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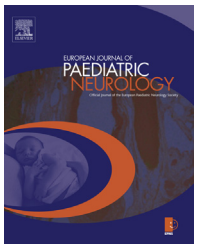

Original article

Language comprehension in nonspeaking children with severe cerebral palsy: Neuroanatomical substrate?

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ABSTRACT

Background and aims: To identify relations between brain abnormalities and spoken language comprehension, MRI characteristics of 80 nonspeaking children with severe CP were examined.

Methods: MRI scans were analysed for patterns of brain abnormalities and scored for specific MRI measures: white matter (WM) areas; size of lateral ventricles, WM abnormality/reduction, cysts, subarachnoid space, corpus callosum thinning and grey matter (GM) areas; cortical GM abnormalities, thalamus, putamen, globus pallidus and nucleus caudatus and cerebellar abnormalities. Language comprehension was assessed with a new validated instrument (C-BiLLT).

Results: MRI scans of 35 children were classified as a basal ganglia necrosis (BGN) pattern, with damage to central GM areas; in 60% of these children damage to WM areas was also found. MRI scans of 13 children were classified as periventricular leukomalacia (PVL) with little concomitant damage to central GM areas, 13 as malformations and 19 as miscellaneous. Language comprehension was best in children with BGN, followed by malformations and miscellaneous, and was poorest in PVL. Linear regression modelling per pattern group (malformations excluded), with MRI measures as independent variables, revealed that corpus callosum thinning in BGN and parieto-occipital WM reduction in PVL were the most important explanatory factors for poor language comprehension. No MRI measures explained outcomes in language comprehension in the miscellaneous group.

Conclusions: Comprehension of spoken language differs between MRI patterns of severe CP. In children with BGN and PVL differences in language comprehension performance is

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attributed to damage in the WM areas. Language comprehension was most affected in children with WM lesions in the subcortical and then periventricular areas, most characteristic for children with PVL.

1. Introduction

Cerebral palsy (CP) is caused by brain damage in the foetal or early infancy period and is characterised by limitations in mobility, often accompanied by co-occurring problems with sensation, cognition and expressive and/or receptive communication.[1,2] Language development, crucial for expressive and receptive communication, is disturbed in up to 100% of children with severe CP.[3] Children with severe CP often have severe dysarthria (unintelligible speech) or anarthria (absence of speech), where severe CP is defined as levels IV or V of the gross motor function classification system (GMFCS).[4] Because of the restrictions in speech and accompanying motor impairments that hinder non-vocal communication in children with severe CP, a discrepancy may occur between expressive communication abilities and comprehension of spoken language (receptive communication ability).[5–7] That is, children with severe CP are restricted in their expressive communication abilities (nonspeaking) but they may show better or even average receptive communication abilities. Comprehension of spoken language is an important premise for continuing intellectual growth and social participation. Therefore, insight in a child's spoken language comprehension skills is important for anticipating functional communication abilities and may facilitate therapeutic and educational processes.[8]

Neuroimaging plays an important role in elucidating the aetiology of CP, with the pattern of brain abnormalities or lesions explaining the major period of pathogenesis in CP.[9–11] Neuroimaging studies have shown abnormalities in 70–90% of children with CP.[12–18] The most common patterns of brain abnormalities in children with CP are periventricular leukomalacia (PVL), basal ganglia necrosis (BGN) and malformations of the brain.[13,14,16,18] To understand the impact of these abnormalities or lesions on brain function and on compensatory mechanisms, MRI findings can help in predicting the neurodevelopmental outcome of the child with CP.[9] However, the nature and scope of the particular lesions are wide ranging and the clinical consequences depend on the topography and extent of the lesions.[9,19,20]

Other than in our preliminary study in which we demonstrated that disturbed integrity of the dorsally and ventrally located language tracts in children with severe CP corresponded with severely hampered spoken language comprehension,[21] so far no study has reported the clinical consequences of brain abnormalities or lesions with regard to spoken language comprehension in children with severe CP who cannot speak. This lack of information between patterns of brain abnormalities and spoken language comprehension in children with severe CP may be (in part) because the assessment of spoken language comprehension remains a challenge in children with severe CP, given the requirements of the currently available psychological and language tests.[6,22,23] Therefore, we developed a Computer-Based instrument for Low motor Language Testing (C-BiLLT) that meets requirements to reliably assess spoken language comprehension in children with severe CP.[23] Comprehension of spoken language was assessed by the C-BiLLT, referring to spoken phrases and sentences of increasing difficulty in vocabulary and in grammar. The C-BiLLT is specifically designed for children with severe CP as the instrument requires minimal if any motor action. The C-BiLLT is equipped with different access methods such as a 19-inch touch screen, adjustable input switches, switch activators on a flexible and bendable shaft, the child's own wheelchair head support, eye gazing or eye-gaze computer control.

The normed data i.e. standardised z-scores for children <6;6 years and age-equivalent scores for children >6;6 years, derived from a large sample of typically developing children (n = 806) of the C-BiLLT provides information on (Dutch) spoken language comprehension performance of children with severe CP relative to their peers without disabilities. The aim of the present study was to investigate the relation between patterns of brain abnormalities in children with severe CP and spoken language comprehension. For this we first prospectively assessed language comprehension in a cohort of nonspeaking children with severe CP and, then, classified the MRI brain pattern of these children. Patterns not classifiable within the range of common patterns of CP (i.e. PVL, BGN and malformations) were classified as 'miscellaneous'. Additionally, because we observed heterogeneity in the children with the different patterns of brain damage (BGN, PVL) we additionally analysed the underlying brain abnormalities in more detail and investigated the relation between brain lesions and spoken language comprehension for the MRI brain patterns of PVL, BGN, and miscellaneous. Brain malformations causing CP are the result of the interruption of migrating neurons during the process of grouping local and distant synaptic connections into cylindrical columns and layers to form the cortex.[24] Therefore, malformations constitute a different developmental sequel of the brain and were excluded from the analyses of the influence of brain lesions on spoken language comprehension. To establish the relation between brain lesions and language comprehension with regard to the patterns of PVL, BGN and miscellaneous type, individual white matter (WM) and grey matter (GM) lesions in predefined areas were analysed.

2. Materials and methods

2.1. *Participants*

In the present study, results of language testing and associations with MRI findings are described in a subset of 80 children