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ORIGINAL PAPER

Deficient language acquisition in children with single suture craniosynostosis and deformational posterior plagiocephaly

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Abstract
Purpose This study examined early language acquisition in children with single suture craniosynostosis (SSC) and in children with deformational posterior plagiocephaly. Our purpose was to determine whether infants with SSC have normal language acquisition at the age of 3 years, and whether infants with deformational posterior plagiocephaly demonstrate parallel development when compared with children with SSC.
Methods The study population includes 61 infants. Twenty of them had synostosis of the sagittal suture, 12 synostosis of other suture and 29 deformational posterior plagiocephaly. Forty-nine of them were operated on a mean age of 10.6 months, and 12 were non-operated children with deformational posterior plagiocephaly. Language skills of participants were prospectively evaluated at the mean age of 3 years 4 months.
Results About one half of the subjects (49%) had normal linguistic development, 30% had slight developmental problems and 21% had severe disorders in speech–language-related skills. These figures showed the prevalence of severe language disorders to be three times higher in our study population when compared with the general population. Children with sagittal synostosis managed better in all language skills compared with other types of SSC. Defective language development was found in deformational posterior plagiocephaly, both operated and non-operated.
Conclusions We found a noticeable developmental risk for specific language impairment in children with nonsyndromic SSC, and that the deviant language development is observable already in early infancy. Contrary to previous beliefs, the developmental risk for defective language development in deformational posterior plagiocephaly was found in both operated and non-operated subjects.

Keywords Single suture craniosynostosis (SSC) · Deformational posterior plagiocephaly · Specific language impairment (SLI) · Language development · Central auditory processing (CAP)

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Introduction

Single suture craniosynostosis (SSC) is one of the most common craniofacial congenital malformations, affecting approximately 1 in 2,000 to 2,500 live births [1]. Craniosynostosis is caused by premature fusion of a cranial suture, such as the sagittal, unicoronal, metopic or lambdoid. In modern diagnosis, 3D computerised tomography (3D CT) imaging is performed to identify the cranial sutures and the bony structures of the head [2]. The most common form of SSC is that involving the sagittal suture. About one half of children with SSC are reported to have speech, cognitive and/or behavioural abnormalities [3].

The causes of SSC are still unknown, and in many cases the aetiology is idiopathic. Researchers have not been able to determine the effect of the fused suture on the underlying cortex. According to neurobiological studies, the dura may send abnormal signals to the overlying suture, and this may lead to the premature closure [4]. Aldridge and colleagues

have reported that morphological changes in the brains of children with SSC are not limited to the areas directly beneath the fused suture [5]. MRI studies have shown changes in brain connectivity both before and after surgical correction [6].

Neurofunctional changes are not the only explanation for developmental delays and specific problems in SSC. Elevated intracranial pressure is estimated to occur in about 25% of patients with sagittal synostosis [7]. Cohen and Persing found intracranial hypertension to be a significant indication for surgery for craniosynostosis [8]. In an earlier study, Renier and colleagues found that increased intracranial pressure (ICP) correlates with the mental level of children with craniosynostosis [9]. The increased ICP can be alleviated by surgery. However, early operations do not seem to prevent neurocognitive delays in children with SSC [10–12].

The diagnosis and treatment of posterior plagiocephaly is one of the most controversial aspects of craniofacial surgery. Deformational plagiocephaly is usually first noticed when an infant is about 2–3 months old. According to a commonly accepted explanation, deformation occurs from the child's lying in the supine position in the early perinatal period. As a consequence the calvarium becomes compressed in the occipital region ipsilaterally and the frontal region contralaterally, resulting in a parallelogram-shaped head [13]. In Panchal's study, severe neurodevelopmental and cognitive delays were reported especially in the group of children with plagiocephaly. The Psychomotor Developmental Index (Bayley Scale of Infant Development-II) indicates significant differences compared with the standard population ($p<0.001$). Kordestani and colleagues reported similar results [14]. According to their study, infants with deformational plagiocephaly showed significant delays in both mental and psychomotor development.

Well-functioning central auditory processing (CAP) is highly related with the cortical bases of language acquisition [15, 16]. CAP refers to the efficiency by which the central nervous system utilizes auditory information. This activity gives rise to electrophysiological auditory potentials. An interesting way to look at the developmental question behind plagiocephaly comes from the event-related potential (ERP) study by Balan et al. [17]. Auditory-evoked responses elicited by sine tones were found to be decreased in infants with abnormal skull shape and unilateral occipital flattening without synostosis. Atypical ERP amplitudes (obligatory responses P150 and N250) were recorded in a non-attend research paradigm. The authors suggest that decreased amplitudes might indicate an auditory processing dysfunction, possibly the result of the delayed or disturbed maturation of the auditory pathways. This observation can in turn be related with earlier reports of decreased auditory-evoked potentials in language-impaired children [18], prematurely born children [19] and children with cleft palate [20].

Neurodevelopmental impairments are commonly reported in SSC [6]. It has been noticed that even though children with SSC may reach normal values during the first year of life, the risk for developmental delay increases in future years [3, 11, 21]. Most of the studies of SSC have inspected cognitive and psychomotor development without a special interest in speech–language development. The explanation for this is that these earlier studies concerned children less than 2 years of age, covered only the period of first word acquisition or focused on more specialised skills in school age children. In the recent study, Chieffo and her fellow researchers [22] reported that one third of the school-aged children with anterior plagiocephaly had special difficulties in processing and planning of speech as evaluated with verbal fluency tests. They also found milder deficiency in speech comprehension and lexical skills. The language-related problems were especially typical of children with unicoronal craniosynostosis.

In the present study, we investigated the risk factors for specific language impairment (SLI) related with SSC. To better understand the developmental profile and special problems related to language acquisition in SSC, we present a population of 32 children with SSC and 29 children with deformational posterior plagiocephaly. This study inspected language acquisition during the most active developmental period.

Material and methods

Study population

Children were eligible if they (1) had nonsyndromic SSC (sagittal, unicoronal, metopic or lambdoid) or posterior deformation of the skull confirmed by computer tomography and (2) came from monolingual Finnish families. Exclusion criteria for cases were (1) the presence of major medical or neurological conditions and (2) the presence of major or minor malformations. In the study group, there was one child of premature birth (before 34 weeks gestation). In this case the corrected age was used for the assessment.

Sixty-one children were prospectively studied at the mean age of 3.4±0.3 years. The study involved 32 children with single suture craniosynostosis and 29 children with deformational posterior plagiocephaly. The operations were performed at the mean age of 10.6±6.6 months. The participants were divided into four subgroups as follows: group I: 20 children with sagittal synostosis, 18 males;

group II: 12 children with synostosis of other suture, 6 males; group III: 17 operated children with deformational posterior plagiocephaly, 8 males and group IV: 12 non-operated children with deformational posterior plagiocephaly, 8 males (Table 1). All the patients in groups I–III were operated in the Department of Plastic Surgery, Cleft Palate and Craniofacial Centre of Helsinki University Hospital. The non-operated children in Group IV had milder deformations without indication to operative treatment or their parents chose continued monitoring instead of the operation. Helmet therapy is rarely used in Finland.

Consecutive cases both in operated and non-operated group of children were offered the possibility of receiving assessment of developmental skills while visiting the hospital for their 3-year check-up. Children were referred to the project at the time of diagnosis by the treating surgeon. A certified speech–language pathologist performed the assessments. The data concerning early language development were collected from the parents and those on general motor development from clinical reports. Forty-eight children were tested and their parents interviewed by PK (the first author) and 13 children by the acting speech–language pathologist of the Cleft Palate and Craniofacial Centre of Helsinki University Hospital. The behavioural testing took about 60 min.

Measures

The parents' questionnaire concerned the following developmental aspects: the child's condition at the time of testing, early motor development, early vocalisation, babbling and first word acquisition, occurrence of first sentences, the child's readiness to use language with other people, and the parents' expectations for the child's future development. The data concerning general motor development were collected from clinical reports. The motor development was graded using a two-step scale, with value of 1 for deviant development or notable delay, and value 2 for age-typical development.

A certified speech and language pathologist examined all participants. The Reynell Developmental Language Scales III [23, 24] was used to measure the child's language status. The standardized scores on the Reynell scales are based on a mean value of 100 (SD, 15), and the clinical cut-off point for this test is 80. Lexical skills were

Table 1 Participant characteristics, groups and test results

Characteristic	Sagittal	Other SSC type	Posterior	Posterior non-operated	All cases	
					n	%
n	20	12	17	12	61	100
Gender						
Female	2	6	9	4	21	34
Male	18	6	8	8	40	66
Age					GA mean±SD	
Operation month	5.4±2.2	16.1±8.6	12.8±3.5	–	10.6±66	
Assessment years	3.4±0.4	3.4±0.4	3.3±0.3	3.3±0.3	3.4±0.3	
First word acquisition						
Month	15.7±6.5	14.3±6.8	12.6±3.3	15.8±6.8	14.5±5.9	
First sentences						
Month	23.6±5.4	25.7±6.1	22.7±5.4	24.2±4.6	23.9±5.4	
Reynell Scales						
Receptive	94.8±14.8	93.3±19.8	104.1±8.2	95.1±15.0	97.1±14.9	
Expressive	108.0±12.6	107.0±7.5	108.9±8.7	103.4±16.0	107.0±12.0	
Mental	102.6±12.8	103.2±8.2	107.4±6.8	99.8±15.1	103.4±11.6	
Language level						
(Scale 1–3)	2.4±0.8	2.1±0.8	2.3±0.8	2.3±0.9	2.3±0.8	
Articulation/phonology						
(Scale 1–2)	1.8±0.4	1.3±0.5	1.5±0.5	1.7±0.5	1.6±0.5	
Gross motor development						
(Scale 1–2)	1.9±0.3	1.7±0.5	1.9±0.3	1.8±0.4	1.8±0.4	

measured with the Renfrew Naming Task [25] and the Finnish version of MacArthur Communicative Development Inventories [26, 27]. For the statistical analysis, quantitative values of lexical development were modified using a three-step scale: value of 1 for more than 1 year below normative scores, 2 for less than 6 months below and 3 for age-appropriate development.

In addition to the formal testing, the speech–language pathologist observed the children in a semi-structured play situation and graded the development of phonological and morphological skills and the child's ability to form sentences. These skills were graded either delayed (value 1) or normal (value 2). The final assessment of the child's developmental level of language-related skills was based on the formal tests scored, in addition to informal observations of the play situation. For the speech–language level, a three-step scale was used: value 1 for severe delay, value 2 for notable delay and value 3 for age-appropriate development.

Data analysis

To estimate the differences between the study groups, we used Fisher's Exact Test. Group-related differences were counted by using one-way ANOVA and Tukey's Studentized Range Test. Comparisons between groups (risk analysis) based on Binary Logistic Regression analysis. We also calculated paired samples *t* test and Pearson correlations to compare the child's performance in single assessments and tests. The statistical significance level was set at 0.05.

Results

Table 1 lists the characteristics and test results of the study population. Of the 61 subjects, 34% were females and 66% males. The diagnoses differed according to the sex of the children, with a larger proportion of males in the sagittal group ($p=0.023$). The groups also differed in terms of the age at the operation time (mean, 10.6±6.6 months), which was highest in group II, i.e. children with synostosis of other suture (mean, 16.1±8.6 months, $p<0.0001$). The age for testing was equally distributed in all study subgroups (mean, 3.4±0.3 years).

About one half of the subjects (49%) had normal linguistic development, 30% had slight developmental problems and 21% had severe developmental delays in speech–language-related skills. In the Finnish population, the SLI is reported to occur in 7% of children under the age of 4 years [28].

Children with sagittal synostosis managed better in all language skills when compared with the other types of SSC (unicoronal, metopic or lambdoid) (Table 2). Narrow lexicon alone did not predict language development, although early first word acquisition and early use of sentences were positive signals for normal language acquisition. Normal, age-appropriate development of artic-

Table 2 Normative frequencies of different sub-skills

Characteristic	Sagittal	Other SSC type	Posterior	Posterior non-operated	All cases
n (%)	20 (32.79)	12 (19.67)	17 (27.87)	12 (19.67)	61 (100)
Language level					
Deviant	3 (15.00)	3 (25.00)	4 (23.53)	3 (25.00)	13 (21.31)
Poor	7 (35.00)	5 (41.67)	4 (23.53)	2 (16.67)	18 (29.51)
Normal	10 (50.00)	4 (33.33)	9 (52.94)	7 (58.33)	30 (49.18)
Lexicon					
Deviant	2 (10.00)	2 (16.67)	1 (5.88)	1 (8.33)	6 (9.84)
Poor	3 (15.00)	2 (16.67)	2 (11.76)	1 (8.33)	8 (13.11)
Normal	15 (75.00)	8 (66.67)	14 (82.35)	10 (83.33)	47 (77.05)
Reynell Receptive Scales					
Deviant	2 (11.00)	3 (25.00)	0 (0.00)	3 (25.00)	8 (13.80)
Normal	16 (89.00)	9 (75.00)	16 (100.00)	9 (75.00)	50 (86.20)
Articulation/phonology					
Deviant	5 (25.00)	8 (66.67)	9 (52.92)	4 (33.33)	26 (42.62)
Normal	15 (75.00)	4 (33.33)	8 (47.06)	8 (66.67)	35 (57.38)
Gross motor skills					
Deviant	2 (10.00)	4 (33.33)	2 (11.76)	2 (16.67)	10 (16.39)
Normal	18 (90.00)	8 (66.67)	15 (88.24)	10 (83.33)	51 (83.61)

ulation and phonological skills were most common in sagittal synostosis.

The language profile for posterior plagiocephaly was quite similar in the operated and non-operated groups. The paired samples *t* test showed that the children's performance in Reynell Receptive and Expressive Scales were bounded up with each other ($t=-4.552$ ($df=46$), $p<0.000$). When interpreting this, one should note that Reynell Receptive Scales was assessed in 58 children, but Reynell Expressive Scales only in 47 children. The reason for the incomplete test performance was in most cases either the child's refusal or becoming overtired. In this study, oral–motor-related development was assessed on the grounds of articulatory skills and phonological development. These skills showed significant co-occurrence with gross motor development ($t=-3.581$ ($df=59$), $p<0.001$).

Table 3 presents results from binary logistic regression analysis. We found a 2.67 times higher risk for impaired language perception in children with unicoronal, metopic or lambdoid synostosis when compared with sagittal cases (Reynell Receptive Scales) and a 10.31 times higher risk when compared with the operated posterior plagiocephaly group. In posterior plagiocephaly, all operated children had normal language comprehension. In contrast, 25% of non-operated children with posterior plagiocephaly had severe problems in receptive language skills, showing a 10.31 times higher risk for this type of language delay when compared with the operated plagiocephalic subgroup.

There was a 6.00 times higher risk of defective development of articulation and phonological skills in children with unicoronal, metopic or lambdoid synostosis when compared with the group with sagittal synostosis. The respective risk was 4.00 times higher when compared with the group of non-operated posterior plagiocephaly. The risk for problems in phoneme articulation was 3.38 times higher in operated posterior plagiocephaly cases when compared with sagittal synostosis.

The language profiles were examined for all subgroups. In this study, the assessment of language level (measure 5 in Table 4) acted as a cumulative value, and for this reason it correlated at a significant level with all specific measures. Early first word acquisition had positive prognostic value for later language development. First word acquisition correlated with language level ($p<0.05$), Reynell Receptive Scales ($p<0.05$), vocabulary size ($p<0.01$) and the age when the child started to use sentences ($p<0.01$). Gross motor skills also correlated statistically with language level ($p<0.05$) and the age when the child started to communicate using sentences ($p<0.01$). Lexical skills at the age of 3 years correlated significantly with receptive skills ($p<0.01$), expressive skills ($p<0.05$) and scores for mental development ($p<0.01$).

Discussion

In our study, one half of the subjects with craniosynostosis or deformational posterior plagiocephaly demonstrated slight or severe defects in early language acquisition. These results are consistent with the previous study of Becker et al. [3] and also in line with the results of Chieffo et al. [22] in unicoronal craniosynostosis. In our experiment, the prevalence of severe language defects was three times higher than in the general Finnish population [28]. These findings suggest a notable developmental risk for SLI in children with SSC and indicate that deviant language development is observable already during early infancy.

Table 3 Risk analysis (OR) for deficiencies in receptive language skills, articulation/phonology, and gross motor skills

	Reynell Receptive Scales			Articulation/phonology			Gross motor skills		
Effect	*p* 0.3720			*p* 0.1040			*p* 0.3796		
Group	*p*	OR	95% CI	*p*	OR	95% CI	*p*	OR	95% CI
Other SSC vs. sagittal	0.328	2.667	0.37–19.06	0.025	6.000	1.25–28.84	0.119	4.500	0.68–29.81
Posterior vs. sagittal	0.403	0.258	0.01–6.19	0.086	3.375	0.84–13.55	0.863	1.200	0.15–9.57
Posterior non-operated vs. sagittal	0.328	2.667	0.37–19.06	0.613	1.500	0.31–7.19	0.521	2.000	0.24–16.67
Other SSC vs. posterior	0.140	10.31	0.46–230	0.462	1.778	0.38–8.23	0.173	3.750	0.56–25.00
Posterior non-operated vs. Posterior operated	0.140	10.31	0.46–230	0.300	0.444	0.10–2.06	0.638	1.670	0.20–13.90
Other SSC vs. Posterior non-operated	1.000	1.000	0.16–6.35	0.110	4.000	0.73–21.74	0.414	2.250	0.32–15.76

Analyses were performed with binary logistic regression analysis

Table 4 Correlations between sub-skills

	1.	2.	3.	4.	5.	6.	7.	8.	9.	10.	11.	12.	13.
1. Group	1.000	−0.237	−0.180	0.514**	0.003	0.109	−0.112	−0.028	0.087	−0.090	−0.082	−0.022	−0.033
2. Sex			−0.077	−0.082	−0.006	−0.006	0.189	0.016	0.113	0.143	0.008	−0.032	0.141
3. Testing age				0.105	0.230	0.036	0.189	0.184	−0.001	0.081	−0.106	−0.250	0.130
4. Operation age					−0.077	−0.029	0.077	0.146	−0.027	0.051	−0.207	−0.023	0.029
5. Language level						0.541**	0.603**	0.638**	0.531**	0.303*	−0.263*	0.413**	0.320*
6. Reynell Receptive							0.482**	0.866**	0.542**	0.007	−0.281*	−0.315*	−0.004
7. Reynell Expressive								0.849**	0.367*	0.022	−0.011	−0.190	0.100
8. Reynell Mental									0.426**	0.015	−0.016	−0.130	−0.028
9. Lexicon										0.024	−0.529**	−0.461**	0.046
10. Articulation/phonology											−0.123	−0.256	0.257
11. First word												0.740**	−0.181
12. First sentence													−0.363**
13. Gross motor skills													1.000

*$p<0.05$, **$p<0.01$

In most cases, language deficiency occurred in speech reception and oral–motor-related skills, including phonological development. However, many children seemed to reach normal developmental scales in expressive language. Comparison between the study subgroups showed that speech, oral–motor and/or phonological abnormalities occurred less frequently in sagittal synostosis. In the SSC group, most of the subjects were males.

A new, interesting finding was that the risk for defective language development was common in deformational posterior plagiocephaly in both operated and non-operated subjects. The risk for deviant language reception was especially high in the group of non-operated deformational plagiocephaly. Our results are consistent with the study of Panchal et al. [13], who showed severe neurodevelopmental and cognitive delays in children with plagiocephaly. Kordestani and colleagues have also reported developmental risks for defective mental and psychomotor development in plagiocephaly [14].

There are many factors that may disturb neural development in the early years of life in children with SSC. Elevated ICP and intracranial hypertension are serious risk factors before skull operation [7–9]. Many researchers have reported that even though early surgical operations alleviate the raised ICP, they do not prevent neurocognitive delays in children with SSC [10–12].

Normally developed and effective CAP is highly correlated with language learning [15, 16]. Decreased amplitudes in non-attend auditory ERPs may indicate an auditory processing dysfunction in SSC [17]. The results of our study support the same theory: defective neurobiological activity and disturbed maturation of auditory pathways might disturb the acquisition of speech and language. The risk for early deficiencies in language learning is important to notice in children with single suture craniosynostosis and also in children with deformational posterior plagiocephaly.

Conclusions

These findings suggest that there is a notable developmental risk for SLI in children with nonsyndromic single suture craniosynostosis, and that the deviant language development is observable already in early infancy. The language deficiency occurred in most cases in language receptive and oral–motor-related skills. Contrary to previous beliefs, the developmental risk for defective language development in deformational posterior plagiocephaly was found in both operated and non-operated subjects. The risk for deviant language reception was especially high in non-operated deformational plagiocephaly.

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