Slovenská zdravotnícka univerzita v Bratislave, 2020, roč. 16, č. 1, s. 1-44. Dostupné na internete: http://casopis.fvszu.sk/ ISSN 1337-1789

RISK FACTORS CONTRIBUTING TO DEATH VERSUS SURVIVAL OF INFANTS WITH A CONGENITAL ANOMALY: A POPULATION-BASED COHORT STUDY OF WELSH LIVEBIRTHS USING DE-IDENTIFIED LINKED DATA BETWEEN 1998 AND 2017

P Ho, MA Quigley, JJ Kurinczuk

National Perinatal Epidemiology Unit, Nuffield Department of Population Health, University of Oxford

Email of corresponding author: Peter.ho@gtc.ox.ac.uk
ICBDSR Registry on which the work is based: Congenital Anomaly Register and Information Service (CARIS), Wales, UK

Background and Objectives: Congenital anomalies are the second leading cause of infant death in the UK, accounting for over one-third of all infant deaths. To date, population-based studies of risk factors for mortality of infants with congenital anomaly have been limited. Estimates of infant mortality associated with congenital anomalies are important to inform planning of healthcare and social interventions aimed at reducing infant mortality. Congenital Anomaly Register and Information Service (CARIS) is a high-quality, population-based register established in 1998 to identify congenital anomalies in all regions of Wales and is known to have high reporting rates due to a robust surveillance system. CARIS collects data about congenital anomalies involving structural, metabolic or genetic defects, present before the end of pregnancy. The aim of this study was to investigate the nature, clinical features, and risk factors associated with the risk of infant death versus survival in infants with congenital anomalies.

Methods: A population-based cohort study was conducted using registry data from CARIS with linkage to data on livebirths and deaths from the UK Office for National Statistics. The study population included all livebirths with birthweight ≥500g, gestational age ≥22+0 weeks, and a diagnosis of a congenital anomaly between 1998 and 2017. The characteristics explored were socio-demographic, maternal, infant, and intervention factors. Univariable logistic regression was performed to obtain crude odds ratios; those with $p<0.1$ were explored in a multivariable model. Multivariable logistic regression was performed using a backward stepwise approach to obtain adjusted odds ratios, and a subsequent likelihood ratio test to examine the independent effect of each significant variable; $p<0.05$ was considered significant in the final model. The analysis was performed separately for: infants with any anomaly; those with isolated anomalies; those with multiple anomalies; and those with cardiovascular anomalies.

Results: Overall, 30,424 livebirths affected by congenital anomalies were identified, of which 20,008 (65.8%) had isolated anomalies and 10,374 (34.1%) had multiple anomalies. Cardiovascular anomalies contributed the highest infant mortality rate (IMR: 1.33 per 10,000 livebirths) among isolated anomalies. Significant increases in adjusted odds of infant mortality were found among infants born to mothers of non-White vs White ethnicity (aOR 2.25; 95% CI: 1.77-2.86); parous vs nulliparous (aOR 1.24; 1.08-1.41); active smokers during pregnancy vs non-/ex- smokers (aOR 1.20, 1.02-1.40); preterm vs term (aOR 4.38; 3.86-4.98); and female vs male infants (aOR 1.28; 1.13-1.46). Infants who required surgery in the first year of life had a lower odds of infant mortality than those who did not (aOR 0.80; 0.68-0.95). Preterm birth was a significant risk factor for all anomaly types but the effect of the other characteristics varied according to the anomaly group.

Discussion and Conclusions: Socio-demographic, maternal, infant, and intervention factors have a significant impact on infant mortality in babies with congenital anomalies by different potential mechanisms depending on the type of anomaly. For example, the excess mortality from congenital anomalies in minority ethnic groups may be due to an interplay of factors resulting in unequal access to antenatal screening and medical and surgical interventions, different attitudes toward congenital anomalies and termination of pregnancy, and consanguinity. Likely smoking-related mechanisms include placental abruption and an increased risk of sudden infant death syndrome. Prematurity increases the risk of comorbidities and related complications. Infants with an anomaly who required surgery in the first year of life had a lower risk of death compared to those who did not or whose anomalies were too severe for surgery.

Ho, P – Quigley, MA – Kurinczuk, JJ. Risk factors contributing to death versus survival of infants with a congenital anomaly: A population-based cohort study of Welsh livebirths using de-identified linked data between 1998 and 2017, s .14.