



Underlying mechanisms of writing difficulties among children with Neurofibromatosis type 1



Yafit Gilboa^{a,*}, Naomi Josman^a, Aviva Fattal-Valevski^b, Hagit Toledano-Alhadeff^b, Sara Rosenblum^a

^a Department of Occupational Therapy, Faculty of Social Welfare and Health Sciences, University of Haifa, Haifa 31905, Israel

^b Pediatric Neurology Unit and the Gilbert Israeli Neurofibromatosis Center (GINFC), Dana Children's Hospital, Tel Aviv Sourasky Medical Center, 6 Weizmann Street, Tel Aviv 64239, Israel

ARTICLE INFO

Article history:

Received 15 January 2014
Received in revised form 9 March 2014
Accepted 9 March 2014
Available online 30 March 2014

Keywords:

Neurofibromatosis type 1
Writing
Underling mechanisms
Cognitive planning
Language skills

ABSTRACT

Writing is a complex activity in which lower-level perceptual-motor processes and higher-level cognitive processes continuously interact. Preliminary evidence suggests that writing difficulties are common to children with Neurofibromatosis type 1 (NF1). The aim of this study was to compare the performance of children with and without NF1 in lower (visual perception, motor coordination and visual-motor integration) and higher processes (verbal and performance intelligence, visual spatial organization and visual memory) required for intact writing; and to identify the components that predict the written product's spatial arrangement and content among children with NF1. Thirty children with NF1 (ages 8–16) and 30 typically developing children matched by gender and age were tested, using standardized assessments. Children with NF1 had a significantly inferior performance in comparison to control children, on all tests that measured lower and higher level processes. The cognitive planning skill was found as a predictor of the written product's *spatial arrangement*. The verbal intelligence predicted the written *content* level. Results suggest that high level processes underlie the poor quality of writing product in children with NF1. Treatment approaches for children with NF1 must include detailed assessments of cognitive planning and language skills.

© 2014 Elsevier Ltd. All rights reserved.

1. Introduction

Neurofibromatosis type 1 (NF1) is an autosomal dominant genetic disorder that affects multiple systems in the human body, including the peripheral and central nervous systems (Lehtonen, Howie, Trump, & Huson, 2013) with an approximate incidence of 1 in 3000. There is a broad spectrum of phenotypic expression in individuals with NF1 that is characterized by café-au-lait spots, multiple neurofibromas, optic gliomas and bone deformities. Moreover, cognitive impairment, learning disabilities and behavioral problems are the most common complications in childhood (Braddock, Kapp-Simon, & Stein, 2011).

Studies have shown that individuals with NF1 have cognitive and perceptual deficits in both language and visuospatial domains. However, visual perception impairment has long been considered a hallmark feature of the disorder with the

* Corresponding author. Tel.: +972 52 3362230; fax: +972 3 9364367.

E-mail addresses: yafitlaytman@yahoo.com, yafitgilboa@yahoo.com (Y. Gilboa), naomij@research.haifa.ac.il (N. Josman), afatal@post.tau.ac.il (A. Fattal-Valevski), drtoledano@gmail.com (H. Toledano-Alhadeff), rosens@univ.haifa.ac.il (S. Rosenblum).

Judgment of Line Orientation test most often cited as showing this deficit in NF1. Moreover, children with NF1 have motor coordination and visual-motor integration difficulties (Levine, Materek, Abel, O'Donnell, & Cutting, 2006). The difficulties with these skills in children with NF1 may interfere with writing competency (Gilboa, Josman, Fattal-Valevski, Toledano-Alhadeef, & Rosenblum, 2010).

Writing is an important task learned during early school years and is indispensable for participation in school activities (Bumin & Kavak, 2010). Two separate components of writing were identified: transcription processes skills (handwriting and spelling) and composition (content and quality of writing) (Berninger, Nagy, & Beers, 2011). Proficient handwriting has been considered a prerequisite for later academic achievement (Volman, van Schendel, & Jongmans, 2006) such as story writing, reading, comprehension, and mathematics (Bumin & Kavak, 2010). Writing is a higher-order skill, demanding coordination of multiple processes simultaneously (Kim, Otaiba, Sidler, & Gruelich, 2013). Recent advances in the understanding of writing problems have identified components of the underlying mechanism (Cheng-Lai, Li-Tsang, Chan, & Lo, 2013; Rosenblum, 2013). It was found that this complex skill requires adequate performance in visual-motor coordination, motor planning, cognitive and perceptual skills, language and tactile and kinesthetic sensitivities (Bumin & Kavak, 2010; Shen, Lee, & Chen, 2012). Since writing involves the interaction of several cognitive and motor processes, it is highly sensitive to neurological disturbances (Kushki, Schweltnus, Ilyas, & Chau, 2011).

Research about writing among children with NF1 is relatively scarce. Our previous study was the first to describe the handwriting performance among children with NF1 in comparison to children with Typical Development (TD) (Gilboa et al., 2010). The study used outcome measures to evaluate both the handwriting process (the mechanical aspects of writing tasks) and two aspects of the written product (legibility and content). The handwriting *legibility* was evaluated by administering the *Hebrew Handwriting Evaluation (HHE)* (Erez & Parush, 1999) and the product's *content* was assessed with the *Six-Trait Writing Method* (Spandel, 2004).

The results of this study indicated that regarding the handwriting *legibility*, the performance of children with NF1 was significantly poorer in comparison to TD in the *spatial arrangement* of their written product: the vertical alignment of letters, the spacing of words and letters, and letter size (HHE variable). Furthermore, their performance was also significantly poorer in the level of the *content* as was measured by the total score of the *Six-Trait Writing Model* in comparison to TDs (as can be seen in Fig. 1) (Gilboa et al., 2010). Following those results, the unique profile of children with NF1 and the complexity of writing, further questions regarding the factors affecting writing skill among children with NF1 were raised.

According to Volman et al. (2006), the components which are required for the writing production were divided into lower-level perceptual-motor processes and higher-level cognitive processes that continuously interact. Lower level perceptual motor processes in writing consist of perception of *either visual or auditory information, fine motor coordination and visual-motor integration*. The higher-level cognitive processes involved in writing include *cognitive planning or working memory* and more specific *language processes* (Volman et al., 2006).

Evidence is accumulating that the writing legibility which was described in Volman's theory, also has an important role in written composition and implies the written content (Medwell & David, 2014). The current research has been enlarged Volman's theory beyond the legibility including the written composition quality of the content. Hence, the aim of the present study is to identify the underline factors that may possibly explain the writing difficulties among children with NF1 and matched controls.

The following research assumptions were addressed: (A) there would be significant differences between children with and without NF1 regarding (1) the lower level processes (i.e., visual perception, motor coordination and visual-motor integration); and (2) the higher level process (verbal and performance intelligence, visual-spatial organization and visual

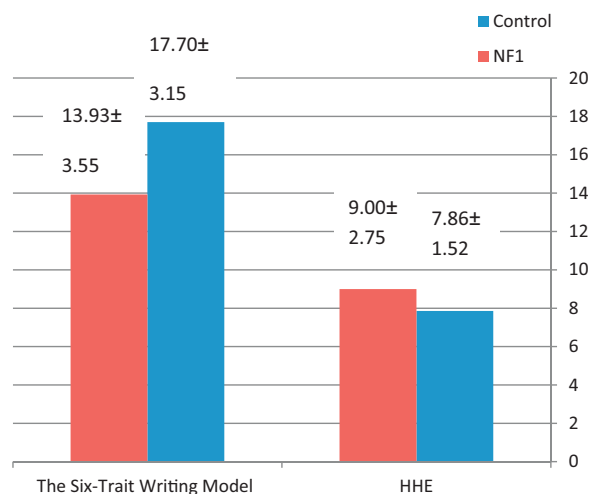


Fig. 1. Differences between the groups for the spatial arrangement and the level of content. Note: Six Trait → score indicates good performance. HHE → score indicates poor performance.

Download English Version:

<https://daneshyari.com/en/article/371287>

Download Persian Version:

<https://daneshyari.com/article/371287>

[Daneshyari.com](https://daneshyari.com)