Patients with Congenital Adrenal Hyperplasia have significantly higher healthcare utilisation than the general paediatric population.

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Introduction

Congenital adrenal hyperplasia (CAH) is the commonest cause of adrenal insufficiency in the paediatric population. Patients have multiple health problems and require specialist endocrine input. Although quality of life , health and stature are known to be impacted by CAH, to our knowledge healthcare utilisation has not previously been assessed in paediatric CAH.

Methodology

Data source

Data were from the Clinical Practice Research Datalink (CPRD), a longitudinal, anonymized research database derived from nearly 700 primary-care practices in the UK. For approximately 50% of these practices, linked secondary-care data are available from the English Hospital Episode Statistics (HES). Linked Office for National Statistics (ONS) data were used for cause of death.

Study design

Patients with a diagnosis of CAH in the primary-care or linked HES data and at least one prescription for a corticosteroid were selected.

CAH patients were matched 1:10 to a randomly drawn control group without CAH on gender, age, general practice, eligibility for HES linkage, and registration status at the CAH patient's index date (defined as the later of their first CAH record and registration date).

Resource use and corresponding financial costs were estimated, using the published NHS tariffs and unit costs of health and social care from the Personal Social Services Research Unit (PSSRU) for general practice contacts, outpatient appointments, and inpatient admissions and compared using chi square test with the matched control population.

Incidence of depression and mortality were examined for **all ages including adults**. Risk of death was compared using a Cox proportional hazards model. As QOL has previously been noted to be impaired in CAH, depression was identified by diagnosis or antidepressant prescription, and compared by chi-square test to the control population.

Results

605 subjects, of which 562 could be matched, had a record of at least one corticosteroid prescription and formed the total CAH study population. Of these 255 (132 female, mean age 5.1yrs (SD 5.0), mean CAH duration 2.5 yrs(SD 3.9)) were aged less than 18 years at time of data entry and were matched to 2550 controls. Of these, 84 cases and 840 controls were eligible for linkage to HES data. Paediatric data for resource use was examined in two age cohorts 0-<7 years and 7-<18years.

For the death analysis, 270 CAH patients could be matched with controls, were eligible for linkage to ONS data and had an index date within the ONS follow-up period.

Healthcare utilisation

In both age cohorts healthcare usage by CAH patients was significantly higher across primary and secondary care, resulting in a significantly increased cost (p<0.001) (see table).

Healthcare Utilisation and Cost by CAH patients and Controls

Events/year	CAH patients aged 0- <7years n=51		Controls aged 0- <7 years n=510		
	Median	IQR	Median	IQR	P value
Primary Care Attendance	9.0	(6.0–17.0)	4.0	(2.0–8.0)	<0.001
Hospital Admission	2.0	(1.0–3.0)	0	(0-0)	<0.001
Outpatient Attendance	8.0	(5.0-13.0)	0	(0-1.0)	<0.001
Total Healthcare Cost (£)	3,185	(1,860–5,187)	700	(465–1,482)	<0.001
Events/year	CAH patients aged 7- <18 years n=33		Controls aged 7- <18 years n=330		
	Median	IQR	Median	IQR	P value
Primary Care Attendance	4.0	(3.0–7.0)	2.0	(1.0–3.0)	<0.001
Hospital Admission	1.0	(0.0–2.0)	0.0	(0.0–0.0)	<0.001
Outpatient Attendance	5.0	(4.0–7.0)	0.0	(0.0–1.0)	<0.001
Total Healthcare Cost (£)	2,183	(1,173–2,850)	491	(290–951)	<0.001

Death

There were 17 deaths in the 270 CAH patients and 41 in the 2,700 controls. The calculated hazard ratio adjusted for age, sex and comorbid conditions (Charlson Comorbidity index) was 5.17 (CI 2.81-9.50).

Depression

188 of 562 CAH patients with data had at least one episode of depression; a lifetime prevalence of 33.5% compared with a 1,467 of 5620 control patients with a lifetime prevalence of 26.1%. (p<0.001)

Limitations

We have used a widely recognised, large, cross-sectional NHS source from the UK for this analysis. There have been reports that coding was not always accurate in the early days of establishing such databases. This would have tended to lead to underreporting of patients with CAH. Costs captured in this analysis may not be complete - for example, the costs of providing specially manufactured paediatric formulations of hydrocortisone are not always clearly captured in the NHS tariff.

Conclusions

Paediatric patients with CAH have significant healthcare needs and access primary and secondary care significantly more than their peers. This results in a significantly elevated cost to healthcare providers.

Despite advances in care, mortality risk is still over 5 times higher in patients with CAH and lifetime depression is increased 28% over the general population attending primary care.

Patients with CAH require improved treatments and care pathways to address the inequalities in outcomes seen.

References

CPRD   https://www.cprd.com/
HES   http://digital.nhs.uk/hes
ONS   https://www.ons.gov.uk/
PSSRU   www.pssru.ac.uk/