School absence and achievement in children with isolated orofacial clefts

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ABSTRACT

Objectives To examine school absence and academic achievement among 7-year-old children with isolated orofacial clefts in England.

Design Analysis of educational data linked to national cleft registry and administrative hospital data.

Setting English state schools.

Patients 3523 children with isolated clefts aged 7 years between 2006 and 2014.

Main outcome measures Annual school absence and reaching the national ‘expected level’ according to teacher-assessed academic achievement.

Results Children with isolated clefts had higher mean annual school absence (10.5 days) than their peers in the national population (8.9 days). Total absence was higher in children with a cleft lip and palate (CLP; 11.3 days) or with a cleft palate only (CPO; 10.5 days) than in children with a cleft lip only (CLO; 9.5 days). The percentage reaching the expected academic level decreased with increasing school absence (from 77.4% (923/1192) with annual school absence ≤5 days to 43.4% (193/445) with annual school absence >20 days). However, differences in school absence did not explain that children with CPO (65.9% reaching expected level) or CLP (66.1% reaching expected level) had poorer levels of academic achievement than children with CLO (73.5% reaching expected level). Children with a cleft were twice as often recognised as having special education needs (40.5%) than their peers (21.6%).

Conclusions School absence and cleft type are both independently associated with school attainment at 7 years. Children with an isolated cleft, especially when the palate is involved, and those with high levels of school absence may benefit from increased support addressing their educational needs.

INTRODUCTION

Around 1 in 700 children are born alive with an orofacial cleft, which may affect only the lip (CLO), only the palate (CPO) or both (CLP). A cleft can affect hearing, language, dental health, psychosocial health, neurodevelopmental health and academic outcomes.

Recently, we have shown that the presence of an isolated cleft (a cleft without additional anomalies or syndromes) negatively affects all areas of learning, especially when the cleft involves the palate (both CPO and CLP). These findings reflect the educational attainment gap in children born with an orofacial cleft, even when treated under modern conditions and supported by a coordinated multidisciplinary team approach.

What is already known on this topic?

Children with clefts tend to have poorer educational outcomes than unaffected children, and outcomes can vary according to cleft type.

What this study adds?

Compared with the general population in England, school absence is higher among 7-year-old children born with a cleft and varies according to cleft type.

What this study adds?

Appointments related to healthcare are primarily contributing to differences in absence between isolated cleft types.

Differences in school absence do not explain that children with a cleft affecting the palate have lower attainment than children with a cleft lip alone.

A study conducted in Western Australia compared school absence rates and academic achievement in 310 children with an isolated cleft and 1722 unaffected children varying in age between 6 and 15 years. This relatively small study found that children with isolated CPO had poorer academic achievement even when school absence levels were considered. However, it found no significant attainment gap for CLO and CLP pupils, in contrast to our previous study of children in England.
METHODS
Study cohort, databases and record linkage
We conducted a cross-sectional study using three national datasets, linked at individual level based on name, date of birth, postcode and National Health Service (NHS) number. The study cohort was identified in the Cleft Registry and Audit Network (CRANE) database. A total of 5976 children born with a cleft in England who were in year 2 of the state primary school system in England (around 7 years old), between 1 September 2006 and 31 August 2014, were identified. Of these, 5117 were eligible for inclusion in the study as their parents had given consent for information to be linked to other records (online supplementary file 1).

There were 4759 children who could be linked to the Hospital Episode Statistics (HES) database, which contains administrative records on all admissions to the NHS, the state-funded healthcare system that treats more than 95% of children born with a cleft in England. A total of 4586 children with CRANE-HES records could also be linked to the National Pupil Database (NPD), which holds individual-level information on educational outcomes, school attendance, special educational needs (SEN) and some sociodemographic characteristics for all children attending state schools in England. Absence data were missing for seven children who were excluded from analyses.

Linkage to HES was used to exclude 1056 (23.0%) children who had additional anomalies or syndromes (online supplementary file 2). The remaining 3523 children were considered to have an isolated cleft and were included in the study.

Patient characteristics
Cleft type was determined by LAHSAL code, obtained from CRANE, and categorised into three groups (from least to most anatomically involved cleft type): CLO, CPO and CLP. The NPD records provided the Income Deprivation Affecting Children Index, an index of socioeconomic deprivation that is calculated for small areas (mean population about 1500). Eligibility for free school meals was whether English was the first language and whether SEN had been recognised, which can relate to physical, cognitive, sensory and behavioural difficulties.

Absence outcomes
The NPD requires schools to report every pupil absence (one session equals half a day) and to state whether absence was ‘authorised’ or ‘unauthorised’. Authorised absence includes absence because of ‘illness’ or ‘medical/dental appointments’. Unauthorised absence includes absence without a justifiable reason, such as arriving late for school or family holidays not approved by the school. For comparison, we provided mean state-school absence levels for the general population in year 2 between 2010 and 2014. We report annual absence levels, categorised into five groups that correspond to school weeks: 0–5 days, 5.5–10 days, 10.5–15 days, 15.5–20 days and >20 days.

Educational outcomes
We analysed teacher-assessed achievement levels across five subject areas: speaking/listening, reading, writing, mathematics and science, which were completed towards the end of year 2 (the ‘Key Stage 1’ assessment). While the NPD shows whether a child meets the ‘expected level’ for each subject area, we created a composite outcome to determine whether children achieved the expected level across all five subject areas.

Statistical analysis
Means and 95% CIs were calculated to describe annual absence levels. A one-sample t-test was used to compare absence levels in the entire cohort against point estimates for the national population. A one-way analysis of variance was used to compare absence levels between cleft types.

Proportions describing achievement levels were compared with χ² tests. Logistic regression was used to compare these proportions and to estimate ORs and 95% CIs with and without adjustment for socioeconomic deprivation, free school meal eligibility, sex, cleft type and absence. We also included a test for linear trend to assess the impact of annual absence levels on academic achievement by including categories of school absence numbered sequentially (1–5) in the regression model.

Post hoc pairwise tests were carried out with Bonferroni correction for multiple comparisons when a statistically significant overall difference was detected between cleft types to establish between which pair significant differences existed. A p value <0.05 was considered to indicate a statistically significant result. Analyses were conducted using Stata V.15.

Ethics
The study is exempt from ethics approval as it involves the analysis of data that are collected for the purpose of service evaluation.

RESULTS
Characteristics of study cohort
Table 1 presents the characteristics of 3523 children with an isolated orofacial cleft and the characteristics of their peers in the national population. Isolated clefts are known to be more common in boys, which explains the higher proportion of boys in the study cohort. The proportion of children with SEN varied according to cleft type (26.7% with CLO, 42.8% with CPO and 47.9% with CLP).

In England, there is a staged approach to providing SEN support. Of the 1426 children receiving SEN support, 36.9% had first stage support, 51.1% had second stage support and 12.1% had final stage support. Only the 900 children with second or final stage support (63.1% of those with SEN) had their primary and secondary SEN type reported. Of these, 70.2% were recorded as having speech, language and communication needs (SLCN), which was the most common SEN among the cohort. All types of SEN were higher in the children with an isolated cleft than in the national population. The greatest differences were observed with SLCN and hearing impairment.

School absence
Out of the included children, 3383 (96.0%) missed at least one school session. Table 2 shows that, on average, overall annual school absence in children with a cleft was greater than in the general population. Children with a cleft missed more school for illness and for medical/dental appointments but not for unauthorised reasons.

Children with a more anatomically involved cleft type missed significantly more school for medical/dental appointments (table 2). As a result, the overall annual absence increased from 9.5 days (95% CI 8.8 to 10.1) in children with a CLO to 11.3 days (95% CI 10.7 to 11.9) in children with a CLP. Post hoc tests confirmed that mean annual medical/dental absence was different between each of the three possible pairwise comparisons of cleft types (p value always <0.001).
School absence and academic achievement

Academic achievement was poorer in children with a cleft if overall annual school absence was longer (figure 1). For all cleft types, the highest proportion of children achieving the expected academic level or above across all five subjects (77.4%) was seen in the children who were absent 5 days or less, and this proportion gradually decreased with increasing overall school absence (43.4% in children who were absent more than 20 days; test for linear trend p<0.001; table 3). This inverse relationship between the length of school absence and academic achievement was similar for all five subject areas (figure 2). Reading and writing were the subjects most affected by absence.

Adjustment for cleft type and additional adjustment for sociodemographic characteristics had little impact on the ORs reflecting the differences in academic achievement according to overall annual school absence (table 3).

Cleft type and academic achievement

The proportion of children achieving the expected level or above was lower in children with CPO (65.9%) and with CLP (66.1%) than in children with CLO (73.5%; table 3). The proportion of children achieving the expected level or above decreased with increasing overall school absence, and this dose–response relationship was similar for each cleft type (figure 1).

The ORs reflecting the differences in academic achievement according to cleft type changed only slightly with adjustment for school absence and with additional adjustment for sociodemographic characteristics (table 3). Post hoc tests confirmed that with adjustment for school absence, sex and socioeconomic characteristics, the proportion of children achieving the expected academic level differed between children with CLO and CPO (p value <0.001) and between children with CLO and CLP (p value 0.030) but not between children with CPO and CLP (p value 0.108).

DISCUSSION

This is the first study to describe school absence for specific reasons across a full academic year in a population-based cohort of 7-year-old children with isolated orofacial clefts. Compared with the national population, children with a cleft had small but significantly increased absence levels for illness and for medical/dental appointments but not for unauthorised reasons. Children who had a more anatomically involved type of cleft had increased levels of school absence.

We did not find evidence that the differences in school absence explain poorer levels of academic achievement among children with a cleft affecting the palate than children with a cleft affecting only the lip. Our results demonstrate that both cleft type and school absence are independently associated with academic attainment in children with a cleft.

Interpretation

Differences in annual school absence levels between 7-year-old children with an isolated orofacial cleft and corresponding children in the general population are due to differences in length of absence for illness and medical/dental appointments. This is in line with our previously published observations that children with more complex cleft types require more interventions from multiple specialties, including grommets insertions and dental procedures requiring general anaesthetic.3 7

It has been suggested that specific cleft-related factors, such as deficiencies in hearing and speech as well as other learning difficulties, such as attention disorder and dyslexia, may contribute to the observed attainment gap.3 11 We found that 40% of the 7-year-old children with an isolated cleft received SEN support, which is even higher than the 25% we observed in our previous study in children aged 5 years.10
Original research

Even children with CLO were found to have higher levels of SEN provision compared with the general population (26.7% compared with 20.9%). Although this could indicate a more generalised cerebral substrate, a recent genome-wide association study of children with isolated CLP compared with parental and unrelated controls found only limited evidence for a shared genetic aetiology or causal relationships between non-syndromic orofacial clefts and educational achievement or intelligence. This highlights the need for further research to explore how the attainment gap in children with a cleft can be addressed.

Our finding that reading and writing seemed to be most affected by absence in children with an orofacial cleft may highlight the need for specific educational support in these areas. Encouraging parents and teachers of children with clefts to seek additional opportunities for reading, writing and learning may prevent lower achievement associated with absence among the cleft population.

In contrast to our results, the study from Western Australia on school absence and academic achievement in children with orofacial clefts found no differences in absence rates between cleft groups and their non-cleft peers of similar age. Another contrast with our results is that the Australian study found the poorest academic achievement in children with CPO rather than in children with CLP.

Possible explanations for these differences in results are that we included a larger number of children with an orofacial cleft, that absence and academic achievement figures were all obtained within the same school year and that all the children in our sample were around 7 years of age. Lastly, the Australian study could only use absence figures obtained for the first half of the school year.

### Table 2

|                         | All absence | Unauthorised absence | Absence for illness | Absence for medical/dental appointments |
|-------------------------|-------------|----------------------|---------------------|----------------------------------------|
|                         | N  Mean 95% CI | N  Mean 95% CI | N  Mean 95% CI | N  Mean 95% CI |
| National population29   | 8.9 1.4 (1.2 to 1.9) | 5.4 0.5 (0.3 to 0.7) | 3234 1.4 (1.3 to 1.5) |
| Isolated orofacial cleft cohort | 3523 10.5 (10.2 to 10.9) | 3523 1.3 (1.1 to 1.4) | 3234 6.5 (6.2 to 6.7) | 3234 1.4 (1.3 to 1.5) |
| P value*                | <0.0001 0.1873 | <0.0001 0.1873 | <0.0001 0.1873 | <0.0001 0.1873 |

By isolated cleft type

|                         | N  Mean 95% CI | N  Mean 95% CI | N  Mean 95% CI | N  Mean 95% CI |
| Cleft lip only          | 920 9.5 (8.8 to 10.1) | 920 1.4 (1.0 to 1.8) | 814 6.0 (5.6 to 6.4) | 814 0.8 (0.7 to 0.9) |
| Cleft palate only       | 1257 10.5 (9.9 to 11.1) | 1257 1.2 (0.9 to 1.4) | 1150 6.6 (6.1 to 7.0) | 1150 1.4 (1.3 to 1.6) |
| Cleft lip and palate    | 1346 11.3 (10.7 to 11.9) | 1346 1.3 (1.1 to 1.5) | 1270 6.7 (6.3 to 7.1) | 1270 1.8† (1.7 to 2.0) |
| P value§                | 0.0002 0.4155 | 0.0662 0.4155 | <0.0001 0.4155 | <0.0001 0.4155 |

*One-sample t-test.
†p<0.0001 for difference with CLO.
‡p<0.001 for difference with CPO.
§One-way analysis of variance test of difference between cleft types. Post hoc comparison using Bonferroni correction.

CLO, cleft lip only; CPO, cleft palate only.

![Figure 1](image-url)

Figure 1  Proportion of children with an isolated cleft achieving the expected academic level or above across all five subject areas, according to cleft type and overall annual school absence.
the academic year, rather than the whole year, which may have affected its results, especially because that study period coincides with summer and autumn, when respiratory illnesses are less prevalent.

We found that adjustment for sociodemographic characteristics had relatively little effect on the differences in academic achievement according to school absence levels and according to cleft type, which suggests that it is unlikely that socioeconomic factors explain our findings.

**Strengths and limitations**

An important strength of our study is that we used three linked national datasets, including 3523 children who represent around three quarters of the total eligible isolated cleft population. This means that the reported results are representative of 7-year-old children in state-funded education in England. Also, the education records indicated the reasons for absence. The administrative hospital records in the HES database could be used to exclude children who had an additional anomaly or syndrome that eliminates the impact of other health issues on school absence and educational achievement.

The first limitation of our study is that academic achievement for 7-year-old children recorded in the NPD is based on teacher assessment, and teachers may have been influenced by the presence of the orofacial cleft. However, a recent study indicates that UK teachers do not perceive the challenges associated with

**Table 3** Proportion of 7-year-old children with an isolated cleft achieving the expected academic level or above across all five subject areas, according to overall annual school absence and cleft type.

| Annual overall school absence | Number (%) of children | Number (%) reaching the national ‘expected level’ of academic achievement* | OR (95% CI) | Adjusted for school absence | Adjusted for school absence, sex, IDACI* quintile and free school meal eligibility |
|------------------------------|------------------------|-------------------------------------------------|-------------|-----------------------------|--------------------------------------------------------------------------------|
| 0–5 days                     | 1192 (33.8)            | 923 (77.4)                                      | 1           | 1                           | 1                                                                             |
| 5.5–10 days                  | 943 (24.8)             | 675 (71.6)                                      | 0.73        | (0.60 to 0.89)              | 0.74 (0.61 to 0.90)                                                          |
| 10.5–15 days                 | 601 (17.0)             | 400 (66.6)                                      | 0.58        | (0.47 to 0.72)              | 0.59 (0.47 to 0.73)                                                          |
| 15.5–20 days                 | 342 (9.7)              | 202 (59.1)                                      | 0.42        | (0.33 to 0.54)              | 0.43 (0.33 to 0.56)                                                          |
| >20 days                     | 445 (12.6)             | 193 (43.4)                                      | 0.22        | (0.18 to 0.28)              | 0.23 (0.18 to 0.29)                                                          |

**Cleft type**

| Cleft type                  | Number (%) of children | Number (%) reaching the national ‘expected level’ of academic achievement* | OR (95% CI) | Adjusted for school absence | Adjusted for school absence, sex, IDACI* quintile and free school meal eligibility |
|------------------------------|------------------------|-------------------------------------------------|-------------|-----------------------------|--------------------------------------------------------------------------------|
| Cleft lip only               | 920 (26.1)             | 676 (73.5)                                      | 1           | 1                           | 1                                                                             |
| Cleft palate only            | 1257 (35.7)            | 828 (65.9)                                      | 0.70        | (0.58 to 0.84)              | 0.72 (0.60 to 0.88)                                                          |
| Cleft lip and palate         | 1346 (38.2)            | 889 (66.1)                                      | 0.70        | (0.58 to 0.85)              | 0.76 (0.63 to 0.92)                                                          |

*See text for further explanation.

IDACI, Income Deprivation Affecting Children Index.

**Figure 2** Proportion of 7-year old children with an isolated cleft achieving the expected academic level or above in each subject area at key stage 1, according to overall annual school absence.
isolated orofacial clefts to be a risk to long-term educational outcomes, which reduces the potential of observer bias.\

Second, the absence data only covers absence in the school year immediately before the assessment of the children’s academic achievement, and absence periods before that year were not considered.

Third, it cannot be ruled out that some included children had additional conditions that were not on our list of prespecified diagnoses to exclude. However, our approach to identify additional anomalies and syndromes has been extensively tested over the years, which strongly reduces its potential to affect the results of our study.

CONCLUSIONS

In England, children born with an isolated orofacial cleft had small but significantly higher levels of school absence than their peers in the national population. The increased school absence was mainly linked to absence for illness and for medical/dental appointments. Although total school absence was inversely associated with academic attainment, school absence levels did not explain the differences in academic attainment between children with different cleft types. Further work involving the linkage of educational data with clinical data may provide further insight into the educational needs of children with an isolated cleft and how these can be addressed.

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Contributors

KJF conceptualised and designed the study, carried out the analyses and interpretation of data, drafted the initial manuscript and revised and finalised the manuscript for submission. JHvD and SAD conceptualised and designed the study, interpreted the data and reviewed and edited the manuscript. LPC coordinated and managed the data collection from the three datasets and processed the Hospital Episodes Statistics data. MHP and JM conceptualised and designed the study, reviewed and edited the manuscript. All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

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Competing interests

None declared.

Patient consent for publication

Not required.

Provenance and peer review

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Data availability statement

Data are available on reasonable request. Data may be obtained from a third party and are not publicly available. Summary school absence data from the general population were obtained from https://www.gov.uk/government/collections/statistics-pupil-absence. Under our data agreements with NHS digital and the Department for Education, we are unable to share the data used for these analyses directly with others. Information on how to request Hospital Episode Statistics data can be obtained from https://digital.nhs.uk/data-and-information/data-tools-and-services/data-services/hospital-episode-statistics/users-uses-and-access-to-hospital-episode-statistics. Information on how to access National Pupil Database data can be obtained from https://www.gov.uk/guidance/how-to-access-department-for-education-dfe-data-extracts. Craniofacial Registry and Audit Network (CRANE) database data are available on reasonable request, which must be approved by the Cleft Development Group on behalf of NHS England (the data controller). Please see http://www.cranedatabase.org.uk/.

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