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Historical Vignette

The history of surgery for esophageal atresia

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ABSTRACT

Until the successful repair of esophageal atresia (EA) and distal tracheoesophageal fistula (TEF) in 1941 by Cameron Haight of Ann Arbor, MI, every infant operated upon for this anomaly died within days and often hours of surgery. A key step was the posterior extrapleural approach to the mediastinum pioneered by Charles Mixter of Boston in 1929 that gave direct exposure of the anomaly without entering the pleural cavity and collapsing the lung. From 1936 to 1939 Thomas Lanman, also of Boston, made five unsuccessful attempts at primary repair of EA. His experience established the basic principles of early radiological diagnosis and prompt surgical intervention to minimize the risks of aspiration pneumonia, dehydration, and inanition.

In 1939 N. Logan Leven of Minneapolis and William Ladd of Boston independently had the first long-term survivors of EA with a series of operations to construct skin-lined tubes on the anterior chest wall that connected an esophagostomy to a gastrostomy. Haight first tried primary repair in 1939, finally succeeding in his fourth case in March 1941.

In their publications Lanman (1940), Haight (1943 and 1944), and Ladd (1944 and 1947) presented case-by-case chronologies. The evolution of surgical management thus can be traced from a fatal condition to one where survival became the expected outcome. History recognizes Haight for his work with EA, not only for its first successful primary repair, but also his lifelong dedication to its surgical management.

Contents

1. Early attempts

In 1697 Thomas Gibson made the first detailed account of an infant born with the most common pattern of esophageal atresia (EA), its combination with a distal tracheoesophageal fistula (TEF; EA/TEF):

> *About November 1696 I was sent for to an infant that would not swallow. The child seem'd very desirous of food, and took what was offer'd it in a spoon with greediness; but when it went to swallow it, it was like to be choked, and what should have gone down returned by the mouth and nose, and it fell into a struggling convulsive sort of fit upon it. [The baby] was fleshy and large, and was two days old when I came to it but the next day died* [1].

Death was especially cruel, with infants dying from aspiration from the EA, pneumonia from reflux of gastric juice through the TEF, or dehydration and starvation from inability to swallow.

The problem was how to reach the anomaly deep in the posterior mediastinum, inaccessible from the neck and abdomen. It defied a straightforward surgical solution. In his 1869 textbook, Thomas Holmes wrote:

> *It becomes worthwhile to inquire whether the occlusion is ever limited to a mere septum, which the surgeon might hope to perforate by cutting*

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down upon it. The evidence…discourages the hope that this malformation is remediable by operation [2].

In 1913 Joseph Brennemann, a pediatrician in Chicago, reported two more fatal cases of EA/TEF: the first after a feeding jejunostomy, the other following a gastrostomy. After watching one of his patients cough and choke to the point of cyanosis after a gastrostomy feeding, he concluded, "[The] utter hopelessness of these cases, if untreated, justifies surgical procedures that would otherwise seem too daring [3]."

It was probably with Brennemann's encouragement that his surgical colleague Harry Richter tried to repair the anomaly through an open thoracotomy in two patients. Surgical exposure of the anomaly proved difficult. Richter wrote:

Lack of familiarity with the surgical anatomy of the parts will obviously be a source of great embarrassment to most general surgeons. It was to the author. The smallness of the parts will in a newborn infant and the peculiarly difficult site of operation made the hazard apparently insuperable [4].

Anticipating problems with ventilation when the chest was opened, he placed a tube into the trachea and attached it to an air pump and rheostat. Richter gave himself only five minutes to complete the operation, knowing his primitive breathing apparatus could not sustain life. It was a testament to his surgical ability that he got both infants off the table alive. Each died only hours after surgery [4].

With the problem of ventilation during open thoracotomy unsolved, indirect operations were devised to address the communication between the trachea and the gastrointestinal tract. In 1936 Mims Gage and Alton Ochsner of New Orleans published their technique, the ligation of the esophagus at the level of the cardia from an upper midline laparotomy [5], an intervention already tried by Isadore Ravdin of Philadelphia and reported in passing by another author in 1933 [6]. In 1937 Hugh Gamble of Greenville, MS, divided the stomach and made two gastrostomies, one to the proximal segment to decompress the distal TEF, and a second as a standard feeding gastrostomy [7]. None of the infants survived more than a few days. From their experience Gage and Ochsner concluded:

The treatment of this unfortunate anomaly is entirely surgical. The ideal operation would be the separation of the esophagus from the trachea and an end-to-end anastomosis of the upper to the lower segment. Such an extensive intrathoracic procedure is not justified in a newborn infant, however, and the operation would always be finished as a postmortem procedure [5].

2. Charles Mixter and Thomas Lanman

In 1929 Charles Mixter, a visiting surgeon at the Children's Hospital, approached the anomaly directly in the mediastinum from the right side of the back with the infant prone. He kept the pleura intact and pushed it aside to keep the lung inflated and still allow spontaneous breathing. After transecting the TEF and suturing the defect in the trachea, he took the distal esophagus and exteriorized it through a separate stab incision in the back as a dorsal esophagostomy that could serve as a site for catheter feedings. His first patient died just as the operation ended; his second and only other patient survived just a little more than two days.

Mixter's procedure became the preferred operation for EA/TEF in Boston (15 of 32 cases reviewed by Lanman in 1940) [8]. More importantly, both Thomas Lanman of Boston (Fig. 1) and Cameron Haight of Ann Arbor, MI (Fig. 2), would use Mixter's posterior extrapleural approach for their attempts at primary repair.

Beginning in 1933, Lanman began to manage the majority of cases of EA in Boston, 18 of 32 (56%) of the total caseload in his review, and 17 of the 24 (71%) during the eight years between 1933 and 1940 [8]. He saw that Mixter's operation only addressed the TEF. A patient surviving any length of time had to contend with aspiration from the proximal EA. He added a proximal esophagostomy in two patients, the first dying at two weeks of age but the second surviving a month.

During the second case, Lanman saw that the proximal esophagus and the TEF lay close enough for an anastomosis had he tried. When the next case arrived on New Year's Day 1936, a two-day-old girl, instead of Mixter's operation, he went ahead and did the first primary repair of an EA and distal TEF in nearly a quarter century. The patient died only three hours after surgery. At postmortem examination the baby had bilateral acute bronchitis and what the pathologist described as moderate atelectasis, an indication that the respiratory status was too fragile to allow the child to survive despite a technically successful operation [8].

Over the next three-and-a-half years, from January 1936 to March 1939, Lanman made four more attempts at primary repair. Like the first, each ended in the death of the infant. Like any pioneer, he experienced for the first time many of the complications that modern surgeons now recognize as hazards of surgery for EA — anastomotic leak, right heart failure from excessive fluid administration, and the classic board examination question: EA/TEF associated with duodenal atresia and imperforate anus. In the last case Lanman deduced that division of the TEF was the priority, an answer that would have earned him a pass in an oral exam today. But without modern neonatal and respiratory care, all of his patients died [8].

As if he was frustrated with his lack with progress to have a patient survive, Lanman abandoned primary repair. In the final five patients in his series he did three ligations of the cardia (the Gage–Ochsner operation) and two stomach transections (the Gamble procedure). From his experience he knew that such indirect operations had no chance of success. That he resorted to such procedures suggests desperation.

He did one final EA case in 1941, one year after his 1940 publication, another death. In all Lanman managed 19 cases, none of them surviving. He never did another operation for EA.

Despite his failures, he and his colleagues at the Children's Hospital set modern principles of the management of EA [8]. In 1929 Edward Vogt at the Infant's Hospital in Boston established that the diagnosis could be made from the inability to pass a tube into the stomach and the presence of air in the gastrointestinal tract, obviating the need for oral radiological contrast [9]. Early operation soon after birth minimized the risk of aspiration pneumonia. And thanks to Mixter, the posterior extrapleural approach kept the lungs inflated during the operation.

Having tried nearly every approach to EA, Lanman's conclusion in his 1940 article was almost forlorn. He wrote that primary repair of EA "should be the method of choice. … That this method will eventually be successful I have no doubt [8]."

3. William Ladd

After Lanman stopped doing EA surgery, Ladd managed most of the EA cases in Boston, more than three-fourths of the EA patients in Boston from 1941 to 1944 (25 of 33 cases, 76%) [10], an abrupt transition that suggests that either Lanman quit or Ladd sidelined his colleague. Apparently disillusioned by Lanman's failed attempts at primary repair, Ladd came up with a different operative strategy that avoided any attempt at esophageal anastomosis. On a baby with EA/TEF born on November 29, 1939 (the birthdate became important later), Ladd did a gastrostomy the following day. The baby proved to be tough, surviving four-and-a-half months of gastrostomy feedings in the presence of a TEF before Ladd divided the TEF and did a proximal esophagostomy.

The child thus protected from aspiration, Ladd then had the time to construct an antethoracic skin-lined tube that bridged the esophagostomy in the neck to the gastrostomy site, a series of operations that he called marsupialization of the esophagus (Fig. 3).

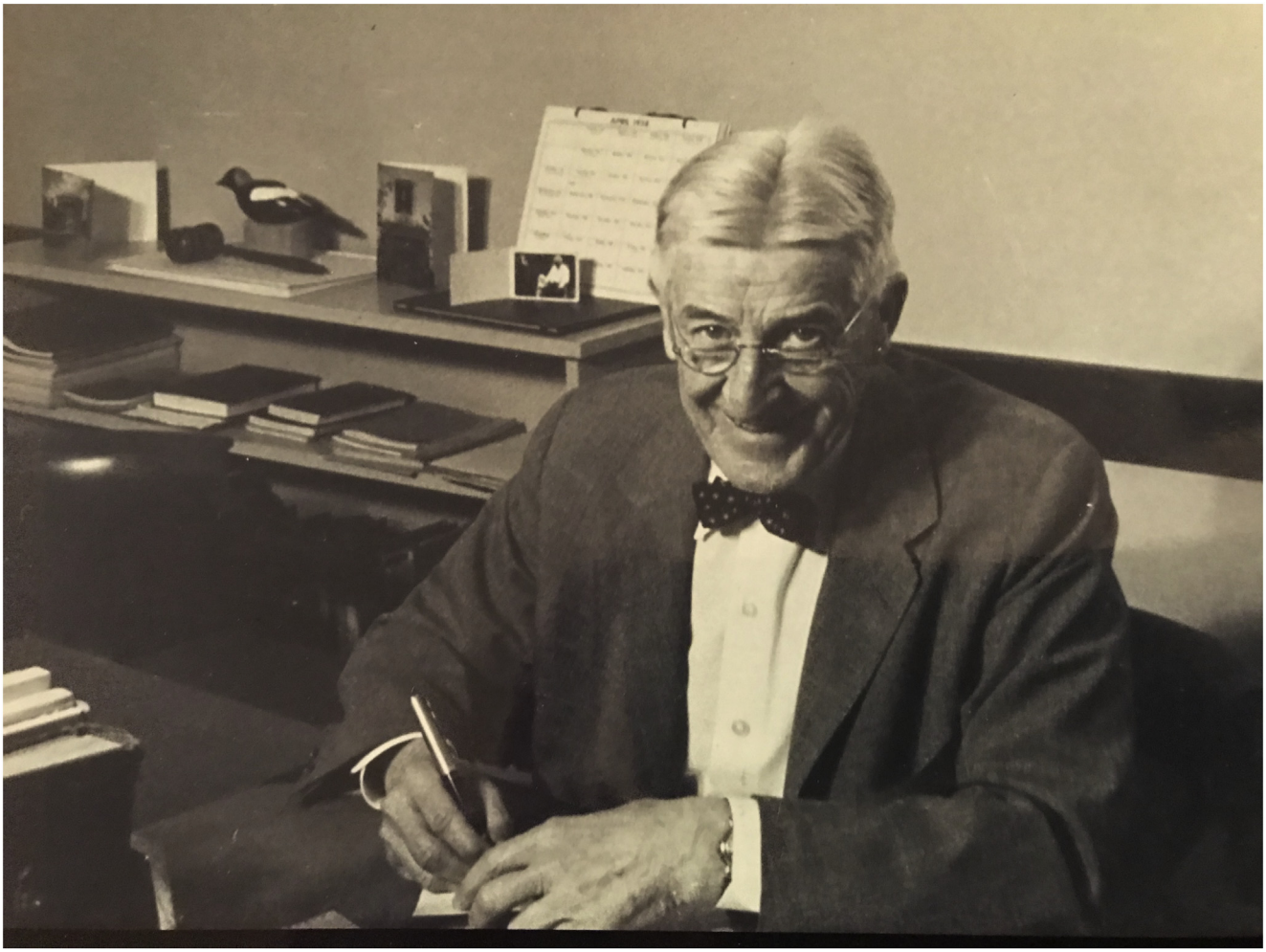

Fig. 1. Thomas Lanman.

Swallowed material drained by gravity into the stomach. The neoesophagus was an unsightly bulge on the child's chest (the first patient was a girl) that extended from the base of the neck to the epigastrium, but Ladd assured his readers that she ate normally [10].

And most importantly, she was alive. Thereafter, he changed the sequence of operations and shortened the intervals between the life-saving operations: First ligate the TEF shortly after birth and do the gastrostomy and esophagostomy days later. Once the respiratory tract was thus protected, construct the esophagus over the ensuing months and years [10].

Despite the problems created by the out-of-sequence operations, his original patient was the first survivor of EA at The Children's Hospital. As far as Ladd knew, he had the first survivor anywhere.

Ladd used marsupialization of the esophagus in six more patients from 1941 to 1942. Clearly, he was making progress: his patients were surviving longer: 35, 106, and 147 days. A patient, born in December 1941, became his second long-term survivor. When he reported his series in the *New England Journal of Medicine* in 1944, he had done 15 marsupialization procedures (13 EA/TEF, 2 isolated EA) with 9 survivors (60%) [10].

4. Cameron Haight

When Lanman was struggling with primary repair in the mid to late 1930s, at the University Hospital at Ann Arbor, MI, four surgeons shared EA cases on the pediatric service with gastrostomy alone as the sole intervention as late as 1937 [11]. There were no cases in 1938, perhaps because the diagnosis was widely held to be fatal and therefore no cases were referred to the University Hospital.

In 1939 and 1940, Haight and John Alexander tried their hand at primary repair and alternated four cases. Both of the Alexander's patients suffered fatal technical complications, one from respiratory insufficiency after entry into the pleura and a second from bleeding from an injury to the aorta. From then, he left the task entirely to Haight.

Haight had no better luck. Hiram Langston of Savannah, GA, then a resident in training in thoracic surgery at Ann Arbor, wrote in a 1984 memoir that Haight had just had another baby with EA die from sepsis when he received word of the transfer of yet another case. “Haight was not eager for the anguish of another disappointment,” Langston wrote [12].

Howard Barkley, another one of Haight's residents, urged him to try. The baby, a girl, had already proved herself to be a survivor. At 12 days of age, she had somehow escaped severe respiratory complications. Aside from a transient episode of bleeding shortly after birth, the baby was free from other medical problems. Her doctor in the upper peninsula town of Marquette had injected fluid into her subcutaneous tissues to maintain her hydration. She received another dose before she embarked on the 500-mile trip to Ann Arbor, and at two more stops along the way for additional doses arranged by her far-sighted physician. On arrival Haight saw that, at 3.66 kg, she “was uncommonly robust for a victim of this defect.” He agreed to make the attempt [12].

On March 15, 1941, with the baby restrained in a lateral position in a rigid frame, Haight used a paravertebral incision on the left side, opposite the right-sided approach used by Richter and the Bostonians. He relied on local anesthesia until he reached the

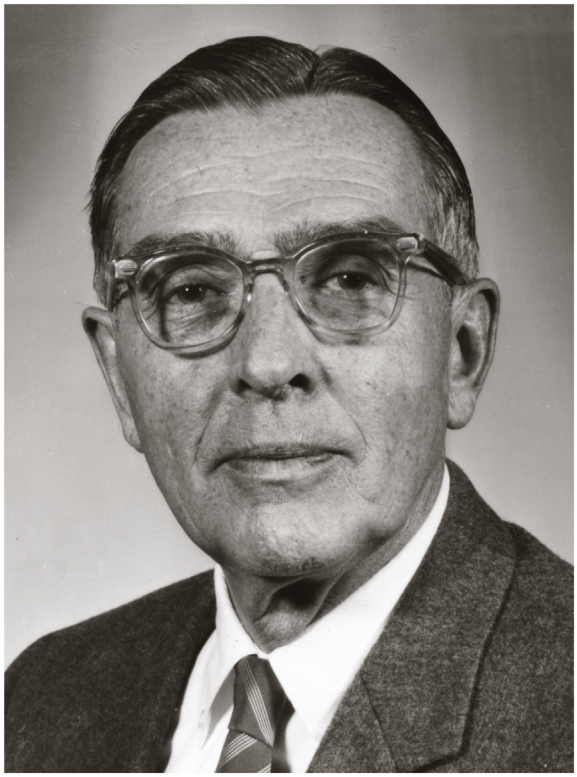

Fig. 2. Cameron Haight. Photo courtesy of the University of Michigan Millennium Project.

chest and needed open drop ether anesthesia to quiet her respiratory efforts. He removed the posterior four centimeters of the second through fifth ribs, staying within the periosteum. Because he was in the left chest, exposure of the anomaly required forward mobilization of the aorta and left subclavian, and ligation of the intercostal vessels, the phase of the operation that likely led to the injury to the aorta that dissuaded his partner from doing any more EA surgery.

Once he saw the TEF, he used a single silk tie to ligate the connection. He performed a single-layered esophageal anastomosis with silk sutures. A tight-fitting mask assured enough positive pressure ventilation to reinflate her lungs at the end of the procedure. Before closing, he left a single rubber drain in the area.

The baby survived the operation in remarkably good shape. Haight gave her 0.25 grains of sulfathiazole by rectum every six hours in an attempt to prevent infection. He relied on subcutaneous injections of saline to maintain hydration. Fluid balance proved difficult, edema developing on the second postoperative day, which eventually resolved. The baby eagerly sucked the two milliliters of sterile water she was offered every two hours.

Just as Lanman was the first to encounter many of the operative complications of esophageal surgery in infancy, Haight had to deal for the first time an anastomotic leak and the development of an esophageal stricture. Saliva leaked from the incision on the seventh day after the operation. On X-ray, all of the swallowed lipiodol radiological contrast drained from the esophagus and came out the wound.

Haight stopped all oral feedings and placed a gastrostomy on the 10th day for feedings of milk, water, and Karo syrup. Three days later, the formula began to leak out the wound. A rubber catheter was passed into the child's mouth in a hope that it could make its way past the anastomosis and into the stomach. One can only imagine Haight's horror when the tube poked out the incision in the baby's back.

Ten days from the gastrostomy, 20 days after her original repair, the baby began to burp milk, a sign that the anastomosis was patent. Another contrast study showed that the leak had resolved, showing for the first time the lesson that most esophageal leaks after EA repair heal. The baby hungrily gulped the 2 mL of formula every two hours offered the next day, a volume she appeared happy to have gradually increased. She thrived on oral feedings, so the gastrostomy was removed when she was six weeks old. Follow-up contrast studies confirmed that a stricture had developed at the level of the anastomosis. Haight was able to wait until she was 17 months old before dilating it [12].

Two years later, in 1943, Haight reported his landmark operation in *Surgery, Gynecology, and Obstetrics*. It included 15 cases at the University Hospital since 1935, of which 10 were his patients and 5 had undergone primary repair. Of the 15, all had died except for his single success. Proudly he included a figure of his surviving patient standing in a playpen (Fig. 4) [11].

After his achievement, more referrals came to the University Hospital: 2 more in 1941, and 6 in 1942. Of the 8, Haight attempted 5 more primary repairs, the first 4 ending in death. Then, in the final case of 1942, he had his second survivor, the 18th case of the University Hospital series and his 10th attempt at primary repair. It sparked a flurry of 14 referrals in 1943, of which nine were operated upon, with four survivors [13].

In May 1944, Haight presented the total University Hospital experience of EA to the American Surgical Association (ASA) meeting in Chicago, a total of 32 cases with his personal caseload of 17 attempts at primary repair with 6 survivors. Notably he finally began to use a right-sided approach on his 11th attempt at primary repair in 1943, case 22 in the University Hospital series [13].

5. Ladd and Gross

Ladd's patrician demeanor was tested during the late 1930s and early 1940s. His trials began in August 1938, when Robert Gross, then chief resident under Ladd and once his favored protégé, did the first li-

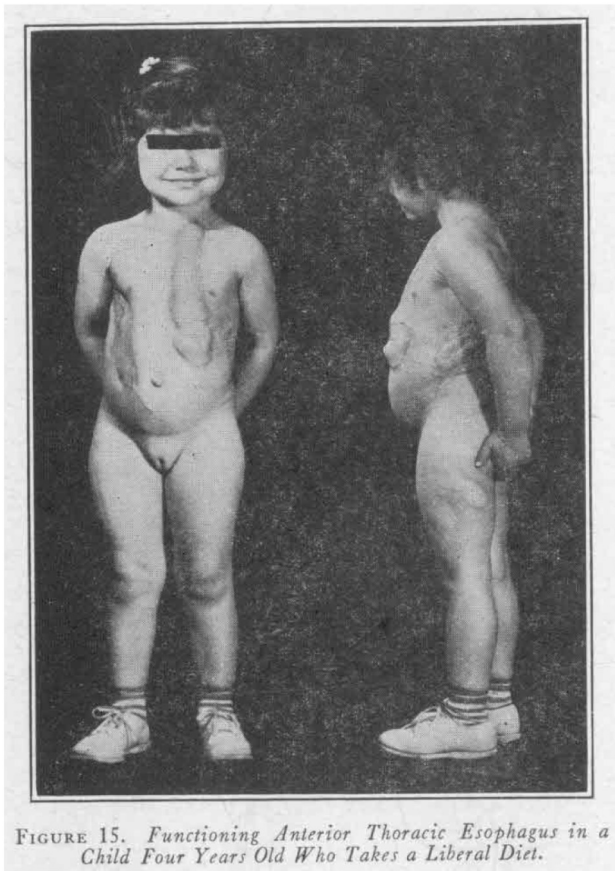

FIGURE 15. *Functioning Anterior Thoracic Esophagus in a Child Four Years Old Who Takes a Liberal Diet.*

Fig. 3. Ladd's original survivor after marsupialization of the esophagus. From Ref. [10]. Copyright Massachusetts Medical Society.

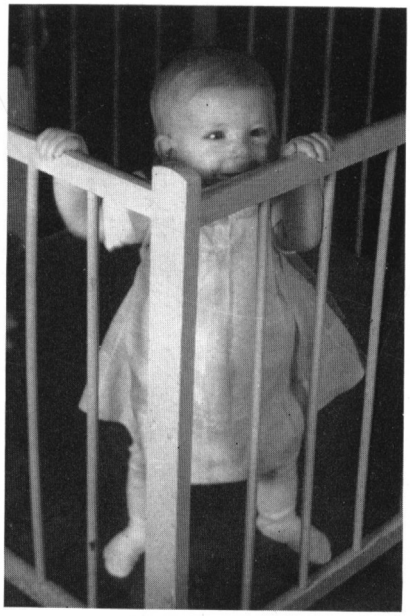

Fig. 8. Photograph of patient 11¼ months after operation.

Fig. 4. Haight's original survivor after surgery for EA/TEF. From Ref. [11]. Copyright American College of Surgeons.

gation of a patent ductus arteriosus without permission during the latter's customary month-long August holiday. When he returned, the chief summarily fired his resident, who remained on staff while the hospital board worked to retain the brilliant, if insubordinate, young surgeon [14].

Lanman, in charge during Ladd's absence, had given Gross the okay to proceed, a role in the imbroglio that may have contributed Ladd taking over EA care from him. Ladd's successful marsupialization operation of 1939 was conspicuously absent from Lanman's 1940 review, which reviewed every other case done in Boston from 1929 to 1940. Either Lanman knew to leave Ladd's case out, or Ladd explicitly told him not to include it.

Time passed and Ladd waited to write up the case, apparently wanting to finish the sequence of marsupialization procedures before publishing. He must have been stunned when he saw an article by N. Logan Leven of St Paul, MN that appeared in the *Journal of Thoracic Surgery* in August 1941 that presented a strategy identical to his own. Unlike Ladd, Leven went ahead and published his operation before construction of the neoesophagus, which he wrote that he planned to do sometime in the future. Instead his article showed a rubber tube connecting the esophagostomy and stomach [15].

Ladd had been scooped. Moreover, Leven's patient was born on November 28, 1939, one day before Ladd's infant. In addition to publishing his technique before Ladd, Leven could lay claim of having the oldest long-term survivor of EA.

By the 1940s, other surgeons were also trying their hand at EA surgery, notably Robert Shaw in Dallas and Rollin Daniel in Nashville. With the largest experience, Ladd and Haight represented the two options: Ladd and marsupialization in Boston *versus* Haight and primary repair in Ann Arbor. Both only had a handful of survivors, but Ladd's approach seemed conservative and safe, while Haight's was bold and risky.

There was an important difference in outcome, however. Each of Haight's surviving patients swallowed food with a native esophagus. Ladd's patients had the unattractive sausage-shaped bulge on their chests that one could imagine easily becoming clogged, despite Ladd's reassurances to the contrary.

In October 1941, under Ladd's own roof at The Children's Hospital, Gross went against his boss once again and made the first attempt at primary repair in Boston in two-and-a-half years, without doubt in response to Haight's success half a continent away seven months before. Gross's patient lived 30 days, longer than Lanman's best of 9 days. Gross tried again one year later in October 1942, the patient dying one day later. Despite the two deaths, it was clear that primary repair would be Gross's preferred option, and not his boss's complicated marsupialization procedure.

Ladd probably felt the pressure to publish his successful experience with marsupialization. Throughout the early 1940s, he still had not published as a number of other surgeons published their experience with EA, including Shaw (1939) [16], Lanman (1940), Leven (1941), Haight's landmark paper of 1943, and Daniel (1943) [17]. On May 25, 1944, Ladd's experience with marsupialization finally appeared in the *New England Journal of Medicine* [10].

But once again Ladd had been preempted. Three weeks previously, Haight made his presentation before the ASA, the most prominent surgical society in the country. Haight's achievement made Ladd's paper irrelevant before it even appeared in print.

Without regard to Ladd's pride, the Association chose him to discuss Haight's paper. With a Boston Brahmin's grace, Ladd congratulated Haight. But he wanted to set the record straight: both he and Leven had four-and-a-half-year-old survivors with antethoracic skin tubes. And while Haight had six survivors, Boston had 11. “It would seem that the multiple-stage operation is a considerably safer procedure,” Ladd said [13].

Ladd went on to show 10 slides of his marsupialization procedure. Whether he was trying to hijack Haight's presentation, it was at least in bad taste. One surgeon of the era, Mark Ravitch, wrote on the rules of etiquette when discussing a paper at a scientific meeting:

> *This [discussion of a paper] is not the occasion to present another paper on the same subject (often one turned down for the same program), still less is it in good taste to drag in by the tail some remotely connected case or observation or experience, even if some tenuous connection can be indicated between it and the paper allegedly under discussion* [18].

6. Survival

Surgery for EA had reached an inflection point to where survival was becoming the expected outcome. In 1947 Ladd and Orvar Swenson updated the Boston experience from 1940 to 1946 (once more excluding Lanman's failures). More than half of the cases (43 of 75, 57%) were managed using Ladd's multistage approach. But now primary repair was performed more frequently, representing more than 40% of their total caseload (32, 43%). Given the small numbers in the two groups, outcomes were equivalent: 37% overall survival for staged reconstruction after proximal esophagostomy, 44% after primary repair, for a 40% overall survival. “The operation of primary anastomosis,” Ladd and Swenson wrote, “done via an approach through the right back, is beyond question the operation of choice when feasible [19].”

In 1952 Leven updated his experience at the University of Minnesota. Abandoning the marsupialization procedure that he and Ladd had tried the previous decade, he embraced primary repair, reporting 43 survivors of 68 patients (63%). Instead of the posterior mediastinal approach through a vertical incision through the posterior segments of multiple ribs, he used a standard posteriolateral thoracotomy through the bed of the fourth rib that stayed in the extrapleural space, the favored approach for open procedures today (but through the interspace without rib resection) [20].

Haight, in his presidential address before the American Association for Thoracic Surgery (AATS) in May 1957 in Chicago, summarized his nearly two decades of work that started nearly 20 years before [21]. He followed each of his cases in detail, which gave him an intimate understanding of each aspect of care and every pitfall and complication.

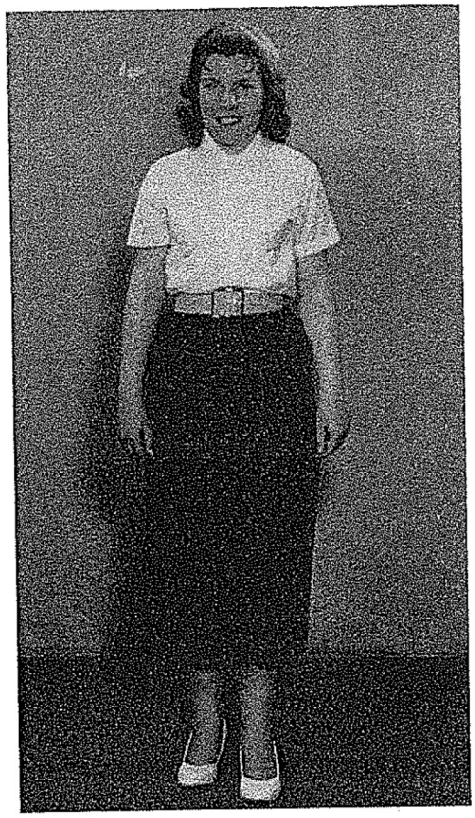

Fig. 5.—First case of recovery following primary anastomosis for esophageal atresia with tracheoesophageal fistula (J. M., Case 10).

Fig. 5. Haight's original survivor, 16 years later. From Ref. [21]. Copyright Elsevier.

His experience was now 200 cases as primary surgeon. Of his second 100 patients, 77 had undergone primary repair, with 52 (68%) surviving the initial operation. He discussed late complications and deaths, providing details that reflected his profound understanding of a lifetime of study and follow-up of his patients. Of the 52 survivors of the initial operation, 41 (79%) were still alive at the time of his address. Late deaths after initial survival after surgery were particularly upsetting to him, as most occurred from complications of surgery, such as recurrent TEF, that in Haight's view could have been avoided [21].

At the 1986 ASA meeting, 51 years after the first recorded operation for EA/TEF at the University Hospital and 16 years after Haight's death, Haight's professional descendants at the University of Michigan, Peter Manning, Arnold Coran, and Herbert Sloan, reported that EA/TEF now had an overall survival rate of 83%, with a 100% survival rate for infants weighing more than 2.5 kg and free from other congenital anomalies. Pneumonia, long a primary cause of death, was no longer an independent risk factor for survival [22].

At his 1957 presidential address Haight showed an updated photo of his first survivor, the baby featured in his first paper on the subject in 1943, now a young woman aged 16 years (Fig. 5) [20]. The epitome of his great achievement, she would be the last patient Haight saw before he died in 1970 at age 70 [12].

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