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Elongation of esophageal segments by bougienage stretching technique for long gap esophageal atresia to achieve delayed primary anastomosis by thoracotomy or thoracoscopic repair: A first experience from China ☆

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ABSTRACT

Objectives: The treatment of long gap esophageal atresia (LGEA) is one of the most challenging congenital malformations in neonatal surgery. A preoperative bougienage stretching technique for elongation of the two segments of esophagus is applied to achieve utilizing the native esophagus to establish esophageal continuity by open or thoracoscopic approach.
Methods: From January 2015 to May 2017, 12 neonates who suffered from LGEA were admitted to our department. They were divided into 2 groups (A and B) according to their admission time. They all accepted bougienage stretching technique before esophageal anastomosis.
Results: Initially the lengths of esophageal gap in 12 infants ranged from 4 to 7.5 vertebral bodies ($M = 5.8 \pm 1.1$). The gap lengths became −1 to 2.5 vertebral bodies after bougienage stretching technique and tension-free anastomosis were performed successfully for all 12 cases: Group A (n = 5) by thoracotomy and group B (n = 7) by thoracoscopic approach. 12 cases have been followed up for 1–25 months ($M = 12.4 \pm 8.5$) after definitive surgery.
Conclusions: Bougienage stretching technique for LGEA is feasible with satisfactory clinical results. Thoracoscopic approach is a good choice for primary anastomosis in LGEA.
Levels of evidence: Treatment Study Level IV

The treatment of long gap esophageal atresia (LGEA) in neonates is one of the most challenging congenital malformations in neonatal surgery. Most pediatric surgeons agree that delayed primary anastomosis is a preferable choice, but the proximal and distal segments of the esophagus are usually too far apart, which leads to primary anastomosis abandoned. Thus, various techniques have been used to bridge the gap between these two segments [1]. Circular and spiral myotomies (Livaditis) were first described in 1972 and 1987 respectively, which can theoretically provide an additional length of 0.5 to 1.0 cm [1]. Various elongation techniques including Foker and Kimura techniques have been widely advocated after the 1990s [1] and a thoracoscopic traction technique was reported recently [2]. Replacement techniques involving gastric and intestinal ones have also been commonly accepted nowadays while they brought various complications caused by complex surgeries and other facts [1]. However, as Meyers's adage in 1974 emphasized, all attempts should be made to conserve the native esophagus for overcoming more complications [3]. This article described a preoperative bougienage stretching technique [4–6] for elongation of the proximal and distal segments to achieve utilizing the native esophagus to establish esophageal continuity by thoracotomy or thoracoscopic approach.

1. Materials and methods

From January 2015 to May 2017, 12 neonates who suffered from LGEA were either admitted or transferred to Department of Pediatric Surgery, Xinhua Hospital Affiliated to Shanghai Jiao Tong University School of Medicine. Gestational age varied from 32.6 to 40.4 weeks ($M = 37.5 \pm 2.7$). Birth weight ranged from 2050 to 3000 g ($M = 2464 \pm 316.4$). Age at time of gastrostomy varied from 1 to 12 days ($M = 5 \pm 3.6$) (Table 1). Among these cases, one was diagnosed of EA Gross type C, who had accepted thoracotomy at a local hospital after birth while the gap distance between the upper and lower esophageal segments was too far to perform anastomosis. Therefore, he only underwent fistula ligated and gastrostomy, and was then transferred to our institute. The 11 others were typically pure EA of Gross type A. 12 patients were fed by gastrostomy to keep nutritional status well

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Table 1
Demographics of patients with LGEA.

Case		Birth histories (weeks)	Delivery mode	BW (g)	Gastrostomy Received Age (days)
A	1	G3P2, 38^{+5}	C-section	2540	3
	2	G5P2, 35^{+6}	C-section	2430	3
	3	G1P1, 37	C-section	2300	2
	4	G2P2, 39^{+5}	Eutocia	3000	2
	5	G1P1, 40^{+3}	C-section	2900	10
B	6	G2P1, 40^{+6}	Eutocia	2650	7
	7	G3P2, 36^{+1}	Eutocia	2350	9
	8	G2P2, 34^{+4}	C-section	2300	5
	9	G1P1, 38^{+6}	C-section	2100	1
	10	G1P1, 40	Eutocia	2800	2
	11	G3P0, 32^{+4}	Eutocia	2050	12
	12	G1P1, 34^{+4}	Eutocia	2150	4

controlled. Continuous suction in the upper pouch was applied to avoid aspiration pneumonia.

Then the stretching technique was performed in the following steps. A bougie (size φ5–6, B10105, Shanghai Medical Instruments (Group) Ltd., Corp. Surgical Instruments Factory) was inserted into the upper esophageal pouch through oral cavity with appropriate downward longitudinal force to increase esophageal length by tissue stretch and growth, while the elongation of distal pouch was achieved via the gastrostomy using the same bougie to give upward pressure. The first insertion for a new patient was performed under X-ray to find the correct pathway by esophagram in order to avoid injury or false passage formation of distal segment while enabling us to evaluate the initial gap distance at the same time. The following insertions and stretches can be performed in wards but usually by a certain doctor for one certain patient because he can roughly know the direction and how to adjust the angle of the bougie. The stretching was performed once a day, each lasting 10–15 min. The gap length was evaluated every 2 weeks by esophagram under applying downward and upward stretching. During the elongation period, continuous upper pouch suctioning and nutritional support were maintained.

We divided these 12 patients into 2 groups according to their admission time: the first 5 patients were group A, and the latter 7 patients belonged to group B.

2. Results

Initially the gap lengths between esophageal segments in 12 infants ranged from 4 to 7.5 vertebral bodies ($M = 5.8 \pm 1.1$) on admission (Fig. 1): group A was 5 to 7.5 vertebral bodies ($M = 5.8 \pm 1.2$), and group B was 4 to 7 vertebral bodies ($M = 5.9 \pm 1.2$). The gap became markedly shortened after stretching technique with a total elongation rate of 0.4–2.3 vertebral bodies every week ($M = 1 \pm 0.6$): the rate of group A was 0.4–0.8 vertebral body ($M = 0.54 \pm 0.2$); the rate of group B was 0.8–2.3 vertebral bodies ($M = 1.3 \pm 0.5$). The final lengths of esophageal gap before anastomosis ranged from −1 to 2.5 vertebral bodies: group A was −0.5–2.5 vertebral bodies and group B was all −1 vertebral body (Figure 2). The duration of elongation process which started from initial stretching till anastomosis performed varied from 3 to 14 weeks ($M = 7.4 \pm 3.2$): group A was 6 to 14 weeks ($M = 10 \pm 3.2$); group B was 3 to 8 weeks ($M = 5.6 \pm 1.7$) (Table 2).

Finally tension-free anastomosis was performed successfully for all 12 cases. 5 patients in group A accepted the surgery by thoracotomy, 3 by Livaditis, 1 by end-to-end and the left one by flip-flap anastomosis. Meanwhile, 7 patients in group B underwent the esophageal reconstruction by thoracoscopic approach: 1 by end-to-end and 6 by Livaditis. And nasojejunal tubes (CH10-145, Flocare) were placed for all 12 patients during the anastomosis.

All 12 cases have been followed up for 1–25 months ($M = 12.4 \pm 8.5$) after definitive surgery. They were initially fed by nasojejunal tube on 4th–10th postoperative days and started oral

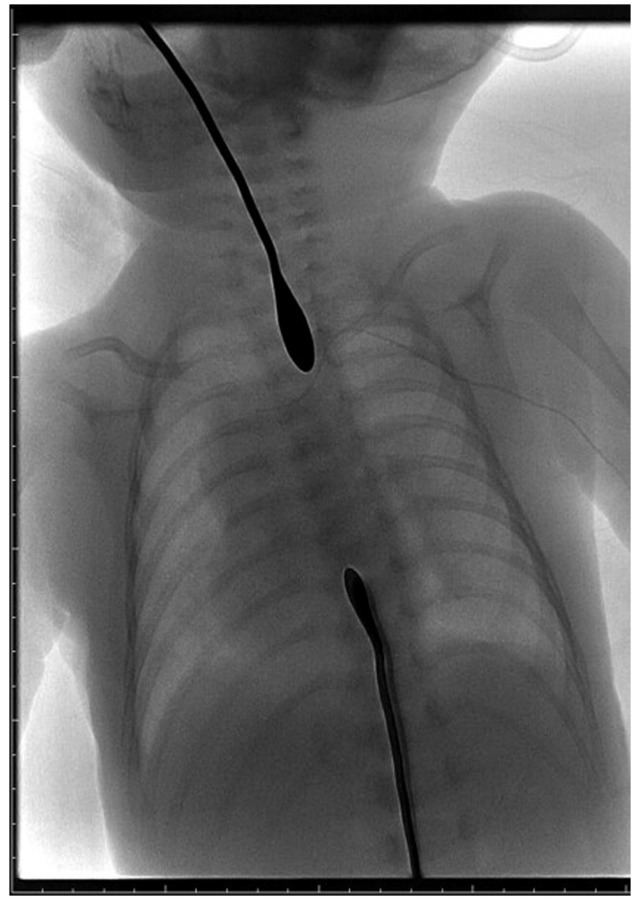

Figure 1. Esophagram showed the gap distance was 5 vertebral bodies before internal traction.

feeding 1–2 weeks later. Among all cases, 6 had pulmonary infection after operations and all were cured by antibiotics and airway caring, 3 in group A and 3 in group B. 1 case was complicated with tracheomalacia which might have been caused by tracheal dysplasia and repeated intubation for severe pulmonary infection. 7 cases (3 from group A and 4 from group B) had anastomotic stricture and accepted dilations every 1 or 2 weeks during the subsequent few months. The strictures of all 7 cases had obvious improvement after around 3–10 times of dilations (Fig. 3). Postoperative esophagrams showed that 2 patients suffered anastomotic leakage but were cured by drainage and supportive treatment, 1 in group A and 1 in group B. During the follow-up, no severe gastroesophageal reflux (GER) occurred in all 12 cases till now. 11 cases have built full oral intake without vomiting or other feeding difficulty. The latest one was fed partially by oral intake and partially by nasojejunal tube now.

Discussion

The definition of long-gap esophageal atresia (LGEA) is in largely controversial and multiple criteria condition. Spitz defined it as "inability to achieve primary end-to-end anastomosis" [7]. In our cases, gap length ranged from 4 to 7.5 vertebral bodies which led to a loss of primary anastomosis that can be defined as LGEA without doubt.

It is uniformly known that the best effect of radical surgery is to achieve utilizing the native esophagus to establish esophageal continuity which could maximize the function with the least complications. However, it requires enough length between two segments to help finish a tension-free anastomosis. And the first choice to achieve increased segments seems to be simply give esophagus time to grow naturally without invasive manipulations [1]. So some centers allow the esophagus of patients whose gap length was over six vertebral bodies to grow for 2 months prior to operation [8]. Usually the maximum natural