

Epidemiology of isolated cleft palate in Wales

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Background

- Cleft lip and/or palate is the most common craniofacial malformation in the newborn.
- Can be defined as isolated, non-isolated and syndromic

Importance of isolated cleft palate



- Isolated cleft palate is more common than isolated cleft lip and is harder to diagnose in the antenatal period
- Infant feeding difficulties and recurrent ear infections
- Implications for the development of speech
- Multidisciplinary care required
- Mean secondary care cost ranges from £6137.49 to £17,004.09 from birth to age 10.¹

1. Secondary care costs to the NHS for children born with unilateral cleft lip and palate, bilateral cleft lip and palate and cleft palate from birth to 10 years of age. Hasanally, S. A. (Author). 26 Nov 2020

Aims

- Describe the epidemiology of isolated cleft palate in Wales
- Investigate historical concern of higher rates of cleft palate in North Wales

Methods

- Population based retrospective cohort study. Utilising routinely collected prospective data.
- Surveillance data from the Congenital Anomaly Register and Information Service (CARIS) for Wales.
- Data available covered births from 1998 to 2021.
- Data sources include hospital data (PEDW) and notifications from health care professionals.
- Verification of data via Welsh Clinical Portal

Exposures and outcomes

- 754 cases
- Patient characteristics
 - Birthweight, gestation at birth
- Maternal characteristics
 - Postcode of residence, regular medication
- Malformation characteristics
 - Isolated, genetic, other malformations present

Statistical tools

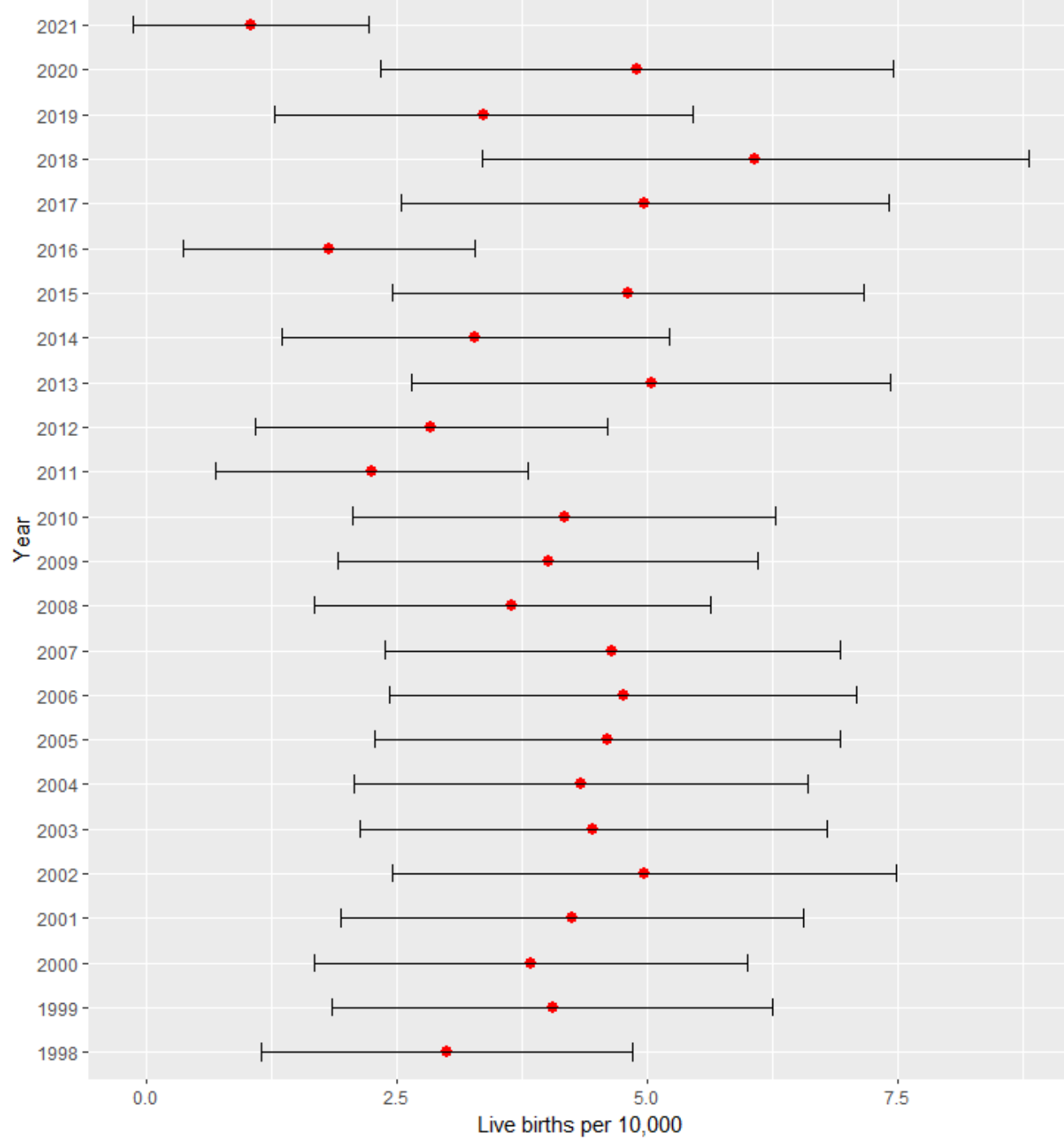
- Analysis performed on R 2022.02.1 Build 461
 - Packages used include dplyr, tidyr, ggplot2, icd, performanceanalytics
- Chi-squared test (deprivation proportions)
- Spearman's Rank Correlation Coefficient (various)

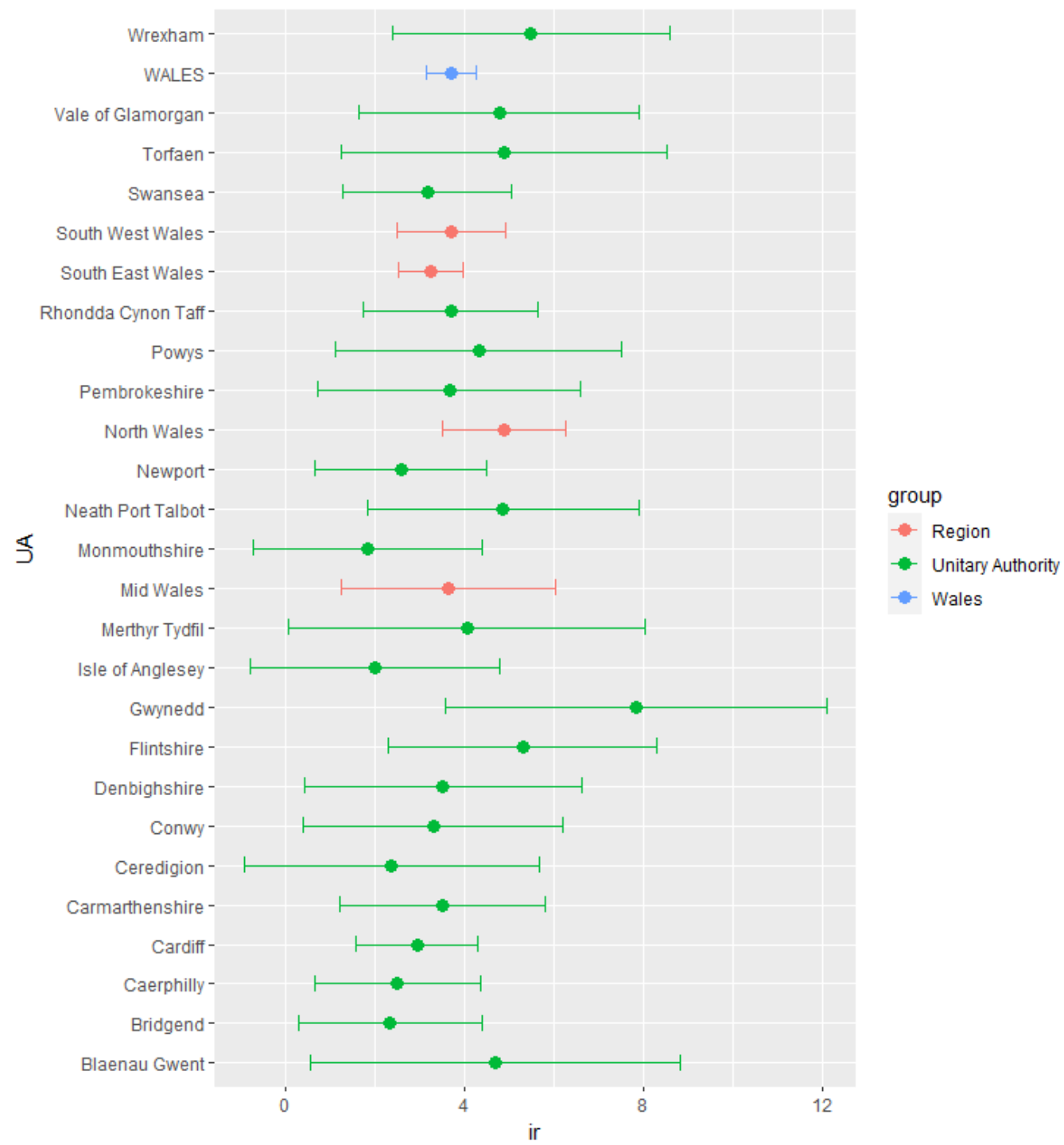
Results

- Isolated cleft: 310
- Isolated cleft with minor anomalies: 38
- Isolated cleft and multiple anomalies in the same system: 84
- Multiple anomalies in different systems: 159
- Chromosomal: 99
- Genetic: 59
- Teratogenic: 5

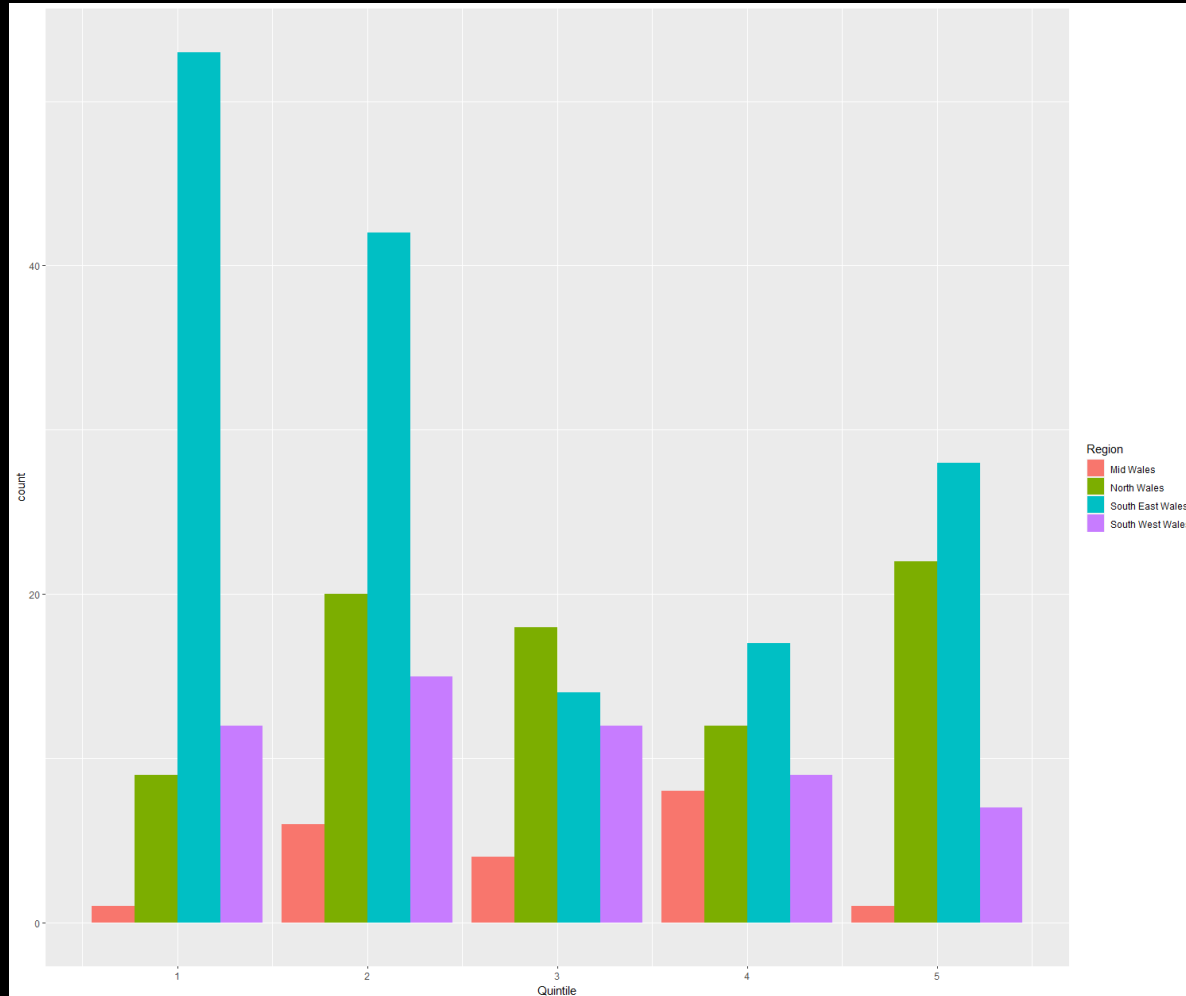
Results

Variable	Value
Sex	43.2% Male
Average birthweight	3377.9 grams
Live births	98%
Mean gestation	39.5 weeks
Average maternal age	28.8 years





Deprivation



- Pearson's Chi-squared test
 - North Wales v South East Wales
- $X^2 = 20.407$
- Degrees of freedom = 4
- p-value < 0.0005

Spearman's rank correlation coefficients

- Birthweight / Deprivation: 0.11 ($p < 0.05$)
- Maternal age / Deprivation: 0.24 ($p < 0.001$)
- Smoking / Deprivation: 0.21 ($p < 0.001$)
 - No statistically significant relationship for drugs and alcohol
- Consanguinity / Migrant status 0.7 ($p < 0.001$)

Review

- Strengths
 - Completeness of cases
 - Limited recall bias
- Limitations
 - Variations in terminology over time
 - Non-homogeneous definitions
 - Missing/incomplete data e.g.
 - Confounders in exposure data

Recommendations

- Separate analysis including isolated cleft lip and palate
- Analysis of cases syndromic/chromosomal aetiology
- Data completeness e.g. Folic acid
- Larger analysis with all UK and/or European data (e.g. EUROCAT data)

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