THE SURGICAL MANAGEMENT OF CONGENITAL ATRESIA OF THE ESOPHAGUS AND TRACHEO-ESOPHAGEAL FISTULA*

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The earliest recorded instance of esophageal deformity appears to be that related by Durston[11] in 1670, a case of simple atresia of the esophagus. Thomas Gibson[14] in 1696 first observed a case of congenital atresia of the esophagus with a tracheo-esophageal fistula, noting the typical feeding difficulties and accurately describing the postmortem findings. Many authors[5, 15, 20, 26, 40, 45, 49, 50, 57] have previously reviewed the literature and collected recorded cases. The increasing number of reports of cases of congenital atresia of the esophagus makes it evident that this anomaly is not rare and has been frequently overlooked in the past.

The occasional occurrence of this deformity in members of the same family might indicate that genetic factors are responsible as the cause. Congenital atresia of the esophagus with tracheo-esophageal fistula has been reported in both a brother and sister in Lanman's[35] series and in two brothers reported by Grieve and McDermott.[16] Mackenzie[45] cited a case of a father, all of whose children by three wives showed this anomaly.

In this series there were 63 male and 40 female infants. No one embryologic explanation of esophageal anomalies will account for all types seen.

The first anlage of the respiratory system appears in the 3-mm. embryo, caudal to the pharyngeal pouches, as a ventral rounded mass of entoderm. Almost complete separation of the trachea from the esophagus may be seen in the 8-mm. embryo. Gruenwald[17] found a typical atresia of the esophagus and tracheo-esophageal fistula in a 9-mm. human embryo. His explanation of the malformation assumed a more or less complete failure of the laryngotracheal tube to separate in proper time from the esophagus. This explains the fistula, and the atresia is a sequel of the fast growth of the trachea drawing out the corresponding part of the esophagus into a narrow strip contributing to its own dorsal wall (Fig. 1).

In the literature various classifications[32, 74, 75] of congenital anomalies of the esophagus have been recorded. The classification suggested by Vogt[75] in 1929 is comparatively simple and frequently used (Fig. 2.) Accordingly, these anomalies are divided into three main groups. In Type 1 there is a complete absence of the esophagus; an extremely rare condition. In Type 2 there is a blind end to both the upper and the lower segments of the esophagus, neither of which communicates with the esophagus; a rare type. Type 3 includes those anomalies with tracheo-esophageal communications. In Type 3a the upper

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segment communicates with the trachea and the lower segment has a blind proximal end; a rare type. In Type 3b the upper segment is blind and the lower segment communicates with the trachea at or about the level of the bifurcation of the trachea. This is the common type of congenital anomaly of the esophagus. In Type 3c both the upper and lower segments communicate with the trachea; a rare type.

Since the first successful case in 1939, 103 cases of congenital atresia of the esophagus and tracheo-esophageal fistula have been seen at the University of Minnesota Hospitals and the Children's Hospital in St. Paul. There were seven cases of simple atresia of the esophagus (Type 2). The obstruction in cases of simple atresia may vary from a membranous diaphragm to complete agenesis.[2, 73, 74] The lower segment of esophagus in the seven cases in this series ended blindly just above the diaphragm and the upper segment appeared as the usual hypertrophic blind pouch. The common type of atresia of the esophagus with tracheo-esophageal fistula (Type 3b) occurred in 93 cases. In two cases the upper and lower segments of the esophagus communicated with the trachea (Type 3c).

One case included in the series was unusual. There was no atresia of the esophagus and two tracheo-esophageal fistulas were found at postmortem examination. Each fistula was 2 mm. in diameter. The lower one was located 1.75 cm. above the carina. The other fistula was 0.5 cm. directly above the lower fistula and 2 cm. below the vocal cords. The trachea and esophagus were intimately attached over an area of 1.5 cm.

Haight[21] reported the first case of tracheo-esophageal fistula without atresia, diagnosed during life, and successfully corrected by surgery. Abbott[1] saw three cases of so-called H-type of fistula in two years.

Associated multiple anomalies are common and complicate the management of these cases, often contributing to an unsuccessful result. In this series there were 17 cases having other major anomalies, 12 of which might have been considered a factor in an unfavorable result. Intracranial hemorrhage was present in three cases.

In the most common type of atresia of the esophagus the upper segment terminates blindly at a variable distance above the bifurcation of the trachea, while the lower segment has a fistulous communication with the trachea. The site of the fistula is usually within 1 cm. of the carina but may occur over 2.6 cm. (Ladd[32]). The upper blind pouch is usually dilated and its walls hypertrophied. This dilatation and hypertrophy are evidently the result of the ineffectual attempts of the fetus to swallow the amniotic fluid. The lower segment of the esophagus, at the cardiac end, is usually of normal size but diminishes in diameter (5–7 mm.) toward its tracheal opening, and may be so small (3–4 mm.) that construction of a primary anastomosis of the esophagus is impossible (Haight[20]—Fig. 3).

The symptoms associated with this anomaly should be readily recognized at birth. An increased amount of mucus may be noted filling the mouth and pharynx and drooling from the side of the mouth. Spells of cyanosis occur due to aspiration of mucus. Severe choking spells occur after a few swallows when fed. Mucus and feedings are regurgitated through the mouth and nose. These infants rapidly lose weight due to starvation, develop aspiration pneumonia, and die if not given the chance of survival now offered by surgery.

The diagnosis may be suggested by the history and confirmed by the passage of a small soft rubber urethral catheter, which meets obstruction usually 10 to 12 cm. from the anterior alveolar margins. The upper blind pouch may be visualized by instilling a small amount of Lipiodol under the fluo-

roscope. The Lipiodol should be removed from the blind pouch to prevent aspiration into the trachea. The presence of a tracheal fistula from the upper segment can thus be determined. The same information may be obtained, less simply, by esophagoscopic examination.[62, 72] The presence of air in the stomach in cases of atresia of the esophagus indicates a fistulous communication of the lower segment with the trachea or bronchus. The absence of air in the stomach indicates but does not prove the absence of a fistula. Haight,[20] Humphreys,[26] and Shaw[63] reported cases in which no air had entered the stomach through a very small fistulous communication between the trachea and lower esophageal segment. Selander[60] and Fuhrman[12] *et al.* have shown that the presence of air in both segments of the esophagus will give a satisfactory outline of the anomaly in the roentgen ray picture.

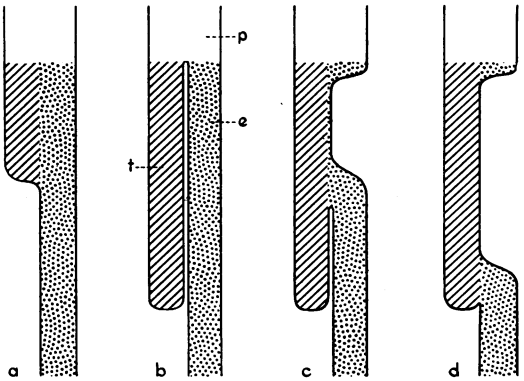

FIG. 1.—Development of the esophagus (after Gruenwald).

The common type of atresia of the esophagus presents three problems: (1) feeding, (2) prevention of regurgitation of gastric contents into the lungs through the tracheo-esophageal fistula, and (3) prevention of aspiration of saliva or liquids. Surgical treatment of these cases has been directed to one or more of these problems (Fig. 4).

Gastrostomy alone[10, 53, 59, 66, 73] for feeding was the most frequent procedure used prior to 1939. Gastrostomy feeding may hasten death by increasing the regurgitation of gastric contents through a patent tracheo-esophageal fistula. To lessen this danger, passage of the gastrostomy catheter through the pylorus and well down into the jejunum had been tried without success.[36] The use of jejunostomy[10, 31, 52, 68] had likewise been ineffectual, since gastric contents may still be aspirated into the lungs.

Various operations at the cardiac end of the stomach have been performed.[4, 8, 36, 65, 72, 73] Ligation of the cardiac end of the esophagus plus gastrostomy has been employed successfully.[47] By this procedure, a blind pouch of esophagus is formed in which secretions will collect, become infected, and empty into the lungs. There is also a definite danger of the occluding ligature cutting through the wall of the esophagus, permitting leakage or recanalization of the lumen.[8] Transection[36, 58, 73] of the upper end of the stomach, abdominal esophagostomy, and distal gastrostomy have been unsuccessful. Exteriorization of the cardiac end of the esophagus and stomach plus gastrostomy has resulted in ulceration of the exteriorized segment.[36] In both of these procedures considerable difficulty presented in the management of the wound due to drainage of tracheal and gastric secretions.

Several methods of direct attack on the fistula have been attempted.

Endotracheal stenosis of the fistula by chemicals has been suggested.[25] In Case 1 of this series the fistulous communication of the esophagus to the trachea was identified through a bronchoscope and was coagulated by an electrode passed into the fistula.[37] This did not effectively stenose the fistula. Injection of caustic solution through a gastrostomy tube passed up into the lower esophagus has been tried.[6]

Division and closure of the fistula at the trachea, together with connection of the esophageal segments by a tube[39] or anastomosis of the segments by a modified Murphy button,[18] have been suggested. Richter[54] in two cases performed a gastrostomy for feeding after ligating the fistulous tract at the trachea through a transpleural approach.

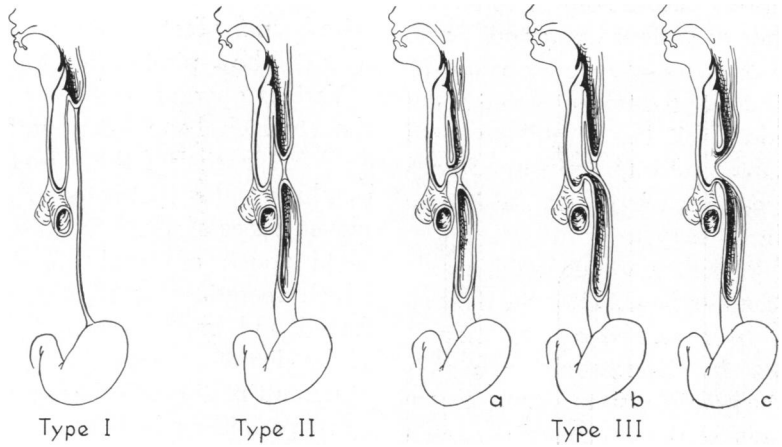

FIG. 2.—Classification of congenital anomalies of the esophagus (after Vogt).

Mixter[46] in 1929 ligated and divided the fistulous communication to the trachea through a right-sided extrapleural approach, bringing out the distal esophagus as a dorsal esophagostomy and placing a catheter in this for feeding. In some cases, proximal esophagostomy was added primarily or secondarily to prevent the overflow of secretions from the blind proximal end of the esophagus. Lanman[35] reported a series of cases in which this procedure was followed, and concluded that dorsal esophagostomy should be abandoned in favor of simple ligation of the fistula in those cases in which a primary anastomosis cannot be carried out.

In 1939 the author successfully carried out a more direct attack on the fistula. This patient was the first case with congenital atresia of the esophagus and tracheo-esophageal fistula that has survived. It is of extreme interest that Ladd[32] came to the same conclusions about methods of attacking this problem about the same time, his first living patient being but a day younger than the author's.

The procedure used in the first successful case will be briefly outlined. After preliminary gastrostomy, the esophageal fistula was ligated at the trachea through a right-sided extrapleural approach, resecting the entire fourth rib. The proximal blind pouch of esophagus was later exteriorized to make a cervical esophagostomy. This plan necessitates construction of an antethoracic esophagus to re-establish the continuity of the gastro-intestinal tract.

The sequence of operations is important. The success in the first case in this series led to incorrect conclusions. Gastrostomy was the initial operation, and ligation of the fistulous tract and exteriorization of the upper blind pouch were considered elective procedures. After losing several patients in whom only gastrostomy had been done, it became evident that early treatment of the upper blind pouch as well as ligation of the fistulous tract are of first importance. In Cases 14 and 28 (Table I) in this series a cervical esophagostomy was done prior to ligation of the fistula because both patients were critically ill with aspiration pneumonia and would not have survived an

intrathoracic exploration at that time. The patient in Case 14 was cyanotic and required continuous oxygen therapy en route to the operating room. Both patients are now alive and well.

Re-establishment of the continuity of the alimentary tract is a major problem in the cases in which a multiple-stage operation has been performed. That no procedure has been entirely satisfactory is indicated by the number of various operations used. Construction of an esophagus by means of a skin-lined tube has been successfully done by Ladd[32] and Ivy *et al.*[29] In these cases Davis and Stafford,[9] Stevenson,[67] and Hanrahan[23] have made a skin-lined tube esophagus for other types of cases.

Use of a jejunal tube for total reconstruction of the esophagus after the method of Roux has been carried out by Yudin[77] and modified by Longmire.[42] Another method is that of Wullstein, using a skin tube esophagus for the upper portion and a jejunal tube to complete the lower portion of the new esophagus. Ladd and Swenson,[33] Lam,[34] and Ochsner and Owens[48] have successfully constructed this type of esophagus to complete these cases. In many cases, difficulties with fistulas or strictures at the jejunocutaneous or esophagocutaneous junction have been encountered. Longmire and Ravitch[41] used a free jejunal graft implanted in a skin tube for construction of an antethoracic esophagus. Longmire[43] has successfully used an antethoracic jejunal transplantation in two infants. Harrison[24] used a transthoracic jejunal loop substitution in high stricture of the esophagus. Robertson and Sarjeant[56] placed the jejunal transplant in the anterior mediastinum.

Sweet[69] described a method of intracervical esophagogastric anastomosis after pulling the stomach up through the chest. This one-stage operation may prove to be the most satisfactory solution to the problem, if the intrathoracic position of the stomach does not cause difficulty during feedings. Cliffton performed a two-stage cervical esophagogastrostomy utilizing a 6-cm. segment of the distal esophagus in the anastomosis. Reinhoff,[55] Bigger,[3] and Potts[51] have carried out an antethoracic esophagogastrostomy in cases where primary anastomosis of the esophagus could not be done. Potts subsequently placed the stomach in an intrathoracic position by dividing the costal cartilages. Swenson[70] and Haight[22] have each recorded successful primary intrathoracic esophagogastrostomy in infants. In one case in this series an esophagogastrostomy was done as a primary procedure. Death occurred on the third postoperative day. Large interauricular and interventricular defects were found in the heart at postmortem. The mortality of this operation as a primary procedure in the new-born is too great to recommend it. The multiple-stage operation with later reconstruction by esophagogastrostomy or esophagojejunostomy as an elective procedure is preferable in the cases of esophageal atresia in which a primary anastomosis of the esophagus is not possible.

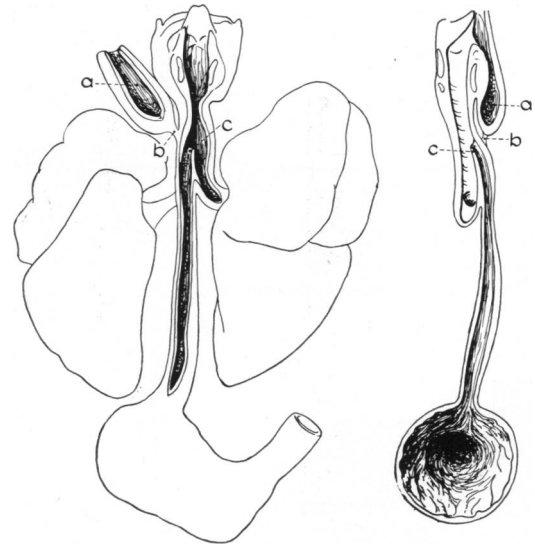

FIG. 3.—Common type of atresia of the esophagus with tracheo-esophageal fistula. Type 3b.

In nine cases in this series antethoracic esophagojejunostomy has been successfully performed. In one case the reconstruction was by a cervical esophagogastrostomy. These cases will be reported in another paper. It was learned early in these reconstructions that by-passing the stomach by esophagojejunostomy created a situation unfavorable to good nutrition. Accordingly, a jejunogastrostomy has been performed in all of these cases, with marked improvement in nutrition, after utilizing the stomach for its normal function.

Because of the difficulties of reconstruction of the esophagus, the multiple-stage operation for congenital atresia of the esophagus and tracheo-esophageal fistula should be used only when primary anastomosis is impossible, because of the distance between the esophageal segments.

Primary anastomosis of the upper blind pouch to the lower segment of the esophagus after ligation and division of the fistula to the trachea is the ideal solution to the threefold problem presented by these cases. It closes the tracheo-esophageal fistula, takes care of salivary secretions, provides for feeding, and restores the esophagus more nearly to normal than any other plan of operation. However, the discrepancy in size of the two esophageal segments and the distance between the segments make this operation often difficult and sometimes impossible.

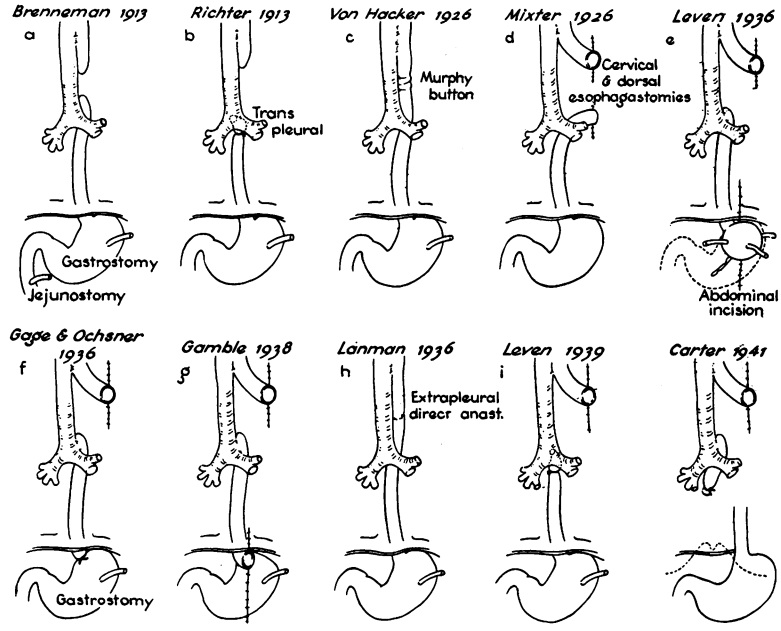

FIG. 4.—Various operations performed in cases of congenital atresia of the esophagus with tracheo-esophageal fistula (after Carter).

In 1936 and 1937 Lanman[35] attempted a primary anastomosis of the esophagus in four cases. Shaw[61] in 1939 first reported the use of this operation. In the period 1939 to 1950, Haight[19, 22] operated on 86 of 93 patients seen at the University Hospital at Ann Arbor. There were 41 operative recoveries, the oldest being the first successful case of primary anastomosis of the esophagus recorded in this anomaly.

The work of Ladd and Swenson,[33] Swenson,[71] Bigger and Potts[51] has been pre-eminent in the management of these cases. Gastrostomy done one or two days preliminary to thoracotomy to deflate the stomach and intestines is advocated by Bigger.[3] In

this series a gastrostomy has been the first procedure in those cases in which there was no air in the stomach or intestines. In such cases the distal esophagus may then be explored by a catheter or bougie to determine if it is adequate for a primary anastomosis with the upper segment. In this series the distal segment of the esophagus was atretic 1.0 to 4.0 cm. above the stomach. A multiple-stage procedure is recommended as indicated previously.

While there are advocates of the transpleural approach in primary anastomoses of the esophagus,[30, 64] it is the consensus of most surgeons that the extrapleural approach is safer in these infants.

Swenson[71] reported 113 cases operated with 53 surviving. In his series of 32 primary anastomoses there were five postoperative deaths. This mortality is a marked improvement over the results from any other series.

PREOPERATIVE CARE

Since most of these infants often come to the surgeon from considerable distances and after a few days delay, careful preoperative preparation is imperative to improve the possibility of a successful result.

The infant is placed in oxygen at once and the upper esophageal pouch aspirated intermittently or continuously by a soft rubber catheter. The prone position with the head dependent aids drainage of salivary secretions.

In all of these cases a polyethylene tube is placed in the saphenous vein at the ankle prior to surgery for the administration of fluids and blood. The total amount of fluids, particularly saline, is strictly limited to avoid pulmonary and generalized edema that so often occurs in these children when the amount of fluids calculated for an average infant has been given. About 75 cc. of fluid per kilogram of body weight is given in 24 hours in these cases. About one-third of this is given as normal saline or Darrow's solution and the remainder as 5 per cent glucose in distilled water. Transfusions of whole blood are given if the hemoglobin is less than 15 Gm. whereas plasma may be given if the hemoglobin is over 15 Gm. The amount of blood or plasma given is substituted for the saline required.

Penicillin, streptomycin, and vitamin K are given in adequate amounts.

OPERATION

In this series of cases the anesthetic of choice has been cyclopropane. In the first portion of this series the anesthesia was given by a closely fitting mask. However, in the latter portion of the series the anesthesia has been by endotracheal method, using a Portex, size 2–0 endotracheal tube. In ten of these cases anesthesia was with Pentothal and curare for introduction of the endotracheal tube. In these cases the patient was maintained on nitrous oxide and Pentothal curare during the remainder of the procedure.

The primary anastomosis of the esophagus is performed through an extrapleural approach on the right side. Through an oblique incision the entire fourth rib on the right side is removed, disarticulating it at the spine. The periosteal bed of the rib is incised and the parietal pleura stripped widely from the chest wall. In some instances where the upper blind pouch is extremely high in its position the third rib is resected instead of the fourth rib. This approach gives adequate exposure and does not injure the thoracic cage. The azygos vein is divided between ligatures central to the entrance of the highest intercostal vein. In some cases it is unnecessary to divide the azygos vein to expose the esophageal segments. The lower segment of the esophagus is then identified just below its communication with the trachea. The hypertrophic upper blind pouch is usually easily identified. However, when it is in an extremely high position the passage of a soft

rubber catheter by the anesthetist into the mouth and pharynx will push the lower end of the pouch into view and aid in its identification. The upper segment is mobilized high into the neck and a traction suture placed in the blind end. Traction on this suture will pull the segment down, and most of the length of esophagus necessary for primary anastomosis is gained by mobilization of this upper pouch. The lower segment of the esophagus is mobilized to the point of its fistulous opening into the trachea and divided. The opening is closed with interrupted sutures. The closure of the fistula is then reinforced by oversewing it with mediastinal fat and areolar tissue to protect against leakage. In some instances the upper portion of the lower esophageal segment is so small that it must be resected before an adequate anastomosis can be carried out. The anastomosis is made by using a single row of interrupted 5–0 silk sutures. It is important that a good mucosal apposition be obtained in the anastomosis. A second row of sutures in the muscularis is used to reinforce the initial row. The use of the fine-toothed ductus clamps, as suggested by Potts, has facilitated making the anastomosis without tension, while placing the sutures, in those cases where there is considerable distance between the two esophageal segments. A soft rubber catheter drain is placed in the extrapleural space and brought out in the first intercostal space below the incision. The chest wall is closed with interrupted silk. The lung is expanded under positive pressure before closure of the chest is completed. Constant suction is applied to the catheter at 10 cm. water negative pressure for a period of eight to ten days.

POSTOPERATIVE CARE

The patient is placed in oxygen after the operation. Fluids, antibiotics, and vitamins are given as in the preoperative period. Salivary secretions are carefully aspirated, using a catheter that is marked so that it will not be pushed down as far as the anastomosis.

As a rule a gastrostomy is performed after 24 hours and feeding started 12 hours later. Because of difficulty with pylorospasm due to the presence of the gastrostomy tube in the stomach, a catheter is passed through the pylorus into the jejunum. Oral feedings are started on the tenth day after checking the continuity and patency of the esophagus with a small amount of Lipiodol instilled into the upper esophagus under the fluoroscope. If there is a leak at the anastomosis, oral feedings are withheld until this closes.

External esophageal fistulas from leakage at the site of anastomosis usually heal without difficulty. However, re-establishment of a tracheo-esophageal fistula is a serious complication.

The present series includes 103 cases of esophageal atresia seen at the University Hospital since 1939, when the multiple-stage operation for correction of this anomaly was first used (Table I). In seven cases (15, 29, 39, 45, 74, 82, and 101) no operation was performed, because four of the patients died before they could be prepared for surgery, in two cases permission for surgery was refused by the parents, and in one case (82) the presence of two tracheo-esophageal fistulas without esophageal atresia was not demonstrated before postmortem. Gastrostomy was the only procedure in five cases following the sequence of operations carried out in the first successful case. These patients died before further stage operations were carried out. In three cases (3, 4, and 5) pneumonia or multiple lung abscesses due to aspiration were found at postmortem examination. In Case 2, death resulted from peritonitis due to perforation of the jejunum. Case 21 was a premature infant, weighing only 1050 Gm.

The death in Case 7 was most discouraging. Gastrostomy, ligation of the fistula,

TABLE I.—*Analysis of 103 Cases of Congenital Atresia of Esophagus.*

Case No.	Hosp. No.	Sex	Type of Anomaly	Operation	Result	Remarks
1	R.L. 688745	M	3 b	Gastrostomy Fulguration of fistula Ligation of fistula Cervical esophagostomy Antethoracic esophagojejunostomy in 3 stages	Living	Excellent functional result
2	S. 695097	F	2	Gastrostomy	Died at 6 days	Peritonitis Perforated jejunum Lower segment ended 2 cm. above cardia
3	W.K. 711782	M	3 b	Gastrostomy	Died at 15 days	Aspiration pneumonia
4	M.J. 711856	M	3 b	Gastrostomy	Died at 9 days	Multiple lung abscesses
5	J.H. 714590	M	3 b	Gastrostomy	Died at 5 days	Bronchopneumonia Mongolism Cerebral sclerosis
6	M.J. 715849	F	3 b	Ligation of fistula	Died at 5 days	Congenital atresia of duodenum and annular pancreas. Spontaneous perforation of stomach
7	D.B. 721398	M	3 b	Gastrostomy elsewhere at 2 days of age Ligation of fistula Cervical esophagostomy	Died at 2-3/12 years	Perforation of stomach by gastrostomy tube
8	R.W. 722543	F	3 b	Ligation of fistula	Died at 10 days	Over-hydration Lung edema
9	D.E. 730647	M	3 b	Gastrostomy and ligation of fistula Secondary ligation of fistula Cervical esophagostomy Antethoracic esophagojejunostomy in 3 stages	Living	Coughed up ligature 1 month after first ligation Excellent functional result
10	H. 732691	F	3 b	Gastrostomy and ligation of fistula	Died at 23 days	
11	D.G. 733867	F	3 b	Gastrostomy and ligation of fistula Cervical esophagostomy Antethoracic esophagojejunostomy in 3 stages	Living	Excellent functional result
12	J.R. 736655	F	3 b	Gastrostomy and ligation of fistula Cervical esophagostomy	Died at 15 days	Respiratory failure due to aspiration of mucus
13	M.M. 739083	M	3 b	Gastrostomy Cervical esophagostomy Ligation of fistula	Died at 14 days	Perforation of stomach by gastrostomy tube
14	D.W. 739320	M	3 b	Gastrostomy elsewhere Cervical esophagostomy Ligation of fistula Secondary ligation of fistula Antethoracic esophagojejunostomy in 3 stages	Living	Re-established fistula 42 days after first ligation Excellent functional result
15	D.H. 739970	M	3 b	None	Died at 4 days	Aspiration pneumonia Critically ill on admission
16	J.G. 740763	M	3 b	Primary anastomosis of esophagus with ligation of fistula	Died at 7 days	Over-hydration
17	R.W. 741227	M	3 b	Primary anastomosis of esophagus with ligation of fistula Gastrostomy	Died at 37 days	Developed esophageal fistula, diarrhea Hypoproteinemia Generalized anasarca
18	J.B. 741913	F	2	Gastrostomy Cervical esophagostomy Antethoracic esophagojejunostomy in 3 stages	Living	Excellent functional result
19	S.J. 744050	F	3 b	Primary anastomosis of esophagus with ligation of fistula Colostomy	Died at 88 days	Re-established fistula Aspiration pneumonia Imperforate anus with rectovaginal fistula Bilateral hydro-ureter and hydronephrosis Subcutaneous abscesses

TABLE I.—(*Continued*).

Case No.	Hosp. No.	Sex	Type of Anomaly	Operation	Result	Remarks
20	R.B. 750804	M	3 b	Primary anastomosis of esophagus with ligation of fistula Gastrostomy Secondary mediastinal exploration	Died at 95 days	Re-established tracheo-esophageal fistula which healed spontaneously Hypertrophic pyloric stenosis Horseshoe kidney
21	G. 752869	M	2	Gastrostomy	Died at 9 days	Premature Weight 1050 Gm.
22	P.N. 753769	M	3 b	Ligation of fistula Gastrostomy and cervical esophagastomy Esophagogastrostomy	Living	No dysphagia Nutrition poor
23	M.H. 756897	M	3 b	Primary anastomosis of esophagus with ligation of fistula Gastrostomy Dilatations of stricture of esophagus	Living	Age 7 years
24	R.S. 756941	M	3 b	Ligation of fistula Gastrostomy and cervical esophagostomy	Died at 30 days	Perforation of stomach by gastrostomy tube Hypertrophic pyloric stenosis
25	W.L. 757232	M (colored)	3 b	Primary anastomosis of esophagus with ligation of fistula Gastrostomy	Living	Developed small esophagopleural leak which closed spontaneously Has stricture of esophagus
26	P.K. 760455	M	3 b	Primary anastomosis of esophagus with ligation of fistula	Died at 3 days	Hemorrhagic bronchopneumonia Pyopneumothorax, right Polycystic kidney, left
27	J.B. 760618	M	3 b	Primary anastomosis attempted. Tension too great. Fistula ligated and cervical esophagostomy Gastrostomy Antethoracic esophagojejunostomy in 3 stages	Living	Excellent functional result
28	O.B. 760878	M	3 b	Cervical esophagostomy Ligation of fistula Gastrostomy Antethoracic esophagojejunostomy in 3 stages	Living	Critically ill on admission with bilateral bronchopneumonia Upper blind segment very short Excellent functional result
29	M. 761813	F	3 b	Surgery refused	Died at 14 days	Aspiration pneumonia Old intracranial hemorrhage
30	G.F. 763630	M	3 b	Primary anastomosis of esophagus with ligation of fistula Gastrostomy	Living	Some trouble with pylorospasm
31	D.B. 768565	M	3 b	Primary anastomosis Gastrostomy Thoracotomy	Living	Anastomosis leak Stricture dilated
32	P.R. 768737	M	3 b	Primary anastomosis	Living	Stricture dilated
33	S.C. 769006	F	3 b	Primary anastomosis	Died at 4½ months Acute respiratory infection	Multiple anomalies Anastomosis excellent No dysphagia
34	D.H. 769553	F	3 b	Primary anastomosis	Living	
35	H. 773065	M	3 b	Primary anastomosis	Died at 7 days	Aspiration pneumonia due to obstruction by catheter in esophagus
36	M.S. 775644	M	2	Exploration of chest Cervical esophagostomy Gastrostomy Antethoracic esophagojejunostomy in 3 stages	Living	Excellent functional result
37	B.M. 775773	F	3 b	Primary anastomosis	Living	Stricture dilated
38	N.M. 776179	F	3 b	Primary anastomosis	Living	Stricture dilated
39	B.L. 777090	F	3 b	No operation	Died at 3 days	Operation refused
40	G.D. 777407	M	3 b	Primary anastomosis Gastrostomy Secondary anastomosis at 7 months	Died at 7 months	Anastomosis leak Impermeable stricture Died of anoxia at completion of secondary anastomosis

TABLE I.—(*Continued*).

Case No.	Hosp. No.	Sex	Type of Anomaly	Operation	Result	Remarks
41	D.S. 777429	M	3 c	Ligation of fistula Tracheotomy Cervical esophagostomy Gastrostomy Antethoracic esophagojejunostomy in 4 stages	Living	Partial loss of jejunal tube. Sufficient jejunum viable to permit completion of anastomosis at subsequent operation Excellent functional result
42	T.B. 777699	M	3 b	Primary anastomosis Gastrostomy	Died at 8½ months	Bronchopneumonia due to a reestablished tracheo-esophageal fistula
43	W.K. 777794	M	3 b	Primary anastomosis	Living	Uneventful
44	J.L. 779988	M	3 b	Primary anastomosis	Living	Uneventful
45	J.A. 780063	M	3 b	No operation	Died at 8 days	Multiple anomalies Bilateral cleft lip and palate Congenital heart disease
46	K.S. 780396	M	3 b	Primary anastomosis Encephalocele repaired at 10 months	Living	Stricture dilated
47	J.S. 781004	F	3 b	Primary anastomosis	Died at 6 days	Bronchopneumonia Anastomosis leak
48	D. 780267	M	3 b	Primary anastomosis Gastrostomy	Living	Anastomosis leak Stricture dilated
49	J.G. 781648	M	3 b	Primary anastomosis Gastrostomy	Living	Anastomosis leak Stricture dilated
50	U. 782470	F	3 b	Primary anastomosis Gastrostomy	Living	Anastomosis leak Stricture dilated
51	J.B. 782542	F	3 b	Ligation of fistula	Died at 6 days	Distance between segments too great for anastomosis
52	E.K. 784870	F	3 b	Primary anastomosis Gastrostomy	Died at 7 days	Anastomosis leak Empyema
53	B.S. 786087	M	3 b	Primary anastomosis	Died at 8 days	Bronchopneumonia Horseshoe kidney Bilateral hydroureter
54	L.B. 786669	F	3 b	Primary anastomosis Gastrostomy	Living	Uneventful
55	P.G. 786875	M	3 b	Primary anastomosis Gastrostomy	Died at 25 days	Cleft lip and palate Premature Weight 1590 Gm. Bronchopneumonia Anastomosis leak
56	J.T. 789239	M	3 b	Primary anastomosis Gastrostomy	Living	Uneventful
57	B.H. 793220	M	3 b	Primary anastomosis	Died at 9 days	Anastomosis leak Extrapleural abscess
58	M.A. 793897	F	2	Gastrostomy Cervical esophagostomy	Died at 7 days	Atelectasis Subarachnoid hemorrhage Ectopic right kidney
59	J.S. 794013	M	3 b	Primary anastomosis Gastrostomy	Living	None
60	C. 794035	M	3 b	Primary anastomosis Gastrostomy	Living	Has twin sister
61	J.D. 794131	M	3 b	Primary anastomosis Gastrostomy	Living	Stricture dilated
62	R.L. 795768	F	3 b	Primary anastomosis Gastrostomy	Living	Stricture dilated
63	R.R. 797249	M	3 b	Primary anastomosis	Living	Stricture dilated
64	L.F. 797394	F	3 b	Primary anastomosis Gastrostomy	Living	Stricture dilated
65	T.S. 799537	M	3 b	Primary anastomosis Gastrostomy	Living	Stricture dilated
66	V.R. 806069	M	3 b	Primary anastomosis Gastrostomy	Living	None
67	L.B. 809626	M	3 b	Primary anastomosis Gastrostomy	Living	None
68	B.J. 810642	M	3 b	Primary anastomosis Gastrostomy	Died at 7 days	Atelectasis Bronchopneumonia

TABLE I.—(*Continued*).

Case No.	Hosp. No.	Sex	Type of Anomaly	Operation	Result	Remarks
69	K.K. 814046	F	3 c	Primary anastomosis Gastrostomy	Living	None
70	S.M. 814283	F	3 b	Primary anastomosis	Died at 18 hours	Twin, second born Tear in tentorium with hemorrhage into falx Bilateral ureteropelvic stenosis Atelectasis Bloody froth in trachea and bronchi Transfusion reaction of kidneys
71	R.H. 808397	F	3 b	Primary anastomosis Gastrostomy	Living	Stricture dilated
72	R.M. 808893	F	3 b	Primary anastomosis Gastrostomy Excision of stricture and re-anastomosis 32 days later	Living	None
73	J.H. 808971	F	3 b	Ligation of fistula Cervical esophagostomy	Living	None
74	R.S. 813425	M	3 b	No operation	Died at 7 days	Horseshoe kidney Imperforate anus Bronchopneumonia *B. coli* meningitis Subdural hematoma
75	D.L. 814656	M	3 b	Primary anastomosis Gastrostomy	Died at 3 months	Stricture dilated
76	M.H. 814863	F	3 b	Primary anastomosis Gastrostomy	Died at 15 days	Anastomosis leak Perforation
77	J.W. 817030	F	3 b	Primary anastomosis Gastrostomy	Living	None
78	T.M. 81973	M	3 b	Primary anastomosis Gastrostomy	Living	Anastomosis leak Stricture dilated
79	P.P. 819965	M	3 b	Tracheotomy Ligation and division of fistula Cervical esophagostomy Gastrostomy	Living	None
80	S.S. 821177	M	3 b	Primary anastomosis Gastrostomy	Living	None
81	M.M. 821745	F	3 b	Primary anastomosis Gastrostomy	Living	Stricture dilated
82	J.J. 822070	F	3 c	No operation	Died at 10 days	Two tracheoesophageal fistulae present. No atresia present
83	R.H. 823012	M	3 b	Primary anastomosis Gastrostomy	Living	Spells of cyanosis Stricture dilated
84	T.F. 823032	M	3 b	Primary anastomosis	Died at 10 days	Critically ill at operation
85	G.Z. 823096	M	3 b	Primary anastomosis Gastrostomy	Living	None
86	M.B. 824282	M	3 b	Primary anastomosis Gastrostomy	Living	None
87	M.W. 824524	F	2	Ligation and division of fistula Gastrostomy Gastroenterostomy	Living	None
88	D.O. 826999	M	3 b	Primary anastomosis Gastrostomy	Living	None
89	S. 830202	F	3 b	Primary anastomosis	Died at 4 days	Congenital heart disease Cor triloculare biventriculatum Two superior and two inferior vena cava
90	K.A. 832069	M	3 b	Primary anastomosis Gastrostomy	Died at 4 days	Pneumonia
91	C.S. 833719	F	3 b	Ligation and division of fistula Cervical esophagostomy Gastrostomy	Died at 17 days	Agenesis of right lung Prematurity Congenital heart disease
92	S. 834171	F	2	Esophagogastrostomy	Died at 3 days	Vesicovaginal fistula Congenital heart disease Large interauricular and interventricular defect Two chamber heart

TABLE I.—(*Continued*).

Case No.	Hosp. No.	Sex	Type of Anomaly	Operation	Result	Remarks
93	A.G. 838024	M	3 b	Primary anastomosis Gastrostomy	Living	Stricture dilated
94	E.J. 839226	F	3 b	Primary anastomosis No gastrostomy	Living	Severe wound infection
95	M.S. 841889	M	3 b	Transpleural primary anastomosis No gastrostomy	Living	None
96	A. MH A42389	M	3 b	Primary anastomosis	Died at 2 days	Died 6 hours postoperative Pentothal curare anesthesia Did not take over spontaneous respiration
97	D.P. CH 29573	F	3 b	Primary anastomosis Gastrostomy	Living	Stricture dilated
98	W.J. CH 29592	M	3 b	Primary anastomosis Gastrostomy	Living	None
99	V.H. CH 30201	M	3 b	Primary anastomosis	Died at 7 days	Bronchial obstruction Unable to effectively clear
100	D.K. CH 30261	F	3 b	Primary anastomosis Gastrostomy	Living	Congenital vascular ring with retrotracheal right subclavian artery
101	G. CH 30458	F	3 b	No operation	Died at 3 days	Pneumonia
102	C.L. CH 30511	M	3 b	Primary anastomosis	Died at 7 days	Premature Weight 3 pounds
103	D.M. CH 30942	F	3 b	Primary anastomosis Gastrostomy	Living	

and cervical esophagostomy had been completed. The patient had returned to the hospital at two years and three months of age for construction of an antethoracic esophagus. In replacing the gastrostomy tube, it was inserted too far. An ischemic necrosis over the tip of the tube resulted in perforation of the stomach wall and peritonitis. This accident has occurred in two other cases. Over-hydration was the probable cause of death in Cases 8 and 16. In Cases 9 and 14 the tracheo-esophageal fistula was re-established four and six weeks after ligation, when the ligatures eroded through the lumen of the esophagus, resulting in recanalization of the esophagus. In both cases the condition was recognized and the esophagus was ligated and divided. Since then the fistulous tract has been divided in all cases after ligation. Ladd[32] previously had noted this complication in two cases after extrapleural ligation of the fistulous communication at the trachea. White and Birdsong[76] reported a similar case in their series. Daniel[8] had one case in which the esophagus became recanalized 24 days after ligating the cardiac end of the esophagus with umbilical tape. The tape ligature was found lying in the lumen of the cardiac end of the stomach at autopsy.

In 68 cases in which a primary anastomosis was carried out there have been 25 postoperative deaths. Patients 33 and 75 died at four and a half months and three months from respiratory infections after successful primary anastomosis. Case 40 died at eight months of age of anoxia at time of secondary operation for resection of a stricture at the site of primary anastomosis. Over-hydration was the cause of death in Case 16, the first primary anastomosis of the esophagus in this series. The patient in Case 17 developed an esophagopleural fistula, diarrhea, hypoproteinemia, and generalized anasarca, with death at 37 days of age. In Case 19 the tracheo-esophageal fistula became re-established, and death at 88 days of age was due to aspiration pneumonia. An imperforate anus with a rectovaginal fistula and bilateral hydroureters and hydronephrosis was also present. In Case 20, roentgen ray studies with

Lipiodol showed that the tracheo-esophageal fistula had become re-established 12 days postoperatively. A gastrostomy was made, and nearly three months later the mediastinum was explored in an attempt to close the tracheo-esophageal fistula. Death occurred five hours after the operation, which was exceedingly difficult due to extensive adhesions about the esophagus. No fistula was found. Postmortem examination showed that the fistula had apparently healed spontaneously. A well-developed hypertrophic pyloric stenosis was present

TABLE II.—*Summary of 103 Cases (1939–1952).*

Procedure	Number of Cases	Living	Dead
No operation	7	0	7
Gastrostomy	5	0	5
Multiple stage	23	13	10
Primary anastomosis	68	43	25
Total	103	56	47

to account for the recurrent spells of regurgitation, choking, and cyanosis that had occurred during the preceding month and had been thought to be due to persistence of the tracheo-esophageal fistula. In Case 24 hypertrophic stenosis was noted at postmortem. Case 30 presented evidence of pylorospasm. Bigger[3] found three cases of hypertrophic pyloric stenosis in his series. He indicated that it might be due to the presence of a gastrostomy tube passed through the pylorus. The patient in Case 42 died at eight and a half months of bronchopneumonia. Postmortem examination showed that the tracheo-esophageal fistula had become re-established.

Leakage at site of anastomosis occurred in 16 cases. A leak was a contributing factor in the death of ten patients. In three of these a tracheo-esophageal fistula had become re-established. In six cases the leak at the site of anastomosis healed with only moderate stricture formation. There have been but three cases of anastomosis leak in the last half of this series, since the importance of good mucosal apposition has been recognized.

In some of the first cases of primary anastomosis of the esophagus in this series a catheter was tied into the lower esophageal segment for traction to relieve tension at the time of anastomosis, as suggested by Daniel.[8] The catheter was left in place for feeding for a week to ten days. The presence of the catheter completely obstructed the esophagus while it was in place. This increased the dangers of aspiration and was the direct cause of death in some cases as well as a probable cause of failure of primary healing of the anastomosis in other cases. Oral feedings were started after a few days, and a gastrostomy was made only after a leak at the site of anastomosis was evident in these earlier anastomoses. Swenson[70] has shown that early gastrostomy and withholding of feedings for ten days after anastomosis of the esophagus rest the esophagus and protect the suture line. Although no gastrostomy was performed in seven successful cases of primary anastomosis in this series, it is now employed routinely.

The high incidence of stricture formation at the site of anastomosis in this series may be more than apparent than real. Roentgen ray evidence of stricture does not always mean a stricture that requires dilatation. Some of the dilatations of the stricture were on the basis of the roentgen ray findings. Many of these strictures improve with growth and function and require no dilatation.

Esophageal strictures in infants have been dilated by the simple method of dilating urethral strictures, described by Hubbard and Leven.[27] Filiforms are passed through the stricture, with Phillips bougies of increasing size used for dilatation. These can be made radiopaque to observe the procedure fluoroscopically if desired. This method has been employed in most of the dilatations in this series. It obviates the

need of a thread through the esophagus for a guide and a gastrostomy for retrograde dilatation.

COMMENT

Earlier diagnosis of congenital atresia of the esophagus and tracheo-esophageal fistula has permitted surgical correction of this anomaly in more cases in recent years. More careful preoperative preparation and postoperative care, as well as improvement in operative technic, have resulted in improved mortality in these cases.

In the patients in whom a multiple-stage operation has been carried out, re-establishing continuity of the alimentary tract presents such a problem that the indications for primary anastomosis should be extended to include all but a few cases.

In this series of cases the operations have been performed by nine different surgeons of the Depatment of Surgery of the University of Minnesota.

SUMMARY

A series of 103 cases seen at the University of Minnesota Hospitals and the Childrens Hospital in St. Paul since 1939 has been reviewed. In 23 patients a multiple-stage operation was carried out and 13 are living. In 68 patients a primary anastomosis of the esophagus was performed, and 43 patients are living. Some of the problems and complications of treatment of these cases are discussed.

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DISCUSSION.—DR. CONRAD R. LAM, Detroit: Dr. Leven is to be congratulated for his pioneering work in this very difficult field, and his continued interest in this same field. I thought there might be some interest in a progress report on two boys whom I presented at the 1948 meeting of the American Thoracic Association in Quebec (J. Thoracic Surg., **18**: 327, 1949). These were infants on whom a multiple-stage operation had been done, for various reasons. I might say that since then I have not had the opportunity to do another because a primary anastomosis has always been possible, except in one or two who did not survive the original exploration.

To review this method very briefly for those who may have forgotten it, I should like to show one or two slides. At the first stage, if an anastomosis is not possible, this stage consists only of closure of the fistula. A day or two later, the second stage, or upper esophagostomy is done. After two days we proceed with the third stage, bringing out the entire lower end of the esophagus, thus avoiding the production of a gastrostomy. Subsequently, feeding for a year is very conveniently accomplished through the distal esophagostomy.

[Slide] At the age of one year, the two esophagostomies were connected by this conventional skin tube.

[Slide] This shows the plastic procedures which were eventually utilized to complete the operation. At the time these children were shown before, their ages were five and three; their ages are now nine and seven. Here is the older one, whose photograph was taken a few days ago. You can see that the development is normal. There are scars on these children, of course.

[Slide] I think I should take advantage of showing this boy with his clothes on, as he is normally seen by his friends. The other child, who is seven years old, is in a similar state of nutrition. He had measles and could not come in for his progress picture. This method is different from those which utilize a portion of the gastro-intestinal tract, such as the jejunum. The result is an all-squamous epithelium esophagus.

DR. CAMERON HAIGHT, Ann Arbor, Mich.: Dr. Leven and his associates deserve great credit for their work in the correction of esophageal atresias. These infants present a major problem, not only because of the nature of the anomaly but also in view of the fact that they are often premature. Furthermore, as Dr. Leven has said, they may have other congenital anomalies which, in themselves, are not compatible with life over a period of time.