

The National Incidence and Outcomes of Gastroschisis Repairs

Abstract:

MJ Barrett^{1,2,3}, O Kozdoba¹, N Al Assaf⁴, J Gillick^{5,6}, A Mortell^{5,6}, A Foran¹

¹Department of Neonatology, Children's University Hospital, Temple St, Dublin 1

²Department of Paediatrics, UCD, Belfield, Dublin 4

³National Children's Research Centre, ⁴Departments of Neonatology and ⁵Paediatric Surgery, Our Lady's Children's Hospital, Crumlin, Dublin 12

⁶Departments of Paediatric Surgery, Children's University Hospital, Temple St, Dublin 1

Abstract

The birth prevalence of gastroschisis worldwide has increased over the past decades. We aim to determine the Irish national incidence of gastroschisis repairs (NIGR) over a 5 year period (2007- 2011) and clinical outcomes by a retrospective cohort review of cases admitted to all Irish paediatric surgical units. Seventy patients were identified. The NIGR per 10,000 live births was 1.96 (SD 0.51) per year. Fifty eight (82%) were antenatally detected. Twenty eight (40%) had primary repair day 1 with the remaining repaired in a median of 3(2-5.75) days. Thirty three (47%) experienced a central catheter related infection. Duration of stay was significantly correlated with decreasing gestational age ($p=0.016$), decreasing birthweight ($p=0.005$), increasing numbers of blood transfusions ($p<0.001$) and co-morbidity or complication ($p<0.001$). This study provides individual centres with patient outcomes and national data that can be provided to parents and clinical staff regarding the clinical course of gastroschisis.

Introduction

The birth prevalence of gastroschisis worldwide has increased steadily over the past few decades, with considerable financial and resource implications for healthcare providers.¹ In 2007, an Irish study by Murphy et al reported 53 gastroschisis repairs from 1998 to 2004, which translates to an average rate of 1.3 per 10,000 live births.² The overall antenatal detection rate in Ireland from 1998 to 2004 for anterior abdominal wall defects is reported as 44%.² Antenatal screening with ultrasound has a high success rate in the detection of anterior abdominal wall defects with successful diagnosis reported in up to 86-90%.³ Advances in neonatal intensive care and the development of parenteral nutrition have reduced infant mortality rates for gastroschisis from 60% in the 1960s to 3-10% in the mid-1990s with no clear evidence of a reduction in mortality since then.^{4,7} Nationally and internationally surgical strategies for gastroschisis have evolved on limited evidence and no consensus on the optimal initial management or an effective clinical pathway.^{1, 2}

Recent studies have stratified neonates with gastroschisis into simple and complex groups which reliably predicts outcome at one year.^{1,5,8} Our aim was to establish the national incidence of gastroschisis repairs (NIGR) in Ireland over a 5 year period (2007- 2011). We describe maternal and infant demographics with clinical course and outcomes of gastroschisis admissions.

Methods

A dual site retrospective cohort review of neonates admitted for gastroschisis repair in the period 2007-2011 to Irish tertiary level paediatric hospitals was performed. Patients were identified using theatre records, the Hospital Inpatient Enquiry System (HIPE) and were cross-referenced with ICU datasets and patient case notes. All data collection was processed by two reviewers and transferred to an electronic database. All data queries were dealt with by consensus. All data originates from medical paper records and databases. Measurements from a Gaussian distribution were compared using means and 2-tailed Pearson's correlation upon meeting assumptions for parametric tests. Non-Gaussian distributions were compared using median (interquartile ranges), Mann Whitney test and 2-tailed Spearman's correlation. Binomials were analysed using Chi Squared test. Data was analysed using SPSS 18.1. Significance was defined as $p\leq 0.05$

Results

In the period 2007-2011 a total of 70 infants had gastroschisis repairs. The NIGR per 10,000 live births is 1.96 (0.51) over the study period. Infants were born as late preterm infants with the median gestational age at 36(2) weeks. (Table 1) Forty one (58%) mothers were 25 years old or younger and 44 (62%) primigravid. Eleven (15%) infants born were considered small for gestational age (SGA). The antenatal detection rate for this cohort was at 83%. Seven (7/58) infants with an antenatal diagnosis were postnatal transfers to the tertiary surgical centres from outside the Dublin maternity hospitals. Four children had concurrent cardiac abnormalities (patent ductus arteriosus, atrial septal defect, mild aortic stenosis, and aortic incompetence) and none required cardiothoracic intervention during their initial admission. Three children had concurrent renal abnormalities (hydronephrosis, dysplasia and vesicoureteric reflux). A single child had a syndrome phenotype which has no associated genetic abnormality. No abnormal karyotypes were detected.

The caesarean section rate was at 48% (34/70). To further subdivide this figure into emergent and elective section we find that the elective rate stood at almost 13%. Caesarean section was only significantly associated with lower 1 minute ($p=0.02$) and 5 minute ($p=0.04$) Apgars scores. The median duration of time for all gastroschisis repairs was 2 days. Further subdividing this group we identified 28(40%) whom had primary repair (PR) day one with the remaining patients (NPR) repaired in a median of 3(2-5.75) days. The median length of stay in the PICU was 7(5-11) days. (Table 2) A significant difference in the duration of stay in the PICU between the PR and NPR groups ($p=0.017$) exists.

For the entire cohort median duration of total parenteral nutrition (TPN) was 20 days with the median length of stay at 31 days from admission to the hospital. There was an observed trend of longer duration of TPN in the NPR group however this was not statistically significant. Just under half of patients (47%) experienced a central catheter related infection (CCRIs) which was equally distributed between PR and NPR groups. There was no significant difference between PR and NPR groups for number of transfusions or duration of stay. Analysis of the entire cohort reveals significant correlations between the duration of hospital stay and gestational age (Pearson's $p=0.016$), birth weight (Pearson's $p=0.005$), duration of TPN (Spearman's $p<0.001$), number of transfusions (Spearman's $p<0.001$) and co-morbidity or complication (Spearman's $p<0.001$). Decreasing gestational age and birth weight correlates to a longer duration of hospital admission. Increasing duration of TPN therapy, number of blood transfusions and co-morbidity or complication correlates to a longer admission. Concurrent bowel atresia was noted in 7 (10%) patients. Symptoms and findings consistent with necrotising enterocolitis (NEC) occurred in 12 (17%) patients. A single death occurred due to overwhelming septicemia and NEC. TPN related adverse events occurred in 5 patients.

Discussion

Gastroschisis is challenging as its causes (a complex biomedical and sociocultural set of risk factors and developmental origin) are largely speculative or unknown. From a public health perspective it appears to disproportionately target children of young mothers and it is increasing in many countries worldwide. This study reveals that the NIGR is 1.96 per 10,000 live births during the study period 2007-2011.² This corresponds to EUROCAT 2006-2010 registry prevalence rates for Gastroschisis of 2.23, 2.05 and 1.82 per 10,000 from Cork and Kerry, Dublin, and South East Ireland respectively.¹⁰ The antenatal detection rate of this condition is at 83% which is significantly higher than previously reported from Ireland.²

This may be due to a multitude of factors including the improved local antenatal screening programs, despite the lack

of a uniform national antenatal screening program. The postnatal benefits of prenatal diagnosis of gastroschisis include family awareness, adequate planning of delivery with alerted paediatric staff, optimal risk categorisation and a personalised protocol for action. The caesarean section rate is almost 49%. To further subdivide this figure into emergent and elective section we find that the elective rate stands at almost 13%. No studies have definitively shown the benefit of routine caesarean section for delivery.¹¹ In the entire cohort, there was a significant correlation between gestational age, birth weight, length of PICU stay, and duration of TPN, number of blood transfusions and co-morbidity or complication with the duration of hospital admission. Risk categorisation allows more informative parental counselling and planning regarding expected potential complications, mortality rate, length of hospitalisation and cost.⁸ The identification of co-factors related to relevant patient outcomes, such as CCRIs, and amenable to improvement reveals future targets for improvement in patient care, outcomes and costs.^{12, 13}

This national cohort compares favourably with other national cohorts.^{1, 2} Unfortunately our retrospective study, being subject to the flaws of such research, did not lend itself to collection of data upon suspected risks factors (due to the absence of uniform data documentation in clinical notes) such as smoking or concomitant illicit drug usage. Therefore a multivariate analysis of risk factors was not undertaken. Early neonatal deaths (albeit rare) at sites of referral could not be included. The strength of this study is its robust data collection of a national cohort over 5 years. This nationally representative study provides a benchmark against which individual centres can compare outcomes and performance. This study provides data which can be provided to parents regarding the clinical course spectrum in relation to gastroschisis repairs. This information may help clinical staff to optimise antenatal delivery, postnatal and tertiary care plans or to define targets upon which improvement is sought. We recommend further local prospective studies with international collaborations to elucidate the risk factors, management strategies and outcomes.

Correspondence: MJ Barrett
Royal Children's Hospital, Melbourne, Australia
Email: mjbarrett@hotmail.com

References

1. Bradnock TJ, Marven S, Owen A, Johnson P, Kurinczuk JJ, Spark P, Draper ES, Knight M, Baps C. Gastroschisis: one year outcomes from national cohort study. *BMJ*. 2011;343:d6749.
2. Murphy FL, Mazlan TA, Tarheen F, Corbally MT, Puri P. Gastroschisis and exomphalos in Ireland 1998-2004. Does antenatal diagnosis impact on outcome? *Pediatr Surg Int*. 2007;23:1059-63.
3. Salihi HM, Boos R, Schmidt W. Omphalocele and gastrochisis. *J Obstet Gynaecol*. 2002;22:489-92.
4. Knutrud O, Bjordal RI, Ro J, Bo G. Gastroschisis and omphalocele. *Prog Pediatr Surg*. 1979;13:51-61.
5. Chang DC, Salazar-Osuna JH, Choo SS, Arnold MA, Colombani PM, Abdullah F. Benchmarking the quality of care of infants with low-risk gastroschisis using a novel risk stratification index. *Surgery*. 2010;147:766-71.
6. Nembhard WN, Waller DK, Sever LE, Canfield MA. Patterns of first-year survival among infants with selected congenital anomalies in Texas, 1995-1997. *Teratology*. 2001;64:267-75.
7. Weinsheimer RL, Yanchar NL, Bouchard SB, Kim PK, Laberge JM, Skarsgard ED, Lee SK, McMillan D, von Dadelszen P. Gastroschisis closure--does method really matter? *J Pediatr Surg*. 2008;43:874-8.
8. Molik KA, Gingalewski CA, West KW, Rescorla FJ, Scherer LR, Engum SA, Grosfeld JL. Gastroschisis: a plea for risk categorization. *J Pediatr Surg*. 2001;36:51-5.
9. McDonnell R, Delany V, Dack P, Johnson H. Changing trend in congenital abdominal wall defects in eastern region of Ireland. *Ir Med J*. 2002;95:236, 8.
10. EUROCAT Gastroschisis (per 10,000 births) for the following registries: All Registries, last 5 years [database on the Internet]. [cited 2/1/2013].
11. Segel SY, Marder SJ, Parry S, Macones GA. Fetal abdominal wall defects and mode of delivery: a systematic review. *Obstetrics & Gynecology*. 2001;98:867-73.
12. Longmate AG, Ellis KS, Boyle L, Maher S, Cairns CJS, Lloyd SM, Lang C. Elimination of central-venous-catheter-related bloodstream infections from the intensive care unit. *BMJ Quality & Safety*. 2011;20:174-80.
13. Keys C, Drewett M, Burge DM. Gastroschisis: the cost of an epidemic. *Journal of pediatric surgery*. 2008;43:654-7.