpful, as mucus or food debris may be seen to extrude from the fistula.

INCREASED INTRAPLEURAL TENSION. — Tension effects after primary anastomosis may be due to leakage of air from the trachea, or a breakdown of the oesophageal anastomosis. A tracheal leak occurred in one baby; during dissection of the upper oesophageal segment a small perforation was made in the trachea. Although this was sutured, and no apparent air leak was demonstrable, this was probably due to close apposition of the endotracheal tube. On removing this at the end of the operation and inflating the lungs through a face-mask a tension pneumothorax developed, causing cardiac arrest; although the tension was relieved and the heart restarted, death occurred later from cerebral damage. In addition to this experience, a tension pneumothorax occurred in another baby, the leak being presumed to occur through the sutured tracheal end of the fistula, but its presence was recognized in time and an intercostal tube inserted; this baby survived.

Oesophageal leakage occurred in two babies after primary anastomosis. In one the condition was treated by intercostal tube drainage and gastrostomy feeds until the oesophagus healed and the baby survived. The other baby had an operative closure of a perforation below the anastomosis (possibly through an avascular area of oesophagus), this being necessary as the intercostal tube repeatedly obstructed with debris so that tension recurred, but the baby died from atelectasis due to inhaled mucus. In a third baby there was a leakage from the anastomosis after reoperation for an oesophageal stricture and recurrence of a fistula, and this baby died from a mediastinal abscess and suppurative pneumonitis, although the lung had re-expanded after drainage.

The acute mediastinal infection which occurs when an oesophageal leak develops is best treated by ensuring lung expansion by underwater-sealed intercostal drainage, and maintaining nutrition by gastrostomy feeds. It sometimes happens that most of a gastrostomy feed is regurgitated into the pleura through the oesophageal perforation, presumably because (for some reason unknown) the cardia becomes incompetent. This occurred in the baby described above and responded to withholding gastrostomy feeds for a week, hydration being maintained parenterally. Should the cardia not become competent at the end of this time, so that all the gastrostomy feed drains via the pleural tube, a feeding jejunostomy may be necessary.

After reconstructive procedures, distension of the intrathoracically placed viscus (stomach or colon) may seriously embarrass respiration. In one baby, distension of the stomach in the left pleural cavity produced the clinical and radiological appearance of a tension pneumothorax (Fig. 12). It proved impossible to pass a tube into the stomach as it was held up at the anasto-

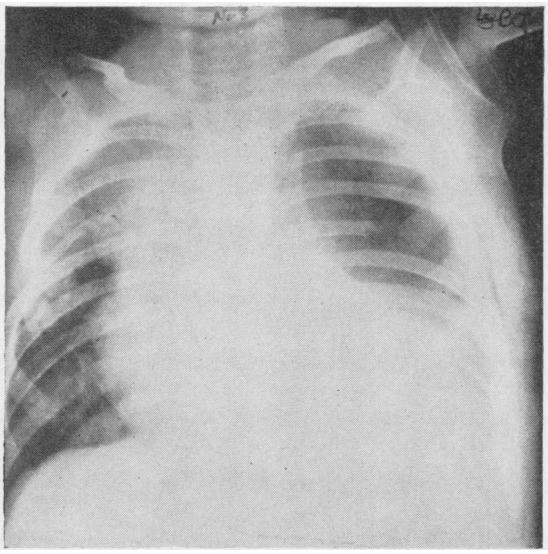


FIG. 12.—Gaseous distension of the stomach following cervical oesophago-gastrostomy. The left lung is collapsed and the mediastinum displaced to the right, causing right upper lobe collapse.

mosis, and the stomach could not be aspirated with a needle, so thoracotomy and transpleural gastrostomy were done; the baby survived. In another oesophago-gastrostomy a pyloro-myotomy was done to prevent post-operative pyloro-spasm and the stomach drained by a transpleural gastrostomy and tension did not occur. A sufficiently large oesophageal tube to evacuate the stomach cannot be left down in a small infant, as this would obstruct the anastomosis leading to saliva spilling over.

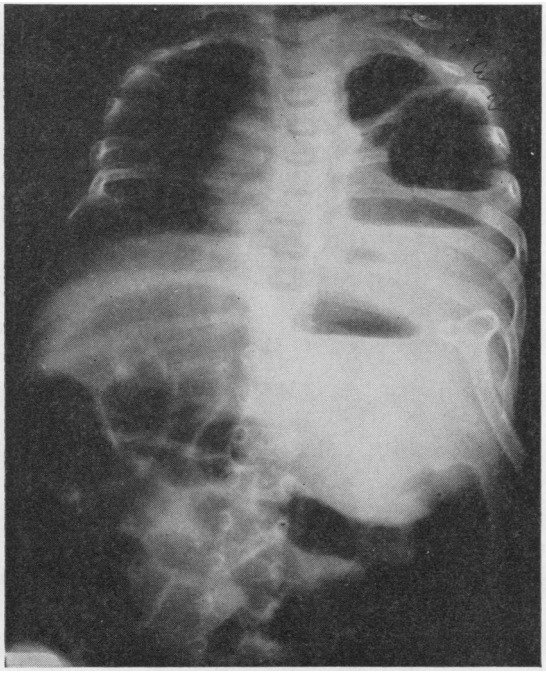


FIG. 13.—Severe distension of the intrathoracic colon. The left lung is totally collapsed.

When a colon graft is used to restore continuity, difficulty may arise from a closed loop obstructing the graft. This is due to three factors: first, to stomal oedema at the oesophago-colic and gastro-colic anastomoses; secondly, to pressure on the graft at the thoracic inlet and oesophageal hiatus; and thirdly, because the graft is largely denervated. In one baby the graft was widely distended, eventually perforating (Fig. 13); the heart stopped, and, although the heart beat was restored by massage, intrapleural tension was relieved, and the graft sutured and drained transpleurally, cerebral damage resulted in the baby's death.

In future cases it is now advised that oesophago-colic anastomosis be delayed, the upper end of the graft being drained on the neck until all danger of obstruction is past.

DISCUSSION OF RESULTS

Humphreys, Hogg, and Ferrer (1956) listed the factors they considered to influence survival as follows: (1) The size and degree of prematurity of the infants; (2) the association with other severe malformations; (3) the awareness of doctors and nurses caring for newborn infants of the possibility of the condition and their alertness in recognizing it; (4) the promptness with which babies are referred for surgery; (5) the care in pre- and post-operative management; (6) the technical aspects of the operation itself.

They considered that, although the first two of these are uncontrollable, the others leave room for improvement. Shaw (1956), in discussing their paper, pointed out that delay in surgical treatment need not necessarily result in death, and then quoted the case of a baby aged 28 days who had been maintained on parenteral fluids and then survived a primary oesophageal anastomosis.

Since it is reasonable to suppose that the presumed high mortality for infants of low birth weight is from lung complications due to a feeble cough reflex, the deaths occurring within two weeks of operation (nine) have been plotted with the survivors for corresponding birth weights in the form of stick graphs (Fig. 14). There does not appear to be any great difference in two weeks'

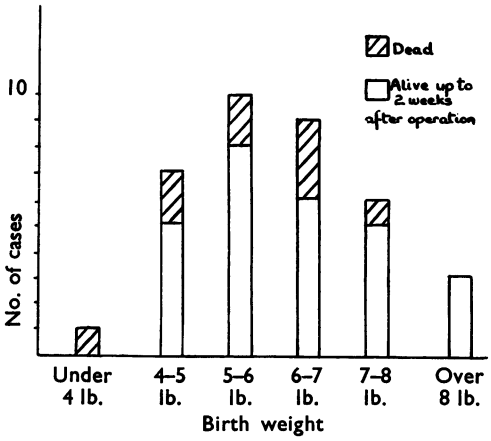


FIG. 14.—Histogram of the mortality and survival rate within the first two post-operative weeks at various birth weights.

survival between any groups. These nine deaths are then plotted with the survivors for two weeks against the age of the baby on admission, since many presume that delay in admission for surgery increases the mortality due to established aspiration pneumonitis (Fig. 15). Again there seems to be no indication that moderate delay is unfavourable, the longest period before admission being five days. The nine early post-operative deaths have been analysed in Table VIII.

Six of the early deaths are attributable to spill-over of oral secretions into the bronchial tree. In one a large persistent ductus arteriosus may have been a contributory factor. One death caused by a tension pneumothorax was due to a technical error and should not have occurred. The cause

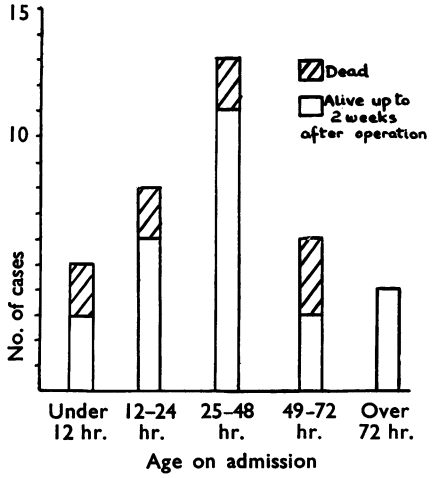


FIG. 15.—The mortality and survival rate within the first two post-operative weeks at various ages of admission to hospital.

TABLE VIII

CAUSE OF DEATH WITHIN TWO WEEKS OF OPERATION

Case No.	Cause of Death
16	Atelectasis: ? persistent ductus arteriosus
17	Atelectasis
19	,,
20	Tension pneumothorax: anoxia of the cardio-respiratory centre
24	Bronchopneumonia
25	Atelectasis
28	? Anoxia of cardio-respiratory centre
34	Probable leakage from thoracic duct
35	Bronchopneumonia

of death was confirmed by post-mortem examination in all save one patient. In this baby good respiratory movements were not resumed after operation; rib recession had been present before operation, and death clinically appeared to be due to a gradual cardio-respiratory failure from anoxia, although intolerance to the anaesthetic agents cannot be excluded.

Out of 18 deaths in hospital, permission for post-mortem examination was obtained in 16. The cause of death in these patients is summarized in Table IX. One other child died at the age of 3½ years from multiple injuries following a road accident, but has been counted as a survival of operation; no post-mortem findings are available for this child.

TABLE IX

CAUSE OF DEATH IN 18 PATIENTS AFTER OPERATION

Case No.	Operation	Necropsy	Cause of Death
16	Primary anastomosis	Yes	Atelectasis; ? persistent ductus arteriosus contributing
17	,,	,,	Atelectasis
18	,,	,,	Unknown
19	,,	,,	Atelectasis; small tentorial tear with intracranial haemorrhage
20	,,	,,	Tension pneumothorax; anoxia of cardio-respiratory centre
21	,,	,,	Empyema; lung abscess
22	,,	,,	Atelectasis
23	,,	,,	Pericarditis; mediastinal abscess; fibro-purulent pleurisy
24	,,	,,	Bilateral confluent bronchopneumonia
25	,,	,,	Atelectasis
28	First stage	No	Anoxia of cardio-respiratory centre
29	,, ,,	Yes	Mediastinal abscess
30	,, ,,	,,	Ventricular septal defect
31	Second stage	,,	Peripheral circulatory failure due to inadequate blood replacement
32	,, ,,	,,	Rupture of colic graft; anoxia of cardio-respiratory centre
33	,, ,,	No	Post-operative respiratory obstruction; ? inhaled mucus. Anoxia of cardio-respiratory centre
34	First stage	Yes	Peripheral circulatory failure due to thoracic duct leakage
35	Primary anastomosis	,,	Bronchopneumonia; unsuspected fistula between upper oesophagus and trachea

Thus, of 18 deaths, seven were due to aspiration of mucus and oral secretions into the bronchial tree (atelectasis, bronchopneumonia), a contributory factor in one case being intracranial haemorrhage from a tentorial tear. Four infants died owing to cerebral damage, anoxia of the cardio-respiratory centre being due to cardiac arrest in two as a result of tension effects (tension pneumothorax in one; rupture of a colon graft in one); one patient failed to breathe properly following operation, and in one the heart stopped a short time after operation.

Sepsis was responsible for death in three babies; one constituted an error of management as a gastrostomy tube had perforated the fundus of the stomach, passed across the diaphragm and into the left lower lobe, causing a lung abscess. Peripheral circulatory failure was due to inadequate blood transfusion in one baby. In another recurrent bilateral pleural effusions were probably due to a thoracic duct injury, but the characteristic chylous fluid was not obtained on aspiration as the baby was being maintained on parenteral fluids, and no fat had been given into the alimentary tract.

One baby's death was unexplained; in view of the possibility of mediastinal leakage from the oesophageal anastomosis, gastrostomy feeds were being given, and death occurred suddenly one hour after a feed; at post-mortem examination no cause of death was found, and the oesophageal anastomosis had healed.

Conclusions

Whenever possible, primary oesophageal anastomosis should be done for oesophageal atresia, and in those cases most suitable for anastomosis (Group C(ii)) a high survival rate may be expected. Where tension is too great to allow a safe anastomosis, a staged procedure should be done. Further improvement in the prevention and treatment of atelectasis in the infants is required to decrease the immediate post-operative mortality. Early recurrence of a tracheo-oesophageal fistula, or early stricture formation, are best treated conservatively. When the prevention of aspiration of mucus into the bronchial tree demands further surgery the cervical oesophagus should be exteriorized, the lower oesophagus closed, and feeding gastrostomy should be done.

Reconstruction by oesophago-gastrostomy gives a good chance of survival to two babies out of three in the series, the one death being due to inadequate blood replacement after operation. When colon oesophagoplasty is done, special care is necessary in post-operative management. The three colon grafts in the series have been unsuccessful. One failed owing to death of the graft at the time of operation because there was traction on the pedicle, and this was the baby that failed to survive oesophago-gastric anastomosis later. One baby died shortly after operation, probably from respiratory obstruction due to inhaled mucus, and in the other a "closed-loop" obstruction of the graft was recognized and treated too late. In future cases it will be advisable to delay oesophago-colic anastomosis until all danger of obstruction of the graft is past.

It is with great pleasure that I record my indebtedness to Mr. A. L. d'Abreu, by whose permission these patients were treated, and whose encouragement was a constant stimulus. I am extremely grateful to the physicians of the Children's Hospital, Birmingham, for referring these babies to me, and especially to Professor J. M. Smellie, to whose ward most of them were admitted. The radiological investigations conducted by Dr. Roy Astley have been an essential feature of the work. A debt of gratitude which cannot be placed too highly is owed to successive resident medical officers and the nursing staff at the Children's Hospital, Birmingham, for their devoted care of these infants. Mr. J. G. Williamson has been responsible for the excellence of the illustrations. To Miss Margaret Wilkie I am especially grateful for typing the manuscript.

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