

## Educational achievements of children aged 10-11 years with cystic fibrosis. A data linkage study in Wales

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### Abstract

#### Introduction

As people with cystic fibrosis (CF) lead longer, healthier lives, educational qualifications and employment prospects are increasingly important. However, little is known about the social consequences of CF, in particular, any impact on educational achievements and the support children with CF receive in schools.

#### Objectives

To assess the educational achievements of children with CF in Wales compared to the general Welsh population, and the additional learning support children with CF receive in schools.

#### Methods

We conducted a population-scale data linkage study of all children born in Wales using the Secure Anonymised Information Linkage (SAIL) Databank. We used anonymised individual-level population-scale health and administrative data sources to identify children with CF born between 2000 – 2015, linked to educational attainment records. We calculated the percentage of children that reached expected levels in statutory assessment at age 10-11, Key Stage 2 (KS2), and compared this to educational outcomes in the general population. We also assessed the percentage of children with CF that received extra learning support.

#### Results

Out of 150 eligible children, 119 had KS2 results. 77% (95% CI: 69%-84%) of children achieved expected levels in English, 81% (95% CI: 73% -87%) in Mathematics and 82% (95% CI: 75% - 88%) in Science. In the comparable general Welsh population, 83.4% to 91.1% achieved the expected level in English, 84.9% to 91.6% in Maths, and 87.1% to 92.2% in Science across the years of the study. 70% of children with CF received extra learning support.

#### Conclusions

Children with CF in Wales may have worse educational achievements than the general population. More research is needed to inform policies and interventions to better support children with CF to reach their full educational potential and employment opportunities.

#### Keywords

data linkage; cystic fibrosis; education; SAIL Databank

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## Introduction

Cystic fibrosis (CF) is a serious inherited condition. One in 2,500 babies born in the UK have CF, with over 10,000 people living with CF in the UK today. CF is caused by variants of the CF transmembrane conductance regulator (CFTR) gene, resulting in the build-up of thick, sticky mucus in various organs, especially those of the respiratory and digestive systems. CF is therefore characterized by pulmonary infections and progressively declining lung function leading to most people with CF dying prematurely from their disease through respiratory failure. However, over the past decades, due to improvements in treatments and care, people with CF are living longer, healthier lives [1]. Half of the babies born with CF in the UK today are expected to live at least into their late forties [1, 2]. Employment prospects and financial stability are therefore becoming increasingly important issues. In 2019 13.5% of people with CF over the age of 16 were unemployed in the UK [2] compared to approximately 4% in the general population [3]. Evidence suggests that the likelihood of having full-time employment is more strongly linked to educational attainment than to disease severity [4]. Ensuring that children with CF achieve their academic potential is thus of utmost importance. However, few studies have investigated educational outcomes of children with CF, with most conducted in the US more than a decade ago. These studies suggest that children with CF show academic achievement levels comparable to the general population [5, 6].

To our knowledge, no study has looked at educational outcomes in the UK CF population. We utilised our recent population-scale data linkage in Wales [7, 8] to assess educational outcomes and the provision of extra learning support in the Welsh CF population, using comparable national data to contrast outcomes for children with CF to the general population of Welsh children. Due to data availability, we focus on educational achievements at ages 10-11 years, which have been shown to be strongly correlated with educational attainment at the end of high school [9, 10].

## Methods

### Study design, setting, participants and data sources

We undertook a population-scale data linkage study of all children born in Wales between 2000 and 2015 using the Secure Anonymised Information Linkage (SAIL) Databank [7, 8, 11, 12]. Every person in Wales registered with the National Health Service (NHS) is allocated an NHS number. Following the standard SAIL anonymization and linkage process, all data acquired into the SAIL Databank have identifiers removed, with the NHS number and all identifiers replaced with an encrypted unique personal identifier, known as an Anonymized Linkage Field (ALF). The ALF is used to link anonymised records across all available health and administrative data sources in SAIL, creating a longitudinal record of health care utilisation. We accessed ALFs for all children born in Wales between 2000 and 2015 through the National Community of Child Health (NCCCH) Database. We used the ALF for each

child to extract individual-level anonymised data from five additional data sources, including: the Congenital Anomaly Register and Information Service (CARIS); Patient Episode Database for Wales (PEDW); Welsh Longitudinal General Practice data (WLGP); the Annual District Birth Extracts (ADBE - also known as the Office of National Statistics (ONS) birth register); and the Welsh Demographics Service Dataset (WSDS).

We used ICD-10 and Read codes to obtain information on whether children had CF [7]. For these children, we further linked their data to education records available in SAIL from the National Pupil Database and the Pupil Level Annual Schools Census, for which we had follow-up to 2016. These data sources contain yearly educational attainment data for all pupils registered in schools in Wales.

## Outcomes

Our primary outcomes were attainment of the expected level in statutory assessment at age 10–11, Key Stage 2 (KS2), in English, Maths and Science. KS2 assessment is carried out by the teacher based on a child's day-to-day work, including classwork and homework. The teacher decides which level on the National Curriculum scale best suits the child's performance in each subject. At the end of KS2 (age 10–11 years), children are expected to achieve level 4 on the National Curriculum scale. We were also interested in whether the children with CF were designated as having 'Special Educational Needs' (SEN), meaning they can access extra learning support from the local authority.

### Expected ability at national curriculum level 4

This information is taken from [13], which gives ability levels for 2007; slight changes may have occurred over the time period of the study.

English:

- Recognise familiar words in simple texts.
- Begin to show awareness of how full stops are used in their own writing.
- Ideas are developed in a sequence of sentences, sometimes demarcated by capital letters and full stops.
- Express opinions about major events or ideas in stories, poems or non-fiction.
- Use their knowledge of the alphabet to locate books and find information.
- Writing is organised, imaginative and clear.
- Handwriting style is fluent, joined and legible.
- In response to a range of texts, pupils understand significant ideas, themes, events and characters, beginning to use inference and deduction.

Maths:

- Sort and classify objects, explaining their reason for doing so.

- Count, order and subtract numbers when solving problems involving up to 10 objects.
- Sort and classify objects in different ways, recording information in simple tables, block graphs and diagrams.
- Identify and use halves and quarters, such as half a rectangle or a quarter of eight objects.
- Use and interpret mathematical symbols and diagrams.
- Mentally solve multiplication and division problems (including those with remainders) using 2, 5 and 10 times tables.
- Multiple and divide whole numbers by 10 and 100.
- Work out perimeter of simple shapes, areas by counting squares and volumes by counting cubes.

Science:

- Identify a range of common animals.
- Recognise that sound and light come from a variety of sources and name some of these sources.
- Describe ways in which some materials are changed by processes such as bending and stretching.
- Know that living things need food, water, air and light to survive, and that different living things are found in different places.
- Explain why some materials are particularly suitable for specific purposes such as copper for making electrical cables.
- Make relevant observations and measure quantities such as length or mass using a range of simple equipment.
- Use scientific names for some major organs of a body system and identify the position of these organs in the human body.
- Choose suitable equipment to observe and measure work then present the results using tables and chart.

## Special educational needs

Special education needs are defined as follows:

“Children have special educational needs if they have a learning difficulty which calls for special educational provision to be made for them. Children have a learning difficulty if they: (a) have a significantly greater difficulty in learning than the majority of children of the same age; or (b) have a disability which prevents or hinders them from making use of educational facilities of a kind generally provided for children of the same age in schools within the area of the local education authority; (c) are under compulsory school age and fall within the definition at (a) or (b) above or would so do if special educational provision was not made for them” [14].

A medical diagnosis or disability does not necessarily indicate that the child has special educational needs; it

is the child’s educational rather than health need that is considered.

Following the identification of special educational needs of a child, help is given in a graduated response moving from School Action, to School Action Plus, to a Statutory Assessment [14]. School Action entails that after consultation with parents and gathering of information on the child, an individual education plan is drawn up which includes the strength and difficulties of the child, the additional help that will be provided by the school, and targets and timescales to assess the child’s progress [14, 15]. If the child has not made sufficient progress under School Action, outside specialists (e.g. educational psychologist, speech and language specialist) are brought in to advise on further changes that could be made within the school to address the child’s needs (School Action Plus). A revised individual education plan is drawn up, which outlines further support in, and possibility outside of, the classroom. Targets and timescales to assess progress are set [16, 17]. If progress under the School Action plan Plus is not satisfactory, the local education authority makes the decision whether a statutory assessment is needed. A statutory assessment involves a number of tests from a multi-disciplinary team (e.g. school, educational psychologist, paediatrician, social services) to assess whether a legal statement on the special educational needs of the child is required. The statement will include the educational and non-educational needs the child has and the arrangements by the local educational authority and external services to provide the child with the support [14, 15].

## Other variables

Demographic characteristics of the population, including year of birth and sex, were extracted from ADBE. CF-related hospital stays in the year of the KS2 assessment were extracted from PEDW.

## Analysis

We summarised the outcomes of interest as the percentage of children who achieved the expected level in KS2 between 2011 and 2016 in English, Maths and Science. 95% confidence intervals were calculated using the Wilson score interval. We repeated the analysis in the sub-population that had no CF-related hospital admissions in the year of the KS2 assessment to explore whether illness requiring hospitalisation may be affecting attainment. We compared the results with the percentage of children who achieved expected levels in KS2 in the general Welsh population during this time period as published by StatsWales [18]. These data were available for each of the years 2011-2016. For ease of comparison, we calculated the average of the percentage of children who achieved expected levels in KS2 in Wales across all years. However, pupil numbers were not available by year and therefore no weighting by population size could be performed.

We also calculated the percentage of children with CF who had a recorded SEN (School Action Plan, School Action Plan Plus, or legal entitlement to any specific package of support).

Table 1: Characteristics of the study population, KS2 results, and frequency of extra learning support provided to the children with CF

Characteristic	N(%) or median [IQR]
Male N(%)	68 (57%)
Year of birth N(%)	
2000	22 (18%)
2001	18 (15%)
2002	14 (12%)
2003	27 (23%)
2004	28 (24%)
2005	10 (8%)
Individuals with CF related hospital admission in KS2 year N(%)	23 (19%)
Number of nights in hospital in those with at least one admission during KS2 year (median [IQR])	13 [5.5–22.5]
<b>Special educational need</b>	
No special provision	38 (32%)
School Action Plan	32 (27%)
School Action Plan Plus	42 (35%)
Legal entitlement to specific support package	7 (6%)
<b>KS2 results across years 2011–2016</b>	
	<b>Percent achieved expected level (95% confidence interval)</b>
English	77% (69%–84%)
Mathematics	81% (73%–87%)
Science	82% (75%–88%)
<b>KS2 results in population with no hospital admission in KS2 year</b>	
English	78% (69%–85%)
Mathematics	81% (72% – 88%)
Science	84% (76%–90%)

## Results

We identified 312 children with a CF code in the linked data sources. 150 of these children were born before 2006 and could therefore have KS2 results by 2016; out of these 15 had moved out of Wales before age 11. 119 children had KS2 outcomes recorded. Table 1 shows the demographics of this population and hospital attendances as a marker for disease severity and potential school absence.

77% (95% CI: 69%–84%) of children achieved the expected levels in English, 81% (95% CI: 73%–87%) in Mathematics and 82% (95% CI: 75%–88%) in Science. There were no statistically significant differences in attainment in the full sample compared to the subpopulation of children that had no CF-related hospital admissions in the year of KS2 assessment (Table 1, Figure 1). The general Welsh population of children who sat KS2 assessment between 2011 and 2016 achieved higher average success in all three areas: 83.4% to 91.1% achieved the expected level in English in each of the years with a crude average across years of 87.3%; 84.9% to 91.6% achieved the expected level in Maths in each of the years with a crude average across years of 88.2%; and 87.1% to 92.2% achieved the expected level in Science in each of the years with a crude average across years of 89.8% [18] (Table 1, Figure 1).

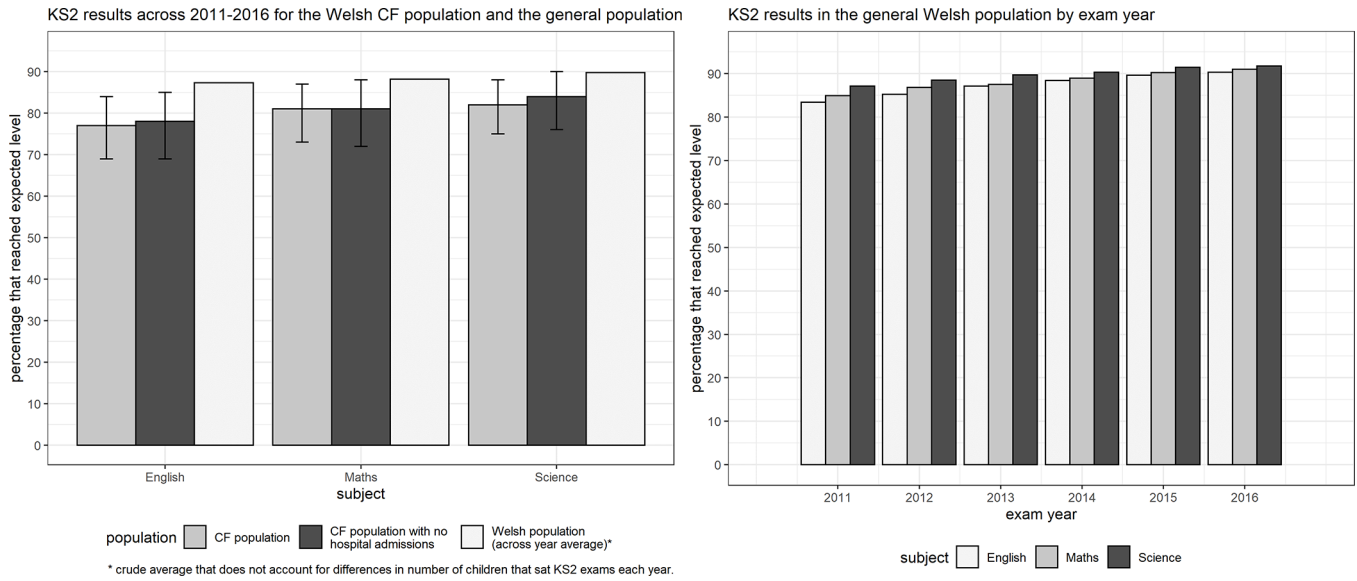
Almost 70% of the children with CF were recorded as receiving extra learning support (Table 1).

## Discussion

We assessed the educational outcomes at age 10–11 of children with CF in Wales. Our results suggest that children with CF may have lower educational achievements than the general Welsh population of children. Almost 70% of the children with CF received additional learning support which compares to 22.5% of all Welsh pupils in 2014/2015 [16].

Our analysis provides proof of principle demonstrating the feasibility of linkage of data on children with CF to educational data on educational progression and attainment, but there are a number of limitations. First, relying on ICD-10 codes can result in misclassification of patients with CF [19]. In a previous study on the same population we used both ICD-10 and Read codes to identify patients with CF across a range of linked data sources and found that 27% of individuals were wrongly classified as having CF when identification was based only on routinely-collected data [8]. Misclassification is therefore likely to be present in this study, and consequently the results have to be interpreted with caution. This is challenging to address because educational attainment data are incomplete

Figure 1: Comparison of Key Stage 2 (KS2) results in the Welsh CF population and the general Welsh population of children



Left panel: KS2 results for the study population and the subpopulation with no hospital admissions in the year of the KS2 exams. For comparison, the average of KS2 results in the general population is given, but the calculation of the average does not take into account the differences between the numbers of children that sat KS2 exams in each year. Right panel: KS2 results by year in the general Welsh population taken from [18] on which the average in the left panel is based.

in the UK CF Registry, the national disease registry which captures data on 99% of people with CF [20]. Linkage of the Registry to non-health data sources is currently not possible since this is not covered within the individual consent for linkage in the UK CF registry. Second, we did not have access to individual-level data on health status, such as lung function measures, or co-morbidities such as CF-related diabetes. We anticipate that we will be able to overcome these limitations in the context of a UK wide linkage of CF Registry data to the type of routine data sources available in SAIL. In the absence of this, the routine data linkage implemented in our study may offer the only possibility to gain insights into the impact of CF on educational attainment in a whole population of children. Third, we did not have individual-level data linked to educational outcomes for the non-CF population. Therefore, we were not able to adjust for potential confounders. However, our previous study on Welsh children showed that the distribution of many demographic variables, such as socio-economic conditions and sex, were comparable between children with and without CF [7]. Fourth, due to the small sample size, we were not able to explore whether there may be a differential impact of CF on educational outcomes by socioeconomic status or sex.

Our study suggests that children with CF may have worse educational attainment, and the reasons for this need further investigation. Children with CF are a heterogeneous group, and more detailed data on health status and co-morbidity is needed to assess health related pathways affecting educational attainment. Long-term school absence may be one of the main mechanisms through which chronic physical health conditions impact educational achievements [17]. Only 19% of children in this study spent any time in hospital during the year of assessment, and we found similar results when we excluded these children from the analysis. However, we did not capture

absences in the years prior to the assessment, or absences from school due to factors other than hospitalization. A recent study highlighted the potential impact CF treatment regimens have on children's home learning and their ability to participate in school activities such as PE and school trips [21]. The same study demonstrated that complex treatment regimens additionally restrict the time children have to develop and maintain friendships. This social disconnectedness has also been suggested as a mechanism through which physical illness can impact educational attainment [17]. With the advent of disease modifying treatments (CFTR modulators) for much of the CF population [22], it can be anticipated that children will spend less time in hospital and have less complex treatment regimens which may positively impact their education. This will increase the importance of contemporaneous research investigating the factors that influence the relationship between CF and educational achievement and the potential barriers that children with CF face.

Previous work has shown that people with CF are more likely to be unemployed if they are from more disadvantaged backgrounds and that disease severity has a larger impact on employment chances in these individuals compared to their less disadvantaged peers [23]. In future work, there is a need to explore the extent to which this can be explained by a differential impact of CF on children's educational attainment. Understanding social consequences of ill health is a key step in elucidating the pathways to health inequalities, since any adverse social outcomes that occur as a result of ill health can create a feedback loop and further damage health status.

In conclusion, we demonstrate that having CF may impact children's educational achievement. More research and better linked data in this area is needed to be able to develop policies and interventions to better support children to reach their full potential.

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This study makes use of anonymised data held in the Secure Anonymised Information Linkage (SAIL) Databank. We would like to acknowledge all the data providers who make anonymised data available for research.

## Declarations of interest

None

## Ethics statement

We use anonymised data in this study, therefore specific ethics approval was not needed. Approval for the use of data in this study, within the SAIL Databank, was granted by an independent information governance review panel (project 0504).

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