Variations in the detection of anorectal anomalies at birth amongst European cities

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Contributors' statement:

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All authors approved the final manuscript as submitted and agreed to be accountable for all aspects of the work. The corresponding author attests that all listed authors meet authorship criteria and that no others meeting the criteria have been omitted.

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Abstract

Aim: The diagnosis of anorectal malformations (ARM) is made at birth by perineal examination of the newborn, yet small series reported late diagnosis in almost 13%[1]. No large series to date have looked into the magnitude of missed ARM cases in the neonatal period across Europe. This study aimed to define the rate of missed ARM at birth across four UK and EU centers.

Methods: All ARM cases treated at two UK tertiary centers in the past 15 years were compared to two tertiary European centers. Demographic and relevant clinical data were collected. Late diagnosis was defined as any diagnosis made after discharge from the birth unit. Factors associated with late diagnosis were explored with descriptive statistics.

Results: Across the four centers (117/1350, 8.7%) were sent home from the birth unit without recognizing the anorectal anomaly. Missed cases showed a slight female predominance (1.3:1), and the majority (113/117, 96.5%) were of the low anomaly with a fistula to the perineum. The rate of missed ARM cases was significantly higher in the UK centers combined (74/415, 17.8%) compared to those in the EU (43/935, 4.6%), ($p<0.00001$), and this was independent of individual center and year of birth.

Conclusion: Significant variation exist between the UK and other European countries in the detection of ARM at birth. We recommend raising the awareness of accurate perineal examination at time of newborn physical examination. We feel this highlights an urgent need for a national initiative to assess and address the timely diagnosis of ARM in the UK.

Key words: anorectal malformations, pediatric surgery, postnatal checks, congenital anomaly

Introduction

Anorectal malformations (ARM) constitute a varied group of congenital anomalies with an estimated incidence of one in every 4000 to 5000 newborns [2]. The diagnosis of an anorectal malformation is usually made at birth through an examination of the perineum of the newborn. However, missed diagnosis (i.e. after hospital discharge) has been reported even in adulthood, and accounting in some pediatric series for almost 13% of cases [1,3,4].

UK-based centres have reported that late referral of ARM cases from the birth centres (after 24h of age) accounted for up to 50% of cases [5,6], although few of the cases in either series had been discharged from hospital. Delays in diagnosis of ARM are associated with higher morbidity and mortality [5–8]. It may predispose to bowel perforation when not defunctioned in timely manner [9], and prolonged inadequate stooling will result in bowel dilatation, mega-rectum formation and will make further reconstruction challenging and more amenable for failure [10].

In the UK in 2006, the National Institute For Care and Health Excellence (NICE) introduced a clinical guideline for postnatal care for women and their babies until 8 weeks of life [11] . The guideline recommends a complete physical examination of the newborn baby within 72 hours of birth; it clearly states that "*the anus must be checked for completeness and patency*". However, unlike other European Countries, these guidelines do not specify who is responsible for the neonatal check. Perineal examination in a neonate can be challenging, particularly in females, and anal patency is often usually assumed by presence of meconium in the nappies rather than introduction of a probe in the anus [12,13].

To provide a measure of the rate of missed anorectal malformations (ARM) in the newborn period, in this study we review all new cases of anorectal malformation born in the UK and presenting to two pediatric surgery tertiary referral centres over a period of 15 years. The data were compared to 2 tertiary centres in Europe (which have different guidelines on postnatal

checks). We aim here to describe the incidence of late presenting ARM at two large-volume UK tertiary pediatric surgery centres, and to test whether the introduction of the NICE guidelines in 2006 in the UK has improved the rate of detection of ARM in the neonate.

Methods

Four tertiary pediatric surgery centres in four major European cities participated in this study. Two UK centres; Great Ormond Street Hospital (centre A) serves the pediatric population of Central/North London and Royal Manchester Children's Hospital (centre B) serves the pediatric population of the North West of England; between them, the two centres serve a population of over 9 million people. Each centre had previously independently audited the rate of missed ARM in their referred patient population. This study was registered as a re-audit between the two centres and appropriate approvals were obtained from the audit office in each hospital. Data from the two UK centres were compared to two tertiary referral centres in Europe; Necker Enfants Malades in Paris, France (centre C) and Bambino Gesù in Rome, Italy (centre D).

To determine the rate of missed anorectal malformation in the UK centres, all cases of infants presenting with ARM over a 15-year span between January 2002 and December 2016 (centre A) and 14-year span between January 2003 and December 2016 (centre B) were identified using medical coding and operating theatre records. We defined a missed diagnosis of ARM as a case in which a newborn baby was discharged home from the birth unit or referred to a pediatric surgery unit, having completed all neonatal checks, without identifying the anorectal malformation or being referred to a pediatric surgery unit. We excluded patients born outside the UK, those with cloacal anomalies and cases of anterior or stenotic anus (solitary or in Currarino triad) within a complete sphincter complex defined by sphincter mapping (examination under anaesthetic using Peña stimulator).

The variables collected for analysis included: patient demographics, mode of presentation, anatomical type of the ARM anomaly, timing of the first surgical intervention (as a surrogate for date of diagnosis which was not reliably recorded in this retrospective study), type of

surgical repair and presence of other VACTERL (vertebral defects, anal atresia, cardiac defects, tracheoesophageal fistula, renal anomalies, and limb abnormalities) anomalies. Binary logistic regression analysis (outcome variable = missed diagnosis) was performed on data from 2009-2016 using SPSS v24.0 and the following co-variates: gender, years from 2009, VACTERL (y/n), hospital (centre A/B) and high anomaly/low anomaly. Fisher's exact test was used to compare proportions. Data are presented as median and interquartile ranges or as numbers with proportion. A p value of $p<0.05$ was considered significant, except for the comparison of UK with European centres, where the Bonferroni corrected p value cut-off of $p<0.017$ (0.05/3) was used.

Results

Over the study period, 415 new cases of ARM were admitted to the two UK centres of which 74 (17.8%) overall (centre A: 26/192 [13.5%]; centre B: 48/223 [21.5%]) were missed at postnatal checks and discharged home. These patients had their first surgery at a median age of 159 days (range 2 days to 2.65 years of age, IQR 19.5-234 days). The number of missed cases and total cases year by year is shown in Figure 1A. Seventeen out of 141 cases (12.1%) were missed between 2002-2006 whereas 57/274 (20.8%) were missed between 2007-2016; thus, the incidence of missing ARM at birth has significantly increased across the two centres ($p=0.03$) despite the introduction of the NICE guidelines in 2006 (Figure 1B).

The male: female ratio in the missed group was 2:3 (29 boys and 45 girls); 72 (97%) had an external fistula (56 [76%] perineal fistula, 16 [22%] vestibular fistula) while 2 cases had an imperforate anus with recto-urethral fistula. In addition, 22 cases (30%) had at least one additional VACTERL association anomaly.

Data for the ARM patients who were diagnosed before hospital discharge was unavailable for the study cohort prior to 2009; we describe all 213 patients with ARM between 2009-2016 in Table 1. Of note, 44% of all perineal fistula patients were missed in this period. There was a significant difference in the proportion of missed cases when comparing low anomalies (vestibular + perineal) (47/138 [34%]) with high anomalies (bladder, prostatic, urethral, vaginal + no fistula) (2/75 [3%]) ($p<0.0001$) (Figure. 2A). Patients with a further VACTERL anomaly (14/108 [13%]) were significantly less likely to be missed than those with isolated ARM (35/105 [33.3%]) ($p=0.0006$) (Fig. 2B), this likely reflects the anatomy of the missed cases as VACTERL is more associated with high than low ARM anomalies as previously reported [14].

In order to examine factors associated with having a missed diagnosis, we performed a binary logistic regression analysis. The only factors associated with missed diagnosis were: low

anomaly (OR 37.0 [4.8-283.2]; p=0.001) and non-VACTERL (OR 3.2 [1.5-7.0]; p=0.003). There was no significant association between having a missed ARM and time (year of birth) (p=0.13), suggesting no improvement in detection of ARM over time from 2009-2016, noting the limited power of this analysis.

The operative management of the 2009-2016 patient cohort is displayed in Table 2. Those patients who had a missed diagnosis were more likely to be managed with a single stage surgery (primary anoplasty, trans-anal proctoplasty [TAP] and posterior sagittal anorectoplasty [PSARP]) than those whose anorectal malformation was diagnosed before discharge (38/49 vs. 93/213, p<0.0001). However, this probably reflects the anatomy of the defect rather than a different attitude towards surgical management of late diagnosed ARM cases. This is supported by the observation that in the low anomaly group (perineal and vestibular fistula), there was no difference in the proportion of single stage repair whether the diagnosis was made or missed prior to discharge from the birth centre (25/48 [52%] vs. 36/91 [40%], *p*=0.15).

In the two European centres, there were 935 new cases of ARM treated over the last 15 years; 43/935 (4.6%) were missed at birth (centre C: 34/696 [4.9%]; centre D: 9/239 [3.8%]). The age at presentation of those with missed diagnosis ranged between 6 days and 8.4 years and all except for two cases (41/43, 95%) were low anomalies with a fistula to the perineum; over half (23/43) of these cases were in males. During the study period the two UK centres had a significantly higher rate of missed anorectal anomalies compared to the two EU centres individually (74/415 [17.8%], 34/696 [4.9%], 9/239 [3.8%] p<0.0001) and as a whole (74/415 [17.8%], 43/935 [4.6%] p<0.00001) (Figure 3 A, B).

Discussion:

Delayed presentation of anorectal malformation remains significant in the NHS. Despite the introduction of a standardised postnatal care guideline by NICE in 2006 [11], we demonstrate here a significant increase in missed diagnosis of anorectal malformation in recent years. Two equivalent European centres are referred a much lower proportion of missed diagnosis, suggesting more effective detection within the immediate postnatal period.

Currently, all babies born in the UK will, ideally, have a systematic examination prior to discharge home or within 72 hours of birth. This newborn physical examination is commonly known as the "baby check" and is performed by trainee pediatricians or by midwifes who completed an Examination of the Newborn accredited course. Historically, the effectiveness of neonatal examinations in detecting birth defects has been questioned in both the UK[15] and overseas [16], and specific data on cataracts has highlighted that a substantial proportion of children with congenital and infantile cataract are not diagnosed by 3 months of age [17].

In this series from two UK centres, we encountered 74 cases of missed ARM, defined as diagnosis after discharge from the birth center. Clinical examination was sufficient to diagnose the anomaly in all missed cases. Delays in diagnosis resulted in surgery being performed outside the neonatal period in a significant proportion of patients, even in childhood in some cases. As one might expect, the missed cases were mostly low-type ARM where an external fistula allows passage of meconium, perhaps suggesting anal patency to the examining professional. We observed milder phenotype in those patients that were diagnosed late, evidenced by the fact that most late diagnoses were amenable to single stage surgery as opposed to requiring a colostomy while awaiting definitive repair.

In order to provide a subjective estimate of the truly missed anorectal anomalies at birth, we excluded in this study cases of anterior anus and anal stenosis which are challenging to diagnose especially in the early neonatal period. In addition, we did not observe any increase

in the total numbers of ARM cases treated at the two UK centres overtime to suggest over treatment of milder cases of ARM anomalies.

Our study represents the largest reported series of late diagnosed ARM cases; and, including only UK-born patients, it is indicative of the delivery of newborn care within the NHS over the past 15 years. This longitudinal data set spans a period where routine baby checks were initially guided by the 1989 RCPCH report "Health for All Children", its 2003 revision (commonly referred to as Hall 4) [18], as well as the 2006 NICE guidelines. More recently, the NHS Newborn and Infant Physical Examination Screening Programme (NIPE) is replacing previous recommendations to ensure a consistence service across all health care providers in England [19] , and how this change will affect the detection of ARM at birth is still to be seen.

One of the limitations of this study is that differences in the health care systems and referral patterns exist between the UK and the studied EU centers. This might have partially influenced the rate of missed cases amongst the centers making the comparison more complex to interpret. In addition, as a retrospective review of referred patients, this study is unable to give as much information as a full epidemiological study. The awaited report from BAPS-CASS (British Association of Paediatric Surgeons – Clinical Anomalies Surveillance System) describing UK nationwide incidence and spectrum of anorectal malformation cases in a calendar year will provide more insight into the nationwide incidence of ARM and the associated missed diagnosis rate as well as delineating those delays due to diagnosis, referral or access.

Although we do not report any serious morbidity or mortality associated with delayed diagnosis in our cohort, other centres report delayed diagnosis to be associated with perforation in 10% of cases [9], and a mortality of approximately 4% [5,6,10]. Moreover, this study was not designed to look at the long-term morbidities such as constipation, incontinence,

urinary problems which can be associated with ARM and could potentially increase in the group of patients with a delayed diagnosis.

Our data presented here indicate that there are 2 possible levels of problems. The first relates to the postnatal care of infants and guidelines on baby checks; contrary to our studied European Countries, babies are often discharged very early after birth (hours), and the newborn examination is therefore rarely performed by a neonatologist. In fact, according to NHS guidelines "The health professional doing the examination could be a doctor, midwife, nurse or health visitor who has been trained to do the examination" [20]. Nevertheless, the perineal examination of a newborn is challenging, even in experienced hands, and our data suggest that the current training pathway for the health professionals currently performing neonatal discharge examination maybe inadequate. One interim solution to overcome this problem is to provide photographic documentation as part of the infant newborn examination. This could be accessed remotely by a paediatric surgeon if required and would prevent unnecessary travel and displacement of families to see a paediatric surgeon. Secondly, in France and Italy, as elsewhere in Europe, primary care for infants and children is performed by a pediatrician rather than a general practitioner. Our data indicate that a third of children with missed diagnosis of ARM may present after 6 months of age, suggesting potential difficulties in forming a diagnosis in the constipated child within the UK primary care setting, where broadly trained general practitioners may lack awareness of rare congenital anomalies to detect the more subtle variants, further delaying a definitive diagnosis.

We believe action is needed to improve the rate of neonatal detection of ARM, similarly to actions improving the quality of hip examination which have improved DDH (developmental dysplasia of the Hip) detection in the UK [21,22]. There is a need to focus on improving the quality of training provided to health care professionals performing routine newborn examinations to avoid the consequences of missing a major congenital anomaly.

Conclusion:

This study of two UK centres highlights a significant issue in the timely diagnosis of anorectal anomalies. Two comparable European centres have a significantly lower rate of missed diagnosis, thus there is a need to improve the newborn detection rate in the UK in order to avoid morbidity and mortality.

Figure Legends:

Figure 1. A: Cases year by year. B: Percentage of cases that were missed before (2002-2006) and after (2007-2016) the NICE guidelines, compared using Fisher's Exact test

Figure 2. A: Distribution of missed cases between high and low ARM. B: Distribution of missed cases in isolated ARM and VACTERL ARM.

Figure 3. Percentage of cases study period in the two UK centers compared to the two EU centers individually (A) or as a whole (B).

Table 1. 2009-2016 ARM missed cases by anatomical type of malformation

Table 2. 2009-2016 Operative management by type of malformation, missed cases vs. not missed. (Single Stage included patients in whom a covering stoma was formed during the definitive surgery)

Supplementary Table 1

Binary logistic regression analysis examining the association between missed diagnosis (dependent variable) and gender, year, VACTERL anomaly, hospital and high anomaly.

References

1. Kim HL, Gow KW, Penner JG, Blair GK, Murphy JJ, Webber EM. Presentation of low anorectal malformations beyond the neonatal period. *Pediatrics*. 2000;105(5):E68. http://www.ncbi.nlm.nih.gov/pubmed/10799632. Accessed November 10, 2017.

2. Levitt MA, Peña A. IMPERFORATE ANUS AND CLOACAL MALFORMATIONS. In: Ostlie DJ, ed. *Ashcraft's Pediatric Surgery*. 5th ed. Elsevier; 2010:468-490. doi:10.1016/B978-1-4160-6127-4.00036-7.

3. Jonker JE, Trzpis M, Broens PMA. Underdiagnosis of Mild Congenital Anorectal Malformations. *J Pediatr*. 2017;186:101-104.e1. doi:10.1016/j.jpeds.2017.03.054.

4. Govender S, Wiersma R. Delayed diagnosis of anorectal malformations (ARM): causes and consequences in a resource-constrained environment. *Pediatr Surg Int*. 2016;32(4):369-375. doi:10.1007/s00383-016-3866-5.

5. Lindley RM, Shawis RN, Roberts JP. Delays in the diagnosis of anorectal malformations are common and significantly increase serious early complications. *Acta Paediatr*. 2006;95(3):364-368. doi:10.1080/08035250500437523.

6. Haider N, Fisher R. Mortality and morbidity associated with late diagnosis of anorectal malformations in children. *Surgeon*. 2007;5(6):327-330. http://www.ncbi.nlm.nih.gov/pubmed/18080605. Accessed November 10, 2017.

7. Tareen F, Coyle D, Aworanti OM, Gillick J. Delayed diagnosis of anorectal malformation--a persistent problem. *Ir Med J*. 2013;106(8):238-240. http://www.ncbi.nlm.nih.gov/pubmed/24282893. Accessed December 4, 2018.

8. Govender S, Wiersma R. Delayed diagnosis of anorectal malformations (ARM):

causes and consequences in a resource-constrained environment. *Pediatr Surg Int*. 2016;32(4):369-375. doi:10.1007/s00383-016-3866-5.

9. Turowski C, Dingemann J, Gillick J. Delayed diagnosis of imperforate anus: an unacceptable morbidity. *Pediatr Surg Int*. 2010;26(11):1083-1086. doi:10.1007/s00383-010-2691-5.

10. Eltayeb AA. Delayed presentation of anorectal malformations: the possible associated morbidity and mortality. *Pediatr Surg Int*. 2010;26(8):801-806. doi:10.1007/s00383-010-2641-2.

11. CG37. Postnatal care up to 8 weeks after birth | Guidance and guidelines | NICE. Clinical guideline [CG37]. https://www.nice.org.uk/guidance/cg37. Published 2006. Accessed November 10, 2017.

12. Aldeiri B, Johal NS, De Coppi P. Meconium is not enough: look for the hole! *Case Reports*. 2012;2012(dec18 1):bcr2012007456-bcr2012007456. doi:10.1136/bcr-2012-007456.

13. Karakus SC, Kilincaslan H, Sarsu SB, et al. The passage of meconium alone is not a sign of correctly positioned anus. *J Matern Neonatal Med*. 2015;28(3):303-305. doi:10.3109/14767058.2014.916267.

14. Totonelli G, Catania VD, Morini F, et al. VACTERL association in anorectal malformation: effect on the outcome. *Pediatr Surg Int*. 2015;31(9):805-808. doi:10.1007/s00383-015-3745-5.

15. Hall DM. The role of the routine neonatal examination. *BMJ*. 1999;318(7184):619-620. http://www.ncbi.nlm.nih.gov/pubmed/10066183. Accessed April 24, 2018.

16. Wilson BE, Etheridge CE, Soundappan SV, Holland AJ. Delayed diagnosis of anorectal malformations: are current guidelines sufficient? *J Paediatr Child Health*. 2010;46(5):268-272. doi:10.1111/j.1440-1754.2009.01683.x.

17. Rahi JS, Dezateux C. National cross sectional study of detection of congenital and infantile cataract in the United Kingdom: role of childhood screening and surveillance. The British Congenital Cataract Interest Group. *BMJ*. 1999;318(7180):362-365. http://www.ncbi.nlm.nih.gov/pubmed/9933197. Accessed April 24, 2018.

18. Hall DMB, Elliman D. *Health for All Children*. Oxford University Press; 2006. doi:10.1093/med/9780198570844.001.0001.

19. Department of Health. Newborn and infant physical examination screening: standards - GOV.UK. https://www.gov.uk/government/publications/newborn-and-infant-physical-examination-screening-standards. Published 2018. Accessed December 1, 2018.

20. NHS Choices. Newborn physical examination - NHS.UK. web info. https://www.nhs.uk/conditions/pregnancy-and-baby/newborn-physical-exam/. Published 2018. Accessed April 24, 2018.

21. Godward S, Dezateux C. Surgery for congenital dislocation of the hip in the UK as a measure of outcome of screening. MRC Working Party on Congenital Dislocation of the Hip. Medical Research Council. *Lancet (London, England)*. 1998;351(9110):1149-1152. http://www.ncbi.nlm.nih.gov/pubmed/9643684. Accessed May 9, 2018.

22. McAllister DA, Morling JR, Fischbacher CM, Reidy M, Murray A, Wood R. Enhanced detection services for developmental dysplasia of the hip in Scottish children, 1997-2013. *Arch Dis Child*. February 2018:archdischild-2017-314354.

343 doi:10.1136/archdischild-2017-314354.

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Figure 1.

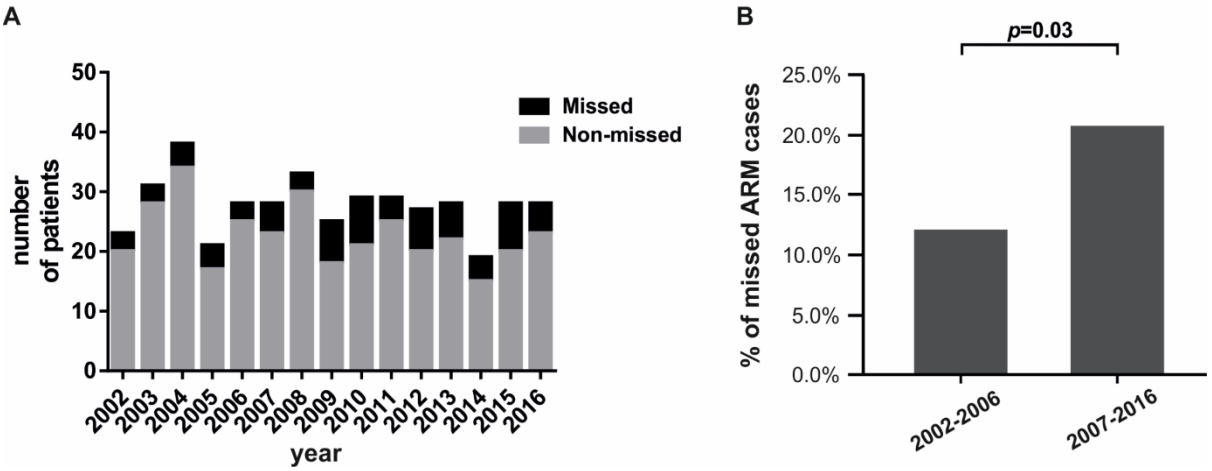
A
number of patients
year
Missed
Non-missed
0
10
20
30
40
50
2002
2003
2004
2005
2006
2007
2008
2009
2010
2011
2012
2013
2014
2015
2016
B
p=0.03
% of missed ARM cases
0.0%
5.0%
10.0%
15.0%
20.0%
25.0%
2002-2006
2007-2016

Figure 2.

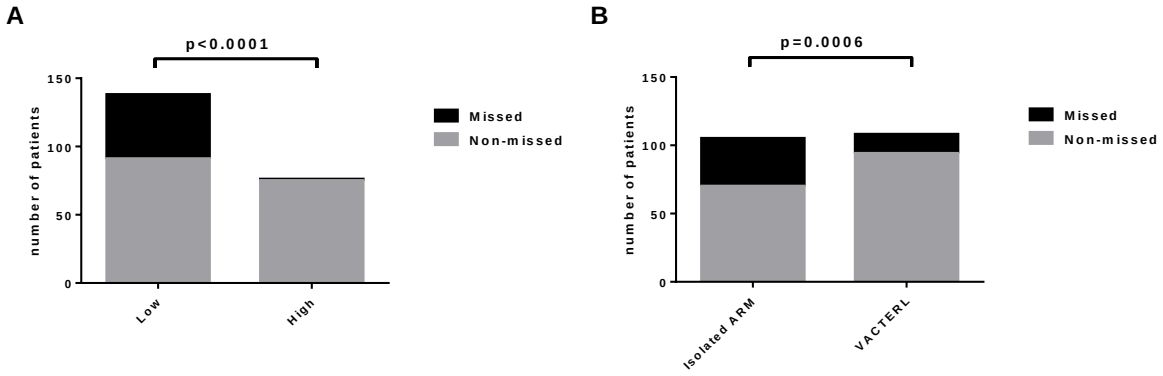
A
p<0.0001
number of patients
150
100
50
0
Low
High
Missed
Non-missed
B
p=0.0006
number of patients
150
100
50
0
Isolated ARM
VACTERL
Missed
Non-missed

Figure 3.

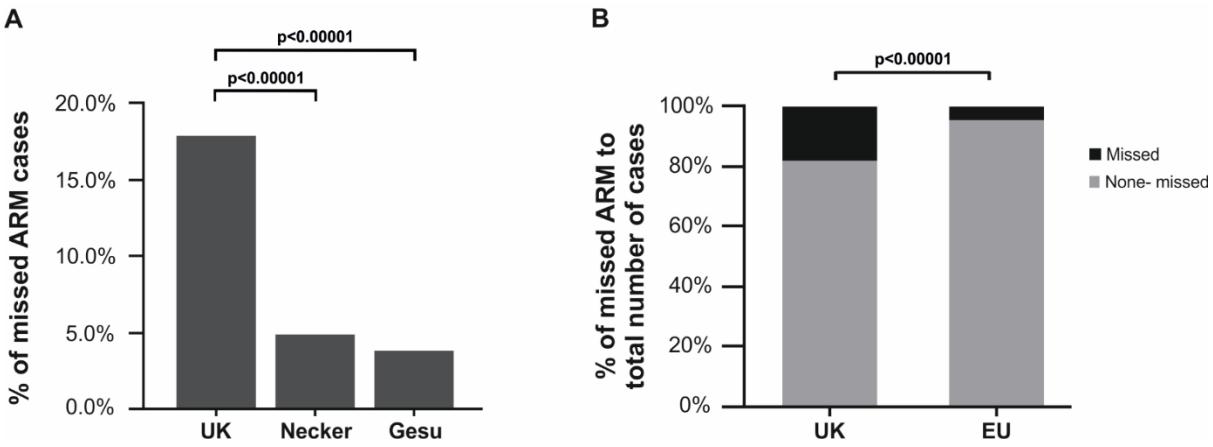
A
p<0.00001
p<0.00001
% of missed ARM cases
20.0%
15.0%
10.0%
5.0%
0.0%
UK
Necker
Gesu
B
p<0.00001
% of missed ARM to total number of cases
100%
80%
60%
40%
20%
0%
UK
EU
Missed
None- missed

Table 1.

	total	missed	proportion
No fistula	14	0	0
Bladder	13	0	0
Perineal	94	41	43.6%
Prostatic urethra	6	0	0
Vaginal	9	0	0
Urethral	33	1	3.0%
Vestibular	44	7	15.9%
Total	213	49	23.0%

Table 2.

ARM Type	Stoma			Primary Repair		
	total	missed	%	total	missed	%
No fistula	12	0	0.0	2	0	0.0
Bladder	12	0	0.0	1	0	0.0
Perineal	32	8	25.0	62	33	53.2
Prostatic urethra	5	0	0.0	1	0	0.0
Vaginal	5	0	0.0	4	0	0.0
Urethral	33	1	3.1	0	0	-
Vestibular	21	2	9.5	23	5	21.7
Total	**120**	**11**	**91.6**	**93**	**38**	**40.9**

Supplementary Table 1

Variable	Exp B (95% CI)	P value
Gender	1.1 (0.5-2.3)	0.875
Year	1.1 (1.0-1.3)	0.125
VACTERL Y/N	3.2 (1.5-7.0)	0.003
Hospital	0.7 (0.3-1.6)	0.444
High anomaly	37.0 (4.8-283.2)	0.001