Educational progress, behaviour, and motor skills at 10 years in early treated congenital hypothyroidism

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Abstract

Aim—To assess educational attainments, behaviour, and motor skills at 10 years of age in a group of children with congenital hypothyroidism identified by neonatal screening.

Subjects—59 children with congenital hypothyroidism born in 1978–81, 31 cases with pretreatment thyroxine (T4) values of 40 nmol/1 or below (group I) and 28 less severe cases with T4 values over 40 nmol/1 (group II), together with 59 classroom control children matched for age, sex, social class, and main language spoken at home.

Methods—The Neale analysis of reading ability; the child health and education study written test of mathematics; Rutter behaviour questionnaires for parents and teachers; the Oseretsky test of motor proficiency (short form).

Results-On all measures the congenital hypothyroidism children in group I had less satisfactory scores for educational attainments, behaviour, and motor skills than those in group II and controls. For reading the differences were small and did not reach statistical significance, but the deficits in mathematics and total motor skills were statistically significant (p < 0.01). There were more striking and statistically significant differences in behaviour scores, particularly with respect to attentional difficulties. Although less striking, these were also apparent in the group II children with mild hypothyroidism.

Conclusions—At the age of 10 years severe congenital hypothyroidism is associated with some mild impairment in educational and motor attainments. Behaviour problems are also common, even in some children with less severe congenital hypothyroidism.

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Keywords: congenital hypothyroidism; educational progress; motor skills; behaviour

Neonatal screening linked with early treatment has improved the prognosis for children with congenital hypothyroidism. However, while follow up studies have shown that most children with congenital hypothyroidism achieve scores for intelligence within the normal range, those with severe hypothyroidism show significant deficits in mean IO scores despite early treatment, and this effect appears to have a threshold.1 There are conflicting results on educational attainment. Essentially normal educational progress at 9-10 years has been reported in a cohort of children with congenital hypothyroidism in North America²; in contrast, a group of children in France were found to have impairment of mathematical skills, as opposed to school tasks covering language and vocabulary,3 and an increased prevalence of learning problems has been reported in Canadian children with congenital hypothyroidism.4 Impaired motor performance⁵ and behaviour problems6 have also been reported in children with congenital hypothyroidism, but the extent of these difficulties is still not clear.

This paper describes attainments in reading and mathematics, motor skills, and behaviour in a group of 10 year old children with congenital hypothyroidism who were born in 1978–81 and compares them with normal classroom controls.

Methods

SUBJECTS

The subjects were 10 year old children born in 1978–81: 59 with congenital hypothyroidism identified by neonatal screening, and 59 classroom controls matched for age (within three months), sex, social class, and the main language spoken at home. Early clinical findings have been given elsewhere, as have the results of psychometric testing at 3, 5, and 10 years of age. 5–10

PROCEDURES

Each child was assessed individually at school and all the tests were carried out by one observer (WFS).

Educational attainments

Reading ability was assessed using the Neale analysis of reading ability (1989 revised British edition). Raw scores were converted to 'reading ages' in months with respect to rate, accuracy, and comprehension.

Mathematical ability was tested using a written test compiled from the child health and education study, Bristol (1980)¹² with 72 questions grouped into six subscores for arithmetic rules, number concepts, measure, algebra, geometry, and statistics.

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Table 1 Mean (SE) scores for reading age (months) and mathematics at 10 years in two groups of children with congenital hypothyroidism and matched controls. Values are means (SE)

	Group I: pretreatment $T4 \le 40 \text{ nmol/l}$		Group II: pretreatment T4 > 40 nmol/l		
	Patients (n=31)	Controls $(n = 31)$	Patients (n=28)	Controls (n=28)	
Reading					
Rate	113.3 (1.9)	115.7 (1.7)	115.3 (2.1)	116.7 (2.2)	
Accuracy	120.4 (1.8)	122.4 (1.8)	122.3 (2.3)	123.1 (2.2)	
Comprehension	117.2 (1.7)	120.4 (1.5)	119.3 (1.7)	120.4 (1.6)	
Mathematics Total score	33.6 (2.3)†	39.9 (2.1)†	43.9 (2.9)	40.9 (2.69)	

†p < 0.01.

Table 2 Parents' and teachers' questionnaire items in which a significantly higher percentage of the hypothyroid children in group I or group II had scores of 1 or 2, compared with matched controls. The percentagesof children with scores of 13 or more on the parents' questionnaire and 9 or more on the teachers' questionnaire are also given

	Group I: pretreatment T4 \leq 40 nmol/l (%)		Group II: pretreatment T4 > 40 nmol/l (%)	
	Patients (n=31)	Controls (n=31)	Patients (n=28)	Controls (n=28)
Parents' questionnaire				
Specific behaviours				
Difficulty sleeping	42*	13	28	21
Cannot settle	35*	6	18	18
Restless	29	16	43*	14
Overall scores of 13 or more	23	6	29	18
Teachers' questionnaire				
Specific behaviours				
Very restless	32*	6	21	14
Fussy, overparticular	29*	6	21	14
Squirmy, fidgets	32*	10	18	14
Unresponsive	39*	16	18	7
Rather solitary	39*	13	35	14
Often appears miserable	32	16	32*	4
Fearful	55*	32	43	28
Overall scores of 9 or more	23*	3	18	11

^{*}p < 0.05 v respective controls.

Behaviour

Parents were asked to complete the Rutter behaviour questionnaire for parents. This has 31 statements concerning behaviour, each with a score of 0–2, indicating whether a particular behaviour 'doesn't apply' (score 0), 'applies somewhat' (score 1), or 'certainly applies' (score 2). A total score of 13 or more is taken to indicate the presence of some behavioural disorder.¹³

Teachers were asked to complete the Rutter behavioural questionnaire for teachers which has 26 questions, each with a score of 0–2 as described above. A total score of 9 or more is taken to indicate the presence of a behavioural disorder.¹³

For both questionnaires the expected rates of behavioural disorder in a community sample would be around 15%.

Table 3 Mean (SE) scores for tests of motor function at 10 years in the two groups of children with congenital hypothyroidism and matched controls

	Group I: pretreatment $T4 \leq 40 \text{ nmol/l}$		Group II: pretreatment $T4 > 40 \text{ nmol/l}$		
	Patients (n=31)	Controls (n=31)	Patients (n=28)	Controls (n=28)	
Balance	9.8 (0.7)	10.8 (0.5)	10.3 (0.6)	11.0 (0.6)	
Coordination	13.5 (0.6)*	15.2 (0.4)	14.0 (0.6)	14.6 (0.5)	
Hand speed	12.5 (0.6)†	14.6 (0.4)	14.8 (0.6)	14.8 (0.5)	
Total score	35.8 (1.3)†	40.6 (0.9)	39.1 (1.3)	40.5 (1.0)	

^{*}p < 0.05, † p < 0.01 v respective controls.

Motor skills

Motor skills were assessed using the short form version of the Oseretsky test of motor proficiency¹⁴ which has 12 subtests, seven scored separately for each limb. Tests were scored from 0 to 3, giving a maximum score of 57 points. Test scores were grouped to give composite scores for balance (five items), coordination (six items), and hand speed (eight items).

STATISTICAL ANALYSIS

As results obtained at 10 years of age indicated that a low pretreatment thyroxine (T4) was associated with impairment of IQ score10 and an apparent threshold effect of pretreatment T4 on IO was found at around 40 nmol/l in a larger study,1 subjects were divided into two groups: group I with pretreatment thyroxine (T4) levels of 40 nmol/l or below (n = 31) and group II with pretreatment T4 levels above 40 nmol/l (n = 28). Results for groups I and II and the corresponding control groups were compared using paired t tests to assess the significance of differences in mean values for educational scores and motor skills, and McNemar's test to assess differences in the prevalence of abnormal behaviours. A p value < 0.05 has been taken to indicate statistical significance.

Results

In all the assessments, children with severe hypothyroidism (group I) had less satisfactory overall scores than the children with milder hypothyroidism (group II) and the controls.

READING AND MATHEMATICS

The mean (SE) scores obtained with the reading and mathematics tests are summarised in table 1. The mean scores for reading rate, accuracy, and comprehension were similar in both groups of hypothyroid children and in the controls. The congenital hypothyroidism children in group I had a significantly lower mean score for mathematical skills than those in group II or the controls (p < 0.01).

BEHAVIOUR

The findings with the parents' and teachers' behaviour questionnaires are summarised in table 2. Parents' ratings of behaviour in the children with congenital hypothyroidism showed generally higher scores for behavioural difficulties than in the controls. The largest differences were for items relating to regulation of activity, that is, restlessness and difficulty settling down to tasks or to sleep. Teachers also reported higher rates of difficulty with regulation of activity, including higher rates of unresponsiveness, as well as restlessness and difficulty in settling. An increased incidence of withdrawn, miserable behaviour in the children with congenital hypothyroidism was also reported by teachers, but not by parents.

'Tends to be absent for trivial reasons' was the only item scored more often in the group II controls than in the children with congenital hypothyroidism.

MOTOR SKILLS

The results for the tests of motor skills are given in table 3. Small but significant differences (p < 0.05 and p < 0.01) were present between group I and group II, and between group I and the controls, whereas the motor skill scores in group II were similar to those for the control group.

Discussion

We have already reported the psychometric results at the ages of 3, 5, and 10 years in this group of patients with congenital hypothyroidism. 8-10 While differences in mean IQ score between the children with hypothyroidism and matched controls were small, children with more severe hypothyroidism-as judged by their pretreatment plasma T4 levels—consistently showed a significant deficit in IQ score of 10-12 IQ points in comparison with children with less severe congenital hypothyroidism and with controls. These latter findings are in line with results reported from other countries, notably Canada¹⁵ and the Netherlands.16 They are also entirely in keeping with the results of a national survey carried out on a large cohort of children with congenital hypothyroidism born in England, Wales, and Northern Ireland in 1982-84, in which there was evidence of a threshold effect of the pretreatment biochemical severity of hypothyroidism on IQ score.1

Given these deficits in IQ score, it was of particular interest to see whether severe hypothyroidism was also associated with poorer educational attainments. The results presented here suggest that there is some impairment but that this is small and probably of minor practical significance. The findings are generally in keeping with those obtained by Rochiccioli et al in 40 children with congenital hypothyroidism who had completed primary school³; tests of vocabulary, grammar, and spelling gave similar results in the children with congenital hypothyroidism and the controls but the hypothyroid children had impairment of mathematical skills. Educational difficulties in congenital hypothyroidism have also been reported from Canada.4 The New England congenital hypothyroidism collaborative2 reported satisfactory educational achievements in 72 children with congenital hypothyroidism who were assessed at the age of 9-10 years, but also reported a normal mean IQ score in this group of children. All these results are encouraging. However, the deficit in mean IQ in the present subjects shows the need to be cautious about the overall educational progress of this group, and further evaluation is required when the subjects have completed their education.

Before the introduction of screening, behavioural problems were common in children and adults with congenital hypothyroidism^{17 18} and our results suggest that mild behaviour problems are common in early treated cases. There is already evidence that early treated hypothyroid children may have social and conduct problems, with increased levels of anxiety or hyperactivity, if they are treated with high doses of l-thyroxine during early infancy⁶; this is not

the explanation for the results in the patients described above. We did not find any evidence of increased aggressive or antisocial behaviour, but 'emotional' features such as solitary behaviour, a miserable appearance, a fearful appearance, and fussy behaviour were common, particularly in subjects with more severe hypothyroidism at diagnosis. Equally striking were the increased rates of problems relating to regulation of activity which were reported by both parents and teachers. This finding supports the presence of persistent attentional problems as reported by Kooistra et al19 and suggests that children with congenital hypothyroidism remain vulnerable to these types of difficulty, but usually in only mild to moderate degree.

We have previously reported impaired motor skills at the ages of 3 and 5 years in the present group of patients with congenital hypothyroidism, particularly in children with low plasma T4 values at diagnosis, and our current study suggests that these motor deficits can still be detected at 10 years of age in the children with pretreatment T4 concentrations of 40 nmol/l or below, although the effects appear to be small. The findings are in general agreement with other reports. For example, impaired performance on a peg board test was demonstrated at 9-10 years in the New England study² and Rochiccioli et al found impaired coordination in children with congenital hypothyroidism aged 6 years,⁵ as did Gottschalk et al20 in a group of children with congenital hypothyroidism aged 5 to 9 years. It therefore appears that minor neurological impairment may persist during childhood in children with severe congenital hypothyroidism, despite early treatment.

In conclusion, our findings suggest that children with severe congenital hypothyroidism have minor difficulties in educational achievement at 10 years of age, some difficulties with coordination and hand control, and may also have more problems with behaviour, including restlessness and inability to concentrate; there seems to be a tendency for these latter difficulties to extend to children with milder forms of hypothyroidism. It is important that paediatricians are aware of these potential problems so that counselling and remedial action can be taken when appropriate.

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