Demographic characteristics in patients with short-gap and long-gap esophageal atresia: a comparative study

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Background **The knowledge of the size of the gap between esophageal ends in esophageal atresia (EA) before surgery is of clinical importance. The aim of this study was to compare the demographic characteristics between patients with short-gap esophageal atresia (SGEA) and long-gap esophageal atresia (LGEA).**

Patients and methods **Medical records of all patients managed for EA spectrum in our department between 2003 and 2012 were evaluated, retrospectively. Demographic data included the maternal age, the number of parities and deliveries, the presence of polyhydramnios and the prenatal diagnosis, sex, the gestational age and prematurity, the type of delivery and the birth weight, age at the time of diagnosis and treatment, the presence of associated anomalies including VACTERL-type and non-VACTERL-type anomalies, the type of EA according to Gross classification, and discrepancies between the diameters of atretic esophageal ends. SGEA and LGEA were defined as a gap of less than three vertebral bodies or three or more vertebral bodies in length between the atretic esophageal ends, respectively.**

Results **There were 99 patients treated for the diagnosis of EA spectrum during the study period: 81 in the SGEA group and 18 in the LGEA group. Most of the parameters studied did not differ between the two groups. Type-C EA was more prevalent in patients with SGEA ($n=77/81$) and type-A was more frequent in children with LGEA ($n=8/18$) ($P<0.05$). The frequency of prenatal diagnosis (2.5% for SGEA vs. 22.2% for LGEA) was more common in the LGEA group ($P<0.05$). Cesarean section compared with normal vaginal delivery was more commonly seen in both groups (56.8% for SGEA vs. 66.7% for LGEA).**

Conclusion **Most of the demographic parameters were similar between the two groups of patients. However, the frequency of prenatal diagnosis was more common in patients with LGEA. Most of the patients in both groups were delivered by a cesarean section. Type-C EA was more prevalent in patients with SGEA and type-A was more frequent in children with LGEA. Further analysis of this topic is warranted and may be beneficial in revealing the true demographic differences between patients with SGEA and LGEA.** *Ann Pediatr Surg* **10:107–111 © 2014 Annals of Pediatric Surgery.**

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Introduction

Described as an interruption of the continuity of the esophagus, esophageal atresia (EA) encompasses a group of congenital anomalies and occurs in association with tracheoesophageal fistula (TEF) or as an isolated entity [1]. The knowledge of the size of the gap between the atretic esophageal ends before surgery is of clinical importance and it is also a prognostic indicator of mortality and morbidity [2,3]. This clinical entity is divided into short-gap esophageal atresia (SGEA) and long-gap esophageal atresia (LGEA) depending on the distance between the atretic esophageal segments.

The distance between the esophageal ends, which constitutes SGEA and LGEA, lacks a strict numerical definition and there are no uniformly accepted criteria that define these entities. It has been reported that LGEA has gap intervals measuring greater than 2 cm, with a cutoff value of 2 cm or 2–3 vertebral bodies (VB) [4–6]. There are other suggested classifications in the literature including the nomenclature of short, intermediate, and long gap intervals with 1, 2.5, and 3 cm as the cutoff points, respectively [3,7]. A 3.5-cm 'ultralong' gap has also been described [8]. Epidemiological studies concerning SGEA and LGEA are limited. Thus, we conducted the present study to analyze the characteristics of patients with SGEA and LGEA and to provide valuable data on this issue. It was also aimed in the current study to compare the demographic characteristics between patients with SGEA and LGEA in the light of relevant literature.

Patients and methods

Institutional ethical approval for the study was obtained. Medical records of all patients managed for EA spectrum in our department between 2003 and 2012 were evaluated retrospectively. In defining the type of atresia, SGEA and LGEA were described as a gap of less than three VB or three or more VB in length between the atretic esophageal ends, respectively. In patients with a gastrostomy tube, by introducing a bougie into the upper pouch and another into the lower pouch through gastrostomy, the gap was evaluated by radiological means (Fig. 1). In patients who underwent thoracotomy for ligation of TEF, assessment of the gap between the atretic esophageal segments was performed during the surgical intervention (Fig. 2). Demographic data included maternal age, the number of parities and deliveries, the presence of polyhydramnios and the prenatal diagnosis,

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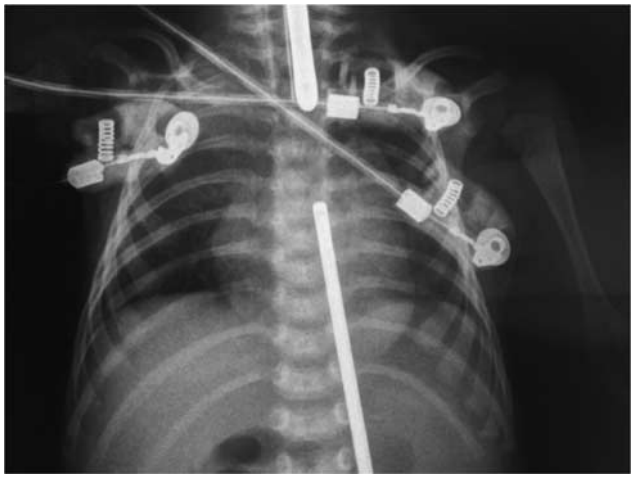
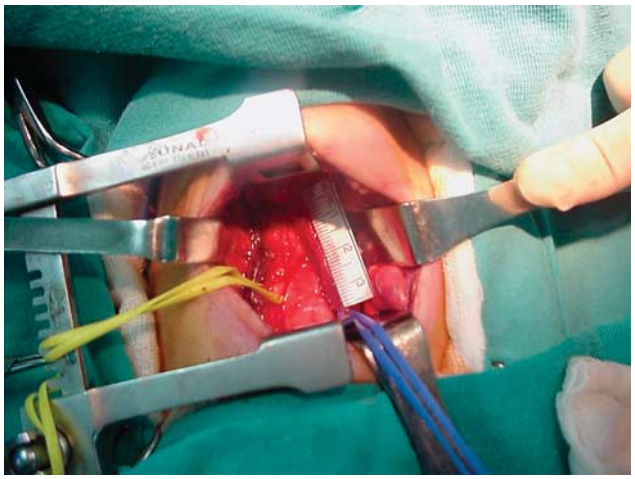
Fig. 1



Measurement of esophageal gap by radiologic examination in patients with gastrostomy. Note a metal bougie in the upper pouch and another in the lower pouch.

Fig. 2



Intraoperative assessment of the esophageal gap between the atretic esophageal segments in a child with esophageal atresia.

sex, the gestational age and prematurity, the type of delivery and the birth weight, age at the time of diagnosis and treatment, the presence of associated anomalies including VACTERL-type and non-VACTERL-type anomalies, the type of EA according to Gross classification, and discrepancies between the diameters of atretic esophageal ends. VACTERL association was defined if the patient had two or more anomalies of the vertebral, anorectal, cardiac (excluding patent ductus arteriosus, patent foramen ovale), renal/genitourinary, and limb systems.

Statistical analyses

Statistical analyses were performed using IBM SPSS (ver. 18; IBM Co., Armonk, New York, USA). Data are presented as mean ± SD, median (interquartile range) according to their distribution. Qualitative variables were expressed as percentages. Distribution of numerical data was assessed by graphical and statistical methods. Statistical analysis was performed with Student's *t*-test to compare numerical values with normal distribution. To compare numerical values with an abnormal dispersal range, interquartile ranges were assessed using the Mann–Whitney *U*-test. Fisher's exact test and χ^2-tests were used for the comparison of categorical data. A *P*-value of less than 0.05 was considered statistically significant.

Results

There were 99 patients treated for the diagnosis of EA spectrum during the study period, and the distribution of anatomical types of EA is shown in Table 1. Of the included patients, 81 were in the SGEA group and 18 were in the LGEA group. The characteristics of patients with SGEA and LGEA are depicted in Table 2. Most of the parameters studied did not differ between the two groups. Type-C EA ($n = 77/81$) was more prevalent in patients with SGEA and type-A ($n = 8/18$) was more frequent in children with LGEA ($P < 0.05$). The frequency of prenatal diagnosis (2.5% for SGEA vs. 22.2% for LGEA) was more common in the LGEA group ($P < 0.05$). Cesarean section was seen to occur more commonly than normal vaginal delivery in both groups (56.8% for SGEA vs. 66.7% for LGEA). Durations of ventilatory support (14.4 ± 2.5 vs. 36.0 ± 8.4 days; $P < 0.05$) and hospital stay (30.0 ± 3.8 vs. 69.7 ± 15.2 days; $P < 0.05$) were longer in the LGEA group. Patients with LGEA had a higher mortality rate (35.8 vs. 55.6%; $P < 0.05$).

Discussion

The rate of EA may be related to the maternal age, and in a study where maternal age was analyzed, both young (< 20 years) and older (> 35 years) mothers were shown to have an increased risk [9]. In a recent study, compared with children of mothers younger than 20 years, children of women giving birth at 35–40 years and above 40 years showed a two-fold and three-fold increased risk of EA, respectively [10]. In a most recent cohort from France, the median age of mothers giving birth to a newborn with EA at delivery was 30 years [11]. In another study, the mean ages of mothers of children with LGEA and SGEA were 30.1 and 28.9 years, respectively [12]. Average ages of mothers in both groups in the current study did not show a statistically significant difference, but as a whole, they were found to be lower compared with those reported previously [9,10,12]. This finding may be explained by true population differences including biologic and environmental factors concerning the ages of the mothers.

Table 1 Distribution of anatomical types of esophageal atresia

Anatomical types of esophageal atresia	*n*
Esophageal atresia with a distal tracheoesophageal fistula	87
Esophageal atresia without tracheoesophageal fistula	8
Tracheoesophageal fistula without esophageal atresia	3
Esophageal web	1

Table 2 Demographic data for patients with short-gap esophageal atresia versus long-gap esophageal atresia

Parameters	Short-gap EA	Long-gap EA	*P*
Maternal age (years)[a]	26.15 ± 5.85	25.17 ± 4.94	0.46
Number of parity[b] (range)	1 (1–4)	2 (1–4)	0.50
Number of deliveries[b] (range)	1 (1–4)	1 (1–4)	0.92
Polyhydramnios ratio	12/81 (14.8%)	5/18 (27.8%)	0.18
Prenatal diagnosis ratio	2/81 (2.5%)	4/18 (22.2%)	0.009
Gender (F/M)	41/40	7/11	0.36
Gestational age[a]	37.23 ± 2.64	36.0 ± 2.97	0.08
Prematurity	15/81 (18.5%)	5/18 (27.8%)	0.37
Type of delivery (NVD/CS)	35/46	6/12	0.44
Birth weight (g)[a]	2504.44 ± 651.16	2325.0 ± 636.14	0.29
Age at diagnosis (days)[b] (range)	1 (1–6)	1 (1–6)	0.07
Age at treatment (days)[b] (range)	3 (1–7)	3 (1–7)	0.30
Days on ventilatory support[b] (range)	14.45 ± 17.24 (0–100)	36.0 ± 31.6 (0–100)	0.002
Days of hospital stay (range)	30.03 ± 33.54 (3–270)	69.72 ± 64.28 (3–270)	0.001
Follow-up (months)[b] (range)	63.46 ± 38.2 (5–81)	48.29 ± 22.48 (5–81)	0.3
Presence of associated anomalies (non/NV/V)	20/8/53	3/1/14	0.59
Type of EA (Gross classification)	C ($n=77$)/non-C[c] ($n=4$)	C ($n=10$)/non-C[d] ($n=8$)	0.001
Discrepancy in diameters of atretic esophageal ends[b] (range)	2 (0–4)	2 (0–4)	0.85
Complications	52/81 (64.2%)	13/18 (72.2%)	0.185
Mortality	29/81 (35.8%)	10/18 (55.6%)	0.047

CS, cesarean section; EA, esophageal atresia; F, female; M, male; NV, non-VACTERL; NVD, normal vaginal delivery; V, VACTERL.
[a]Data are presented as mean ± SD.
[b]Data are presented as median (interquartile range).
[c]Class E + class web.
[d]Class A.

Parity may be an independent risk factor for birth defects. Associations between maternal parity, the number of deliveries, and birth defects have been observed previously: compared with primiparous mothers, nulliparous mothers were more likely to have infants with a number of congenital birth defects including EA [13]. Our series is dissimilar to this finding in that there were only one and three nulliparous mothers giving birth to patients with LGEA and SGEA, respectively. Although there is no definite explanation for this finding, variability in population compositions might have played a role. Although the mean number parities of mothers giving birth to children with LGEA was found to be higher than those of mothers with babies having SGEA, the median number of parities and deliveries in this study did not show statistical significance between the two groups. Research on the biologic or environmental factors that may be associated with the number of maternal parities and birth defects may be helpful in explaining some or all of these possible associations.

Polyhydramnios is an important clinical sign in the diagnosis of EA, but usually it is not apparent until the third trimester [14]. The rate of polyhydramnios in mothers who give birth to a newborn with EA ranges from 20 to 30% [14–16]. In a recent study, it was reported that EA associated with chromosomal or structural anomalies was associated with greater occurrence of polyhydramnios with a rate of 53% [17]. In a recent cohort comprising 307 new esophageal cases, polyhydramnios was present in 53.5% of the cases [18]. The 17.2% rate of polyhydramnios in the present study was lower than those reported previously [11,14–16]. This may be explained by the relatively low number of prenatally diagnosed cases included in our series than have been included in previously published series. Although not statistically significant, another finding in our series was that children with LGEA were associated with a greater occurrence of polyhydramnios than patients with SGEA (27.8 vs. 14.8%).

Prenatal diagnosis of EA before the third trimester is difficult and the prenatal detection rate of EA is low [18]. It was reported that prenatal detection was possible in 9.2 and 24% of cases, respectively [19,20]. In a recent report comprising 23 registries, it was found that prenatal detection rates of EA varied by registry from more than 50% to less than 10% of cases, and it was also claimed that the prenatal detection rate increased from 26 to 36.5% over the last two decades [21]. In the current study, the rate of prenatal diagnosis of LGEA was much higher than that of SGEA ($P < 0.05$), a finding similar to a recent report [11]. The ratio of 22.2% in the prenatal diagnosis regarding LGEA patients in our study is comparable to the literature, whereas that of patients with SGEA (2.5%) is lower than that reported [18,21]. We do not have an explanation for this finding, which demands more aggressive attempts to conduct prenatal diagnosis. As regular antenatal check-up programs increase, it is anticipated that the prenatal detection rates of EA will increase in the future.

The ratio of male to female was 1.06 in our study. In 15 EUROCAT registries covering 1 546 889 births for EA, 62% of the cases were male [22]. Other studies have reported either a small or no excess of male patients [9,23,24]. In a most recent report, the sex ratio (male/female) was found to be 1.3 [10]. We found a sex ratio of 1.57 for LGEA in favor of male patients, a result similar to that found in the literature [25]. In contrast, our finding of a sex ratio of 0.98 against male patients among children with SGEA is unlike that of Bianca and Ettore [25], who reported a ratio of 1.5 in favor of male patients. Although there is no definite explanation for this finding in the present study, variability of causes linked to genetics and the gene–environment interaction may be involved.

The current study did not show differences between the groups with regard to the gestational age and the age at diagnosis and treatment. This finding is consistent with that of the studies by Bagolan *et al*. [6]. In contrast, studies by Al-Shanafey and Harvey [12] and Lopes and Botelho [2] demonstrated that patients with LGEA were born at an earlier gestational age. In a most recent cohort, children with LGEA had a lower birth weight than children with SGEA [11]. Although children with LGEA had a lower birth weight and a high rate of prematurity compared with patients with SGEA in this study, we did not find any statistically significant difference between the groups, a finding unlike the study by Lopes and Botelho [2]. Dissimilarities in the present series concerning the birth weight and prematurity may be attributed to discrepancies pertaining to the demography of the study population.

Concerning the type of delivery in our study, most of the patients in both groups were delivered by cesarean section. This finding is consistent with that of the study by Chang *et al*. [15], who reported that 44 out of 72 babies with EA were born through cesarean section. In contrast, in a study by Aslanabadi *et al*. [26], it was found that patients with LGEA but not SGEA were delivered more often by a cesarean section. We speculate that materno-fetal factors might have been involved in the type of delivery in these patients. Further comprehensive investigations on the choice of the delivery type in these patients may give clear knowledge on this issue.

Full or partial VACTERL spectrum defects have been reported in 10–67% of the neonates with EA [27–32]. Similar to this range, we found VACTERL association in 67% of the children with EA in our study. The prevalence of non-VACTERL-type anomalies in patients with EA ranges from 20 to 70%, and our rate of 9% for non-VACTERL-type anomalies is lower compared with previous studies [27,33–35]. Our study revealed that the prevalence of associated anomalies including VACTERL type were comparable between the groups. This finding is consistent with that of the study by Lopes and Botelho [2]. Concerning the number of ribs, two of the patients in the LGEA group had 13 pairs of ribs, whereas none of those with SGEA had such a skeletal anomaly. Also, both groups in the current study showed similarities concerning the prevalence of associated non-VACTERL-type anomalies. This finding is different from that of study by Aslanabadi *et al*. [26], which reports more frequent non-VACTERL-type anomalies in patients with LGEA. Variations in our series may be due to genetic and geographic differences pertaining to our study population.

The other noticeable finding of this study regarding the type of EA according to the Gross classification was that type-C was more prevalent in patients with SGEA and type-A was more frequent in children with LGEA. Although there are conflicting reports, this finding has been considered as a description of LGEA by some authors in that there may be an association between the absence of TEF and LGEA [12]. Apart from the type of EA, both groups of patients did not show any difference with regard to the discrepancy in the diameter of atretic esophageal ends.

As may be expected, both the duration of ventilatory support and the length of hospital stay were found to be higher in children with LGEA compared with those of patients with SGEA in the current study ($P < 0.05$). In a previous study, the median length of hospital stay for patients with LGEA was found to be 83 days, a finding higher than that of ours [36]. In a recent cohort, the median durations of ventilation and of first hospital stay were 3 and *22* days, respectively [11]. Nevertheless, infants with LGEA spend a long time in hospital and need more time under ventilatory support compared with those with SGEA. Complications and long-term problems are frequently seen in patients with EA during treatment. The incidence of early and late complications during treatment and the follow-up duration did not differ between the two groups in the current study. Although of a limited time period, the follow-up duration in our study was comparable to that reported previously [14]. Long-term multidisciplinary follow-up seems to be beneficial in these patients [37,38].

Nevertheless, there are limitations in this report. The retrospective design and the limited number of patients could impact the validity of the data. Moreover, eight different surgeons managed our patients. This could bring up individual variations between the surgeons with different degrees of experience and technical skills. However, our discrimination of LGEA and SGEA was objective considering a gap of three VB as a cutoff value. We believe that using uniform criteria to measure the gap length in prospective studies involving more patients may allow us to compare results from different institutions. With this approach, it may be possible to shed light on this issue, providing fundamental information.

Conclusion

Most of the demographic parameters studied were similar between the two groups of patients in this series. However, the frequency of prenatal diagnosis was more common in patients with LGEA. Most of the patients in both groups were delivered by a cesarean section. Concerning the type of EA according to the Gross classification, type-C was more prevalent in patients with SGEA and type-A was more frequent in children with LGEA. The duration on ventilatory support and the hospital stay were higher in children with LGEA. Further prospectively designed population-based registries are necessary and may be beneficial in revealing the true demographic differences between patients with SGEA and LGEA.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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