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Subspecialization may improve an esophageal atresia service but has not addressed declining trainee experience☆,☆☆

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Abstract

Background: Subspecialization defined pediatric surgery using Alder Hey innovations in neonatal surgical units (Rickham) and anesthesia (Jackson-Rees). In neonatal surgery, United Kingdom subspecialization for cloacal extrophy and biliary atresia acknowledges their dependence on multidisciplinary management and the desirability of caseload for training. We phased in regional subspecialization for esophageal atresia (EA) repair and replacement surgery while trainee numbers increased nationally to reduce hours. We examined EA outcomes and training during subspecialization.

Methods: We analyzed EA cases (1999-2009) treated at Alder Hey Children's Hospital in two 5-year cohorts, the first early phase of incomplete subspecialization and the later near-total or "comprehensive" subspecialization phase. These periods approximated those before and after trainee numbers rose sharply to reduce working hours.

Results: Of 119 cases, 60 in the early cohort shared similar characteristics with the 59 in the later cohort. Near-complete subspecialization was achieved in the second 5 years with 97% of cases performed under a surgeon with an EA specialty interest; in the earlier cohort, 25% of surgeries were undertaken by surgeons without EA subspecialty interest. With near-complete subspecialization, pediatric intensive care unit stay fell from 5 (4-11) to 4 (2-7) days (median (IQR); $P = .005$), and approaches such as the Bianchi axillary repair and Bax single-stage jejunal interposition were introduced; hospital stay went from 25 (12-63) to 17 (13-28) days ($P = .27$), and deaths, from 6 to 3 children ($P = .49$). Mortality was 7.6% (9/119) compared with 14% (19/134) in our previous institutional series ($\chi^2 = 2.8$, $P = .09$), and neonatal mortality fell from 6% to 0 ($P = .008$). Near doubling of trainee numbers accompanied an approximately 3-fold fall in repairs per trainee over the study.

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☆☆ The study was registered with the Clinical Audit Department of Alder Hey Children's Hospital NHS Foundation Trust in September 2009 with Registration Number: 2248.

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Conclusion: Near-complete subspecialization for EA coincided with reduced pediatric intensive care unit stay, successful introduction of cosmetic axillary approaches, and extension of our replacement service to offer all interposition types. It has not reversed the steep decline in trainee experience of EA that has been associated with the greater numbers of trainees that have been employed to reduce working hours.

Surgery was recently described as "a profession adrift" because of limited quality research, inadequate audit of outcomes, and persistent preventable harms [1]. Similarly, quality improvement in surgery has been compared with "a lame horse" after revelations that approximately 80% of adverse outcomes go unreported by surgical trainees shouldering this responsibility [2]. In this context, pediatric surgery may be vulnerable because of low volumes of index cases and trainees' limited exposure to them.

Pediatric surgery arose from subspecialization to improve neonatal surgical outcomes [3,4]. Whether subspecialization within pediatric surgery can improve outcomes by increasing case volumes is debated [5-10]. In the United Kingdom, numbers of consultant pediatric surgeons and specialty trainees have risen sharply, in part to accommodate the European Working Time Directive (EWTD) [11,12]. This expanded workforce performs more general surgery of childhood, but neonatal caseload has not increased commensurately.

We describe our experience as perhaps the only United Kingdom specialist unit to phase in near-total subspecialization for esophageal atresia (EA). We anticipated that this would protect surgeon caseload and postulated that this could enhance outcomes and service development. At the same time, we increased our trainee numbers to meet the EWTD; by enhancing consultant case volume, we hypothesized that subspecialists would be more likely to take trainees through the case than an occasional EA surgeon under pressure to maintain their skills. This study analyzes our experience with this subspecialist mode of service delivery and the contemporaneous change in trainee experience.

1. Methods

1.1. Choice of time cohort

We analyzed data on children identified with EA between September 1, 1999, and August 31, 2009. This cohort follows a previous report of EA management in our unit from 1986 to 1997 when most or all consultant pediatric surgeons undertook repairs. Our study encompasses the phased introduction of subspecialist repair, approximated as an early phase (first 5 years, limited subspecialization) and a later phase (second 5 years, near complete subspecialization); EWTD restrictions were introduced over this period.

1.2. Patient selection

Cases were identified using codes from the World Health Organization's *International Classification of Disease Codes version 10* and the *Office of Population, Censuses and Surveys Classification of Surgical Operations and Procedures Version 4.4*.

Records were retrieved and reviewed by 2 authors (BC and WBJ). Cases that had initial EA management at Alder Hey were included in our analysis; any that had their primary surgical repair elsewhere were excluded. Operation notes, Picture Archiving and Communication System (PACS), and hospital information systems were used to complete our pro forma. Data were collected on demographics, gestational age, birth weight, antenatal problems, delivery, Apgar scores, associated anomalies, EA type, surgical procedures including operating surgeon, postoperative complications, and mortality. Data were transcribed into a PASW Statistics 18 (Chicago, IL) database.

1.3. Statistical analysis

The series was split into two 5-year ranges, September 9, 1999, to August 31, 2004, and September 1, 2004, to August 31, 2009, representing early and late subspecialization eras. Patient characteristics and the distribution of independent measurements were assessed using Q-Q plots and Shapiro-Wilk testing. Further statistical tests were conducted and reported accordingly. χ^2 tests, Fisher's Exact test, and independent-samples Mann-Whitney U tests were used as appropriate. All tests were 2 tailed unless stated otherwise. $P < .05$ was considered significant.

2. Results

We identified 119 cases in total: 60 in the early cohort and 59 in the later cohort. Demographic characteristics were similar between cohorts, as were the numbers of each EA type per Spitz category (Table 1) [13]. Vertebral, Anorectal, Cardiac, Tracheo-Esophageal, Renal and Limb (VACTERL) sequence was identified in 21 patients. The cardiovascular system was the commonest site for comorbidity, and anorectal malformation was the commonest associated gastrointestinal anomaly; 9 patients (7.6%) had a right-sided aortic arch.

In the early cohort, a quarter of cases (15/60) were performed by surgeons not identified as subspecialists; by

Table 1 Demographics and EA type by era

	Overall			Early			Late		
No, of patients	119			60			59		
No. of females	51			26			25		
Birth weight (g), mean (SD)	2581 (671)			2517 (668)			2648 (672)		
Gestational age (wk), median (IQR)	38 (36-40)			38 (36-40)			38 (36-39)		
Patients with cardiac malformations	57			29			28		
No. of VACTERL patients	21			8			13		
No. of patients with trisomy 21	4			4			0		
EA type (Gross) by Spitz category	I	II	III	I	II	III	I	II	III
A	5	5	0	1	5	0	4	0	0
B	1	0	0	1	0	0	0	0	0
C	69	29	1	32	15	0	37	14	1
D	1	1	0	1	0	0	0	1	0
H	6	1	0	5	0	0	1	1	0
Total	82	36	1	40	20	0	42	16	1

There were no differences in baseline characteristics: IQR indicates interquartile range (all $P > .05$); in this and the following table, the "early" cohort refers to the first 5-year era of incomplete subspecialization, the "late" cohort to the second 5-year era of near-total subspecialization. In this table and Table 3, anatomical EA types are classified according to the Gross classification, and their prognostic group, by the Spitz classification.

the second 5 years, 97% of EA surgery was performed by subspecialists ($\chi^2 = 11$, $P = .001$). In the later cohort, pediatric intensive care unit (PICU) stay was significantly reduced from a median (IQR) of 5 (4-11) to 4 (2-7) days; hospital stay also showed a similar trend changing from 25 (12-63) to 17 (13-28) days ($P = .27$) (Table 2).

Overall survival by Spitz category was 98% (I), 81% (II), and 100% (III) with no difference between early and late cohorts ($P = .235$, $P = 1.0$, and P not calculable for Spitz I, II, and III, respectively). Overall mortality was 7.6% (9/119) because of deaths at 3 to 12 months that were predominantly infection related (Table 3). Deaths halved from 6 (10%) of 60 in the early cohort to 3 (5.1%) of 59 in the later period ($P = .49$). Major leaks occurred in 4.5% of neonatal or delayed primary EA repairs with 4 of 5 requiring revision and no significant difference between cohorts (Table 2).

Over both cohorts, 100 patients had primary repair, 1 with isolated EA died before a definitive esophageal procedure and 4 had delayed primary repair (2 isolated EA, 1 EA with proximal tracheo-oesophageal atresia (TOF), and 1 long-gap EA with distal TEF (tracheo-esophageal fistula) that ultimately needed interposition). One required emergency fistula ligation. Five esophagostomies and 13 gastrostomies were constructed.

Since introduction of near-total subspecialization, 19% (11/59) of repairs were performed via the Bianchi axillary approach compared with none in the earlier cohort (Fig. 1) [14]. There were no deaths among those repaired using this approach. In one of these patients, a primary anastomosis could not be fashioned. The initial axillary approach did not preclude the use of a standard posterolateral thoracotomy for both the delayed primary repair and the subsequent salvage with colon interposition.

Nine EA patients (4 early cohort, 5 late cohort) had esophageal replacement finally using stomach (3), colon (4), or jejunum (2), with 1 late nonsurgical death overall and 1 case in each era requiring reoperation: the first required multiple surgeries, the other, a repair of leak after jejunal interposition. During near-total subspecialization, 3 jejunal interpositions were undertaken: 2 with prior ligation of mesojejunal vessels (gastric interposition preferred intra-operatively in 1) and 1 using the "Bax" method [15].

All repairs were either performed by, or supervised by, a consultant. The number of repairs performed by supervised trainees fell from 40% (24/60) to 24% (14/59) since near-

Table 2 Outcomes by subspecialization era

	Early	Late	
Anastomotic leak	3/55 (5.5%)	4/57 (7.0%)	$\chi^2 = 0.12$, $P = .73$
In primary repair	2/51 (3.9%)	3/53 (5.7%)	
In replacement	1/4 (25%)	1/5 (20%)	
TEF recurrence	3/54 (5.6%)	1/55 (1.8%)	$\chi^2 = 1.1$, $P = .30$
Dilatations	36 (60%)	34 (58%)	$\chi^2 = 0.069$, $P = .79$
5 or more	12 (20%)	9 (15%)	
Fundoplication	13 (22%)	7 (12%)	
Tracheomalacia	14 (23%)	8 (14%)	
Tracheostomy	5 (8.3%)	3 (5.1%)	
Aortopexy	2 (3.3%)	1 (1.7%)	
Deaths	6 (10%)	3 (5.1%)	$\chi^2 = 1.0$, $P = .31$
Related	1	0	
Unrelated	5	3	
Any complication	20 (33%)	13 (22%)	$\chi^2 = 1.9$, $P = .17$

Table 3 Summary of deaths

Type; Spitz	Management	Associated anomalies	Age and causes of death
C; II	Day 3, repair. Day 89, fundoplication, gastrostomy.	CHARGE; tetralogy; coloboma; choanalatresiae	Day 158, pneumonia.
C; II	Day 1, repair, pneumothorax. Day 32, aortopulmonary repair.	DA; subglottic stenosis; ASD; VSD; aortopulmonary window	Day 89, airway compromise, sepsis, and cerebral ischemia.
C; I	Day 1, repair.	Single left kidney	Day 166, meningococcemia
C; II	Day 2, repair. Day 15, leak, reanastomosis. Day 18, leak, esophagostomy, gastrostomy. Day 31, cholecystotomy. Day 32, retained swab removed.	ARM; left MCDK, hydronephrotic duplex right; DORV; subpulmonary stenosis; VSD	Day 145, endocarditis, liver failure.
A; II	Day 3, gastrostomy.	Laryngeal cleft; tracheomalacia; trisomy 21; AVSD; RVOTO	Day 107, pneumonia
C; I	Day 1, repair. Day 117, fundoplication and gastrostomy.	Duodenal atresia; ARM; asplenia; R aortic arch; bilateral VUR.	Day 170, sepsis.
C; II	Day 2, repair—tiny leak managed conservatively. Day 109, colectomy.	Pulmonary artery stenosis; tricuspid stenosis; ASD; VSD; NEC.	Day 114, NEC.
C; II	Day 1, fistula ligated, gastrostomy, DA repair and colostomy. Later LAARP. Day 272, gastric pull-up converted intraoperatively from jejunum.	Retrotracheal SCA, upper pouch at pharyngeal level, DA, ARM, single kidney, tethered cord, craniosynostosis. VACTERL.	Day 352, Klebsiella, pulmonary hemorrhage.
C; I	Day 2, repair. Day 63, aortopexy.	Giant exomphalos; large ASD; tracheomalacia	Day 107, pulmonary hemorrhage

ARM indicates anorectal malformation; ASD, atrial septal defect; AVSD, atrioventricular septal defect; DA, duodenal atresia; DORV, double outlet right ventricle; LAARP, laparoscopic-assisted anorectal pull-through; MCDK, multicystic dysplastic kidney; NEC, necrotizing enterocolitis; RVOTO, right ventricular outflow tract obstruction; SCA, subclavian artery; VSD, ventricular septal defect; VUR, vesicoureteric reflux.

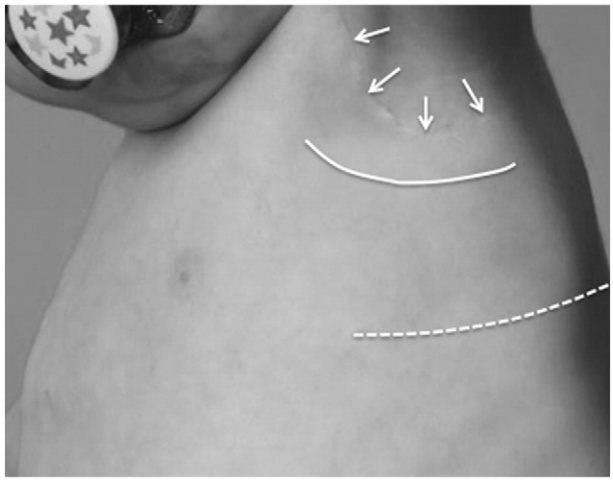

Fig. 1 Lateral view showing the healed incision (arrowed) after left axillary thoracotomy for EA repair in the presence of a preoperatively diagnosed right-sided aortic arch. Our incision is positioned higher and hence needs to be more curved than depicted by Bianchi in his original study (illustrated by the solid line) or the traditional approach (dashed line); however, it is in accordance with Bianchi's current recommendation. This left-sided version also emphasizes the incision's utility for example, for ligation of patent ductus arteriosus. Image used with parental permission and courtesy of the corresponding author.

total subspecialization was phased in ($\chi^2 = 3.623$, $P = .057$). However, trainee numbers increased from 5 at the start of the study to 9 at its end. Hence, repairs per trainee fell approximately 3-fold. Trainees performed 2 axillary repairs but no replacements.

3. Discussion

Historically, subspecialization was integral to development of pediatric surgery. Now, increasing surgeon numbers and limited neonatal caseloads prompted us to subspecialize within neonatal surgery. We postulated that preserving surgeon caseload would protect outcomes, support service development, and enhance training. Although near-total subspecialization for EA surgery coincided with reduced PICU stay and introduction of, for example, the Bianchi axillary repair and Bax single-stage jejunal interposition, we recorded an approximately 3-fold fall in repairs performed per trainee.

Mortality, leak and fistula rates were similar to contemporary series [16-20].The current mortality rate of 7.6% compares with a 14% mortality in our previous institutional series ($\chi^2 = 2.8$, $P = .09$) [21]. Compared with that series,

neonatal mortality fell from 6% (from cardiac defects) to 0 (P = .008). In the present series, deaths occurred mostly because of late sepsis. Further opportunities to reduce mortality therefore include intensified microbiological surveillance and early-warning strategies for when these patients leave high-dependency environments. Our greater frequency of balloon dilatations may reflect a low threshold for day care dilatations: our lower fundoplication rates indicate that recalcitrant strictures, that require protection from reflux, are less common.

There is continuing tension between obtaining adequate surgical experience and compliance with a limit on working hours. In neonatal surgery, with its low-volume practice, reduced operative experience may constitute a particular risk. However, the long working hours traditionally used to obtain such exposure have been associated with increased physician errors [22-24]. In our series, despite comprehensive subspecialization, trainees performed fewer repairs because of both the EWTD but also perhaps a changing recruitment pattern: in the past, a greater proportion of trainees were overseas graduates with sometimes considerable prior EA experience, and previous trainees had undergone longer basic surgical training with higher weekly hours.

Given good historical results, our study indicates that subspecialization for EA may have a limited benefit clinically and in terms of service development, but it has not ameliorated the decline in trainee experience. Although our findings indicate that subspecialization is not overtly detrimental, we acknowledge less tangible costs: consultants not performing EA repair can become deskilled, and this could be problematic when urgent fistula ligation is required. Furthermore, dependence on subspecialists for EA and lung surgery has, because of leave and sickness, required prolonged cover by single surgeons. Nevertheless, declining trainee exposure may mean subspecialization for EA surgery progresses by default: should the public accept new consultants doing occasional EA surgery after 2 or fewer in training?

Strategies to improve trainee experience include (1) rotation to overseas centers with bigger neonatal caseloads, for example, in India; (2) early demarcation of trainees wanting to pursue neonatal surgery from those content with children's general surgery alone (as in a recent United Kingdom survey); and (3) replacing pediatric surgery trainees on rotas with general surgery trainees that are newly mandated to obtain pediatric experience like their anesthetic equivalents. Involving general surgery trainees could reduce the drift of general surgery of childhood from district hospitals to specialist centers, while allowing the latter to furnish EWTD-compliant rotas with only 1 or 2 pediatric surgical trainees to share EA cases.

This is a retrospective study with consequent caveats: we cannot prove that subspecialization reduced PICU stay or similar nonsignificant trends in mortality. Given the latter's scarcity, the potential for an overlooked difference (type II error) has to be considered. Although subspecialization facilitated our adoption of different techniques, we recognize that it is not essential for initiating such developments. Further experience with subspecialized practice will also better determine the overall impact of such techniques.

In conclusion, subspecialization for EA surgery coincided with reduced PICU stay and introduction to our unit of different techniques, such as the axillary thoracotomy and single-stage jejunal interposition. Trainees' exposure to EA surgery appears to have declined sharply since the workforce expansion to meet the EWTD. Hence, future subspecialization within neonatal surgery may be driven less by outcomes and more to compensate for training limitations.

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References

[1] Horton R. Editorial: what is the point of surgery? Lancet 2010;376:1025.
[2] Dindo D, Hahnloser D, Clavien PA. Quality assessment in surgery: riding a lame horse. Ann Surg 2010;251:766-71.
[3] Forshall I, Rickham PP. Experience of a neonatal surgical unit - the 1st 6 years. Lancet 1960;2:751-4.
[4] Rickham PP. Neonatal surgery—early treatment of congenital malformations. Lancet 1952;262:332-9.
[5] Safford SD, Pietrobon R, Safford KM, et al. A study of 11,003 patients with hypertrophic pyloric stenosis and the association between surgeon and hospital volume and outcomes. J Pediatr Surg 2005;40:967-72.
[6] Dasgupta R, Kim PC. Relationship between surgical volume and clinical outcome: should pediatric surgeons be doing pancreaticoduodenectomies? J Pediatr Surg 2005;40:793-6.
[7] Grushka JR, Laberge JM, Puligandla P, et al. Effect of hospital case volume on outcome in congenital diaphragmatic hernia: the experience of the Canadian Pediatric Surgery Network. J Pediatr Surg 2009;44:873-6.
[8] Ly DP, Liao JG, Burd RS. Effect of surgeon and hospital characteristics on outcome after pyloromyotomy. Arch Surg 2005;140:1191-7.

[9] Bucher BT, Guth RM, Saito JM, et al. Impact of hospital volume on in-hospital mortality of infants undergoing repair of congenital diaphragmatic hernia. Ann Surg 2010;252:635-42.
[10] Gutierrez JC, Cheung MC, Zhuge Y, et al. Does children's oncology group hospital membership improve survival for patients with neuroblastoma or Wilms tumor? Pediatr Blood Cancer 2010;55:621-8.
[11] Children's Surgical Forum. General Paediatric Surgery: Survey of service provision in district general hospitals in England. London: The Royal College of Surgeons of England; 2010. http://www.rcseng.ac.uk/publications/docs/general-paediatric-surgery-service-provision-survey/attachment_download/pdffile.
[12] Skipworth RJE, Terrace JD, Fulton LA, et al. Basic surgical training in the era of the European Working Time Directive: what are the problems and solutions? Scott Med J 2008;53:18-21.
[13] Spitz L, Kiely EM, Morecroft JA, et al. Oesophageal atresia: at-risk groups for the 1990s. J Pediatr Surg 1994;29:723-5.
[14] Bianchi A, Sowande O, Alizai NK, et al. Aesthetics and lateral thoracotomy in the neonate. J Pediatr Surg 1998;33:1798-800.
[15] Bax KM. Jejunum for bridging long-gap esophageal atresia. Semin Pediatr Surg 2009;18:34-9.
[16] Lopez PJ, Keys C, Pierro A, et al. Oesophageal atresia: improved outcome in high-risk groups? J Pediatr Surg 2006;41:331-4.
[17] Mortell AE, Azizkhan RG. Esophageal atresia repair with thoracotomy: the Cincinnati contemporary experience. Semin Pediatr Surg 2009;18:12-9.
[18] Lilja HE, Wester T. Outcome in neonates with esophageal atresia treated over the last 20 years. Pediatr Surg Int 2008;24:531-6.
[19] Tönz M, Köhli S, Kaiser G. Oesophageal atresia: what has changed in the last 3 decades? Pediatr Surg Int 2004;20:768-72.
[20] Holcomb GW, Rothenberg SS, Bax KMA, et al. Thoracoscopic repair of esophageal atresia and tracheoesophageal fistula: a multi-institutional analysis. Ann Surg 2005;242:422-8.
[21] Driver CP, Shankar KR, Jones MO, et al. Phenotypic presentation and outcome of esophageal atresia in the era of the Spitz classification. J Pediatr Surg 2001;36:1419-21.
[22] Privette AR, Shackford SR, Osler T, et al. Implementation of resident work hour restrictions is associated with a reduction in mortality and provider-related complications on the surgical service: a concurrent analysis of 14,610 patients. Ann Surg 2009;250:316-21.
[23] Levine AC, Adusumilli J, Landrigan CP. Effects of reducing or eliminating resident work shifts over 16 hours: a systematic review. Sleep 2010;33:1043-53.
[24] Olson EJ, Drage LA, Auger RR. Sleep deprivation, physician performance, and patient safety. Chest 2009;136:1389-96.