Research in Developmental Disabilities 36 (2015) 303–310

Contents lists available at ScienceDirect

Research in Developmental Disabilities

# Persistence of visuo-constructional and executive deficits in adolescents after open-heart surgery

Michael von Rhein [a,c,*], Julietta Kugler [a], Rabia Liamlahi [a,b], Walter Knirsch [b,d], Beatrice Latal [a,d], Liane Kaufmann [e]

[a] Child Development Center, University Children's Hospital, Steinwiesstrasse 75, 8032 Zurich, Switzerland
[b] Division of Congenital Cardiovascular Surgery, University Children's Hospital Zurich/Children's Reseach Center, Switzerland
[c] Department of Pediatrics, Kantonsspital Winterthur, Brauerstr. 15, 8401 Winterthur, Switzerland
[d] Childrens Research Center, University Children's Hospital, Steinwiesstrasse 75, 8032 Zurich, Switzerland
[e] Department of Psychiatry and Psychotherapy A, General Hospital, Milser Straße 10, 6060 Hall in Tyrol, Austria

ARTICLE INFO

*Article history:*
Received 23 May 2014
Received in revised form 13 October 2014
Accepted 15 October 2014
Available online

*Keywords:*
Congenital heart defect
Open-heart surgery
Adolescent
Visuo-constructional
Rey figure
ROCFT

ABSTRACT

*Aim:* This study assesses whether previously reported performance deficiencies in visuo-constructional and executive functions, using the Rey-Osterrieth Complex Figure Test (ROCFT) in pediatric patients with congenital heart disease (CHD), persist into adolescence.
*Methods:* 53 adolescent CHD patients (mean age 13.7) and 39 healthy controls (mean age 14.1) participated. ROCFT performance was measured by three different scoring methods, focusing either on quantitative (Meyers & Meyers, 1995), qualitative (Wallon & Mesmin, 2009), or both performance aspects (Bernstein & Waber, 1996). Potential confounders (i.e., intelligence and visuomotor integration) and surgery-related risk factors were included in the data analysis.
*Results:* Adolescents with CHD demonstrated immature copy and recall approaches on the ROCFT using the qualitative system by Wallon and Mesmin ($p < .001$). Memory performance was also predicted by Bernstein and Waber scores ($p < .03$), whereas group differences were not significant according to the other scoring methods. Intelligence and visuomotor skills, but not surgery-related risk factors, were positively correlated with ROCFT performance (each $p < .02$). Interpretation: Visuoconstructional and executive deficiencies could be found in adolescent patients with CHD. However, not all ROCFT scoring methods were equally apt to detect group differences: especially the qualitative scoring method developed by Wallon and Mesmin seems sufficiently sensitive to detect long-lasting visuo-constructional and executive deficiencies in CHD patients.


## 1. Introduction

Severe congenital heart disease (CHD) occurs in six out of thousand live born children (Hoffman & Kaplan, 2002). Thereof, about 50% require cardio-pulmonary bypass surgery. Major improvements in surgical procedures and techniques have led to a significant decrease in mortality and improved cardiac outcome (Nieminen, Jokinen, & Sairanen, 2007). Nonetheless,

* Corresponding author at: Child Development Center, University Children's Hospital of Zurich, Steinwiesstrasse 75, CH-8032 Zurich, Switzerland. Tel.: +41 44 266 77 51/52 266 3708; fax: +41 44 266 71 64.
*E-mail address:* Michael.vonRhein@kispi.uzh.ch (M. von Rhein).

http://dx.doi.org/10.1016/j.ridd.2014.10.027
0891-4222/

children with CHD who have undergone open-heart surgery are at risk for developmental and neuropsychological deficits in several domains. Previous investigations have revealed mild to severe neurological impairments and motor deficits (Hovels-Gurich, Seghaye, Dabritz, Messmer, & von Bernuth, 1997; Limperopoulos et al., 2002; Majnemer et al., 2006), intellectual abilities in the low-average range (Forbess et al., 2002; Hovels-Gurich et al., 1997), and specific cognitive deficits (Bellinger, Wypij, et al., 2003; Brosig, Kuhn, & Tweddell, 2007) including visuomotor, visuospatial (Bellinger, Bernstein, Kirkwood, Rappaport & Newburger, 2003; Kirshbom et al., 2005; Majnemer et al., 2008) and executive dysfunctions (Majnemer et al., 2008). For the examination of visuospatial skills Bellinger, Bernstein, et al. (2003) used the Rey-Osterrieth Complex Figure Test (ROCFT) in 8-year old children with transposition of the great arteries (Rey, 1941). They reported poor ROCFT performance in their cohort of 8-year-old CHD patients: 52% of the children in their cohort achieved the lowest level on a five-category clinical rating in the copy task (Bernstein & Waber, 1996). Interestingly, these particular deficits were no longer present in the same cohort at 16 years of age, even though visuo-spatial deficits were detected when other tests were used (Bellinger et al., 2011). However, the respective literature is somewhat inconsistent: While some authors described below-average visual motor integration scores in 21.4% of 3–8-year-old children with severe CHD that were unrelated to intellectual abilities (Uzark et al., 1998), others did not find significant impairments regarding visual-spatial skills in 6–12-year-old CHD patients in a design copy test (Miatton, De Wolf, Francois, Thiery, & Vingerhoets, 2007).

### 1.1. Scoring systems of the Rey-Osterrieth Complex Figure Test (ROCFT)

The ROCFT is a standardized test requiring participants to copy and reproduce a complex abstract figure composed of many details. Currently, different scoring methods are applied that either focus on quantitative (i.e., presence or absence of figural elements) (Meyers & Meyers, 1995) or qualitative performance aspects (i.e., drawing approaches) (Wallon & Mesmin, 2009) or both (Bernstein & Waber, 1996). Generally, the copy condition ought to assess visuoconstructional, visuospatial, planning/organizational and graphomotor skills, while the recall condition additionally assesses visual memory capacities (Ruffolo, Javorsky, Tremont, Westervelt, & Stern, 2001). To the best of our knowledge, so far only one study has compared different scoring systems of the ROCFT in a clinical sample: Frank and Landeira-Fernandez (2008) found performance differences on the ROCFT between patients with left versus right temporal lobe epilepsy upon using a qualitative scoring system, while quantitative scoring approaches failed to detect significant group differences.

### 1.2. Aims and hypotheses

The aims of the present study were threefold: *First*, we hypothesized that adolescent patients with CHD would perform poorer on the ROCFT (regarding executive, visuoconstructional and visual memory functions) compared with age-matched healthy controls. *Second*, we aimed to compare different scoring methods of the ROCFT. While the quantitative scoring system (Meyers & Meyers, 1995) should be especially useful to detect visuoconstructional and visuospatial performance aspects, qualitative scorings (Bernstein & Waber, 1996; Wallon & Mesmin, 2009) should be superior in assessing executive control processes (i.e., planning/organizational skills). *Finally*, correlations with other cognitive domains and surgery-related risk factors will be performed.

## 2. Materials and methods

### 2.1. Participants

The present study is part of a longitudinal cohort study documenting neurodevelopmental outcomes of children with different types of CHD who underwent full-flow bypass surgery at the University Children's Hospital Zurich between 1995 and 1998 (mean age at first surgery: $1.4 \pm 1.4$ years). Out of the initial sample of 117 children examined at a mean age of 10.4 years (Landolt, Valsangiacomo Buechel, & Latal, 2008; von Rhein, Valsangiacomo Buechel, Landolt, & Latal, 2012), 87 participants were eligible for the follow-up examination at 14 years (74.4%). Exclusion criteria were a diagnosis of chromosomal or genetic syndromes, congenital and acquired neurological diseases (i.e., congenital rubella syndrome or neurofibromatosis) and an age older than 17 years at the current follow-up examination. 23 adolescent patients refused participation and five further patients were lost to follow-up. Of the remaining 59 adolescents six were excluded as they had a full-scale IQ below 85 as indexed by the current German version of the Wechsler Intelligence Scale for Children (WISC-IV (Petermann & Petermann, 2010)). Thus, the final sample of study participants consisted of 53 adolescents with CHD (for sample characteristics see Supplementary Table). Though demographic and surgical characteristics were not significantly different between our patients and those lost to follow-up, our patients represented a subgroup with fewer neurological abnormalities (33 versus 60%, $p < .001$) and higher IQ (107/SD 11 versus 86/SD 14, $p < .001$) compared to those lost to follow-up.

Supplementary Table related to this article can be found, in the online version, at http://dx.doi.org/10.1016/j.ridd.2014.10.027.

The control group consisted of 39 healthy adolescents (19 males/20 females) closely matched according to age, sex and socio-economic status (Table 2). All controls went to regular school and did not suffer from any chronic or neurological disease.