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School absence and achievement in children with isolated orofacial clefts

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SCHOOL ABSENCE AND ACHIEVEMENT IN CHILDREN WITH ISOLATED OROFACIAL CLEFTS

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).[1] A cleft can affect hearing,[2,3] language,[4,5] dental health,[6,7] psychosocial health,[8] neurodevelopmental health,[9] and academic outcomes.[9,10]

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).[10] These findings reflect the 'educational attainment gap' in children born with an orofacial cleft, even when treated under modern conditions and supported by a co-ordinated multidisciplinary team approach.[11] A study conducted in Western Australia compared absence rates and academic achievement in 310 children with an isolated cleft and 1,722 unaffected children varying in age between six and 15 years.[12] 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.[10]

We tested the hypothesis that differences in academic achievement in children according to cleft type can be explained by differences in school absence, using linked national datasets. First, we compared differences in school absence for specific reasons reported for children with different types of cleft with corresponding published national figures. Second, we examined the association between school absence and academic achievement. Third, we

explored to what extent differences in academic achievement according to cleft type could be explained by differences in school absence.

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 NHS number. The study cohort was identified in the Cleft Registry and Audit Network (CRANE) database.[13] A total of 5,976 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, 5,117 were eligible for inclusion in the study as their parents had given consent for information to be linked to other records (Supplementary File 1).

There were 4,759 children who could be linked to the Hospital Episode Statistics (HES) database, which contains administrative records on all admissions to National Health Service (NHS), the state-funded healthcare system that treats more than 95% of children born with a cleft in England.[14]

A total of 4,586 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 1,056 (23.0%) children who had additional anomalies or syndromes (Supplementary File 2). The remaining 3,523 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 (IDACI), an index of socioeconomic deprivation that is calculated for small areas (mean population about 1500),[15] eligibility for free school meals,[16] whether English was the first language, and whether special educational needs had been recognised, which can relate to physical, cognitive, sensory and behavioural difficulties.[17]

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.[18] 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% confidence intervals (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 chi squared tests. Logistic regression was used to compare these proportions and to estimate odds ratios 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 to 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 (Stator, College Station, Texas, USA).

Ethics

The study is exempt from ethics approval as it involves the analysis of data that are collected for the purpose of service evaluation,[19].

RESULTS

Characteristics of study cohort

Table 1 presents the characteristics of 3,523 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 1,426 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, 3,383 (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.[20] 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 five 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 socio-demographic characteristics had little impact on the odds ratios 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 odds ratios reflecting the differences in academic achievement according to cleft type changed only slightly with adjustment for school absence and with additional adjustment for socio-demographic 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 to 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] Page 9 of 22

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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.[3,21] 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 5-year-olds.[10]

Even children with CLO were found to have higher levels of SEN provision compared with the general population (26.7% compared to 20.9%). Although this could indicate a more generalised cerebral substrate, a recent genome-wide association study of children with isolated CLP compared to 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.[22] 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 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.[12] 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 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 3,523 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 provided 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 which 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 isolated orofacial clefts to be a risk to long-term educational outcomes, which reduces the potential of observer bias.[23]

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.

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Third, it cannot be ruled out that some included children had additional conditions that were not on our list of pre-specified 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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Abbreviations: CLO, cleft lip only; CPO, Cleft palate only; CLP, Cleft lip and palate; CRANE, Cleft Registry and Audit Network Database; HES, Hospital Episode Statistics; IDACI, Income Deprivation Affecting Children Index; NHS, National Health Service; NPD, National Pupil Database.

Contributors: Kate Fitzsimons 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.

Jan Van Der Meulen and Scott Deacon conceptualised and designed the study, interpreted the data and reviewed and edited the manuscript.

Lynn Copley coordinated and managed the data collection from the three datasets, and processed the Hospital Episodes Statistics data.

Min Hae Park and Jibby Medina 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.

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.
- Only one study has explored school absence in a cleft population and it reported inconsistent findings.
- Limited evidence indicates that school absence is associated with school performance • but reasons for absence in the cleft population are unknown.

What this study adds

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

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<text>

Table 1. Characteristics of study group: children born with an oral cleft in England who had school absence data for the academic year in which they turned 7 years of age (academic years 2006/7 to 2013/14 included).

	Isolated clo (n=35		National figures according to th National Pupil Database[24]
Characteristic	number	(%)	(%)
Cleft type			
Cleft lip only	920	26.1	-
Cleft palate only	1257	35.7	-
Cleft lip and palate	1346	38.2	-
Sex			
Female	1458	41.4	51.0
Male	2065	58.6	49.0
IDACI* group (quintiles of national distribution)			
1 (least deprived)	549	15.7	17.4
2	666	19.0	18.0
3	670	19.1	18.4
4	774	22.1	20.5
5 (most deprived)	843	22.1	20.5
missing	21	-	23.1
Eligible for free school meals			
No	2739	78.1	80.6
Yes	769	21.9	19.4
missing	15	-	
Ethnic group			
White	3096	87.1	76.8
Non-white	426	12.1	23.2
Missing	1		
English as first language			
Yes	3209	91.2	82.5
No	309	8.8	16.9
missing	5	-	
Recognised special educational needs (SEN) and	approach		
No	2091	59.5	79.1
Yes (Total)	1426	40.5	20.9
Yes - School Action	526	15.0	12.4
Yes - School Action Plus*	728	20.7	6.6
Yes – Statement*	172	4.9	1.9
missing	6	-	
D			
Primary type of SEN**	51 0	14.5	2.5
Speech, language & communication needs	510	14.5	2.5
Hearing impairment	73	2.1	0.2
Learning difficulties	142	4.0	2.8
Behavioural, emotional & social difficulties	77	2.1	1.6
Other	98	2.8	1.4

* IDACI, Income Deprivation Affecting Children Index. See text for further explanation

**Type of SEN is documented for only those with School Action Plus or a Statement of SEN

		All al	osence	U	nauthorise	ed absence		Absence f	or illness	Abse	ice for n appoin	nedical/dental tments
(Ν	Mean	95% CI	Ν	Mean	95% CI	Ν	Mean	95% CI	Ν	Mean	95% CI
National population[20]		8.9			1.4			5.4			0.5	
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			< 0.0001	
By isolated cleft type												
Cleft lip only	920	9.5	(8.8 to 10.1)	920	1.4	(1.0 o 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	1.4ª	(1.3 to 1.6)
Cleft lip and palate	1346	11.3ª	(10.7 to 11.9)	1346	1.3	(1.1 to 1.5)	1270	6.7	(6.3 to 7.1)	1270	1.8 ^{ab}	(1.7 to 2.0)
P value**		0.0002		10	0.4155			0.0662			< 0.0001	

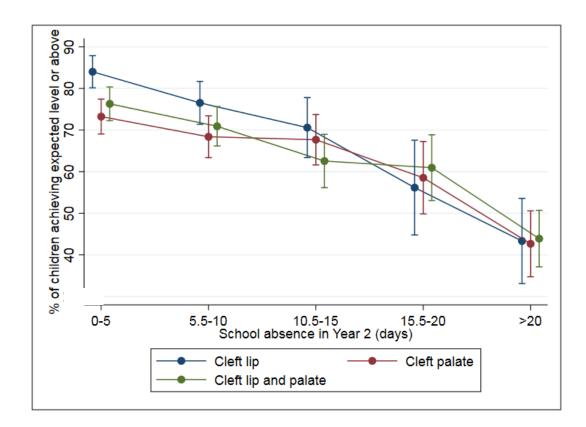
* One-sample t test

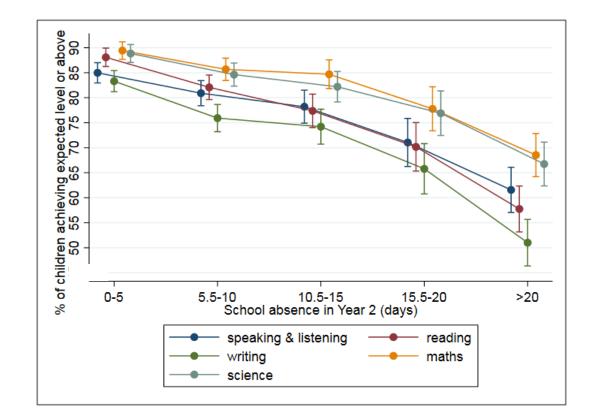
** one-way analysis of variance test of difference between cleft types. Post hoc comparison using Bonferroni correction: *P<0.0001 for difference with CLO; ^b P<0.001 for difference with CPO

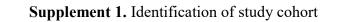
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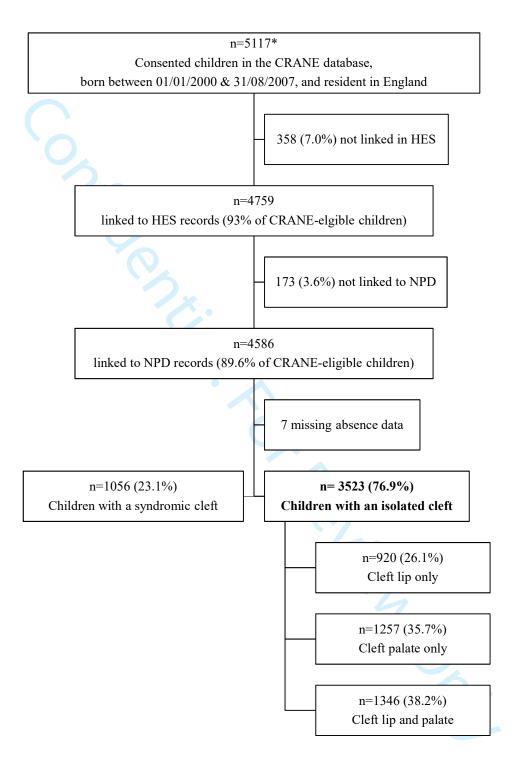
Table 3. Proportion of 7-year-old children with isolated orofacial cleft reaching national	'expected levels'	of academic achievement across five
subject areas, according to overall annual school absence and cleft type.		

		Number (%) reaching the	Odds ratio (95% CI)						
Annual overall school absence	Number (%) of children	national 'expected level' of academic achievement*	Unadjusted Adjusted for cleft type				Adjusted for cleft type, 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)	0.75	(0.61 to 0.92)	
10.5-15 days	601 (17.0)	400 (66.6)	0.58	(0.47 to 0.72)	0.59	(0.47 to 0.73)	0.63	(0.50 to 0.79)	
15.5-20 days	342 (9.7)	202 (59.1)	0.42	(0.33 to 0.54)	0.43	(0.33 to 0.56)	0.52	(0.40 to 0.68)	
>20 days	445 (12.6)	193 (43.4)	0.22	(0.18 to 0.28)	0.23	(0.18 to 0.29)	0.30	(0.23 to 0.38)	
						Odds ratio (95% Cl	f)		
Cleft type	Number (%) of children	Number (%) reaching the national 'expected level' of academic achievement*	U	nadjusted	Adjusted 1	for school absence	IDACI*	for school absence, sex, 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)	0.64	(0.52 to 0.78)	
Cleft lip and palate	1346 (38.2)	889 (66.1)	0.70	(0.58 to 0.85)	0.76	(0.63 to 0.92)	0.77	(0.63 to 0.94)	









*There were 5,976 children born with a cleft in England between 01/01/2000 and 31/08/2007 who were notified to the national cleft registry, CRANE. 5117 (85.6%) had consent confirmed and formed our eligible cohort.

Supplement 2: Diagnostic codes for syndromes and anomalies used to identify children with additional medical anomalies.

Patients were defined as having additional medical anomalies if there was a record of any of the following codes in any of the fourteen diagnosis code fields for any of that patient's HES episodes. i.e. the syndromic diagnosis mention is not necessarily associated with a record containing the patient's first cleft diagnosis/repair.

Code	Description
D821	Di George's syndrome
	Congenital malformations of the nervous system (Q00-Q07)
Q00	Anencephaly and similar malformations
Q01	Encephalocele
Q02	Microcephaly
Q03	Congenital hydrocephalus
Q04	Other congenital malformations of brain
Q05	Spina bifida
Q06	Other congenital malformations of spinal cord
Q07	Other congenital malformations of nervous system
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Q16	Congenital malformations of ear causing impairment of hearing
Q18	Other congenital malformations of face and neck
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	Congenital malformations of the circulatory system (Q20-Q28)
Q20	Congenital malformations of cardiac chambers and connections
Q21	Congenital malformations of cardiac septa
Q22	Congenital malformations of pulmonary and tricuspid valves
Q23	Congenital malformations of aortic and mitral valves
Q24	Other congenital malformations of heart
Q25	Congenital malformations of great arteries
Q26	Congenital malformations of great veins
Q27	Other congenital malformations of peripheral vascular system
Q28	Other congenital malformations of circulatory system
Q380	Congenital malformations of lips, not elsewhere classified
Q75	Other congenital malformations of skull and face bones
Q86	Congenital malformation syndromes due to known exogenous causes, not
-	elsewhere classified
Q87	Other specified congenital malformation syndromes affecting multiple systems
	Chromosomal abnormalities, not elsewhere classified (Q90-99)
Q90	Down's syndrome
Q91	Edwards' syndrome and Patau's syndrome
Q92	Other trisomies and partial trisomies of the autosomes, not elsewhere classified
Q93	Monosomies and deletions from the autosomes, not elsewhere classified
Q95	Balanced rearrangements and structural markers, not elsewhere classified
Q96	Turner's syndrome
Q97	Other sex chromosome abnormalities, female phenotype, not elsewhere classified
Q98	Other sex chromosome abnormalities, male phenotype, not elsewhere classified
099	Other chromosome abnormalities, not elsewhere classified