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Behavioral Development is better after Early Repair of Tetralogy of Fallot

Manuel CHIRA[1], Dana Florica CIOTLAUS[2], Adrian STEF[3], Olivia VASILE[4], Grigore TINICA[5], Adrian MOLNAR[6]

Abstract

Psychological development of patients with surgically corrected Tetralogy of Fallot could be affected by the chronic preoperative cerebral hypoxia; therefore early repair is beneficial for the outcome of these patients. 71 Tetralogy of Fallot patients were operated in Heart Institute between September 1st, 2001 and July 1st, 2006, all of them underwent corrective surgery, without prior palliations. Corrective surgery consisted of: transannular patch (46), infundibular patch ± PA patch (17), and transatrial and transpulmonary correction (8). The patients were divided into 2 groups, correction below age of 1 year and above 1 year, for comparative study of results. 13 patients were excluded for various reasons, and 58 patients were followed up, at 82.66±17.269 months, performing a Child Behavior Check List test. All the components of CBCL test were studied comparatively between the 2 groups: affectivity, anxiety, somatic, ADHD, opposing behavior and conduct problems, as absolute values and divided into normal, subclinical and clinical levels. Major differences were noticed between the two groups, as followed: affectivity and anxiety values (absolute values and normal, subclinical and clinical levels) were higher in patients operated after 1 year of age. Interestingly, affective and anxiety problems and opposing behavior were also correlated (statistically significant) with the preoperative hematocrit. Delay in surgical correction beyond 1 year of age in patients with Tetralogy of Fallot could have deleterious effects over psychological development (affective, anxiety, somatic, ADHD, opposing behavior and conduct problems) of these patients, due to longer period of preoperative chronic cerebral hypoxia.

Keywords: Tetralogy of Fallot, child behavior check list, psychological testing; TOF early repair.

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Introduction

Tetralogy of Fallot (TOF) is the most common cyanogenic congenital heart disease (CHD) and a milestone in pediatric cardiac corrective surgery, for every surgeon and for every growing pediatric cardiac center. Thus, Tetralogy of Fallot surgical correction was subject of major improvements during time, especially in the 80`s and 90`s. Therefore, lowering the age and weight of patients at the time of correction was possible, avoiding in this manner the deleterious effect of prolonged preoperative hypoxia on the brain and other organs. Prolonged preoperative hypoxia could have negative impact on postoperative cognitive and behavioral development of TOF patients. There are very few papers in international literature studying behavioral development in patients with surgically corrected TOF. Most of these papers study psychological development of corrected TOF patients, with no regard to surgical timing. Thus the aim of this paper tries to fill a gap in the international research, regarding psychological development of TOF patients related to the timing of corrective surgery.

Materials and Methods

A group of 77 patients was subjected to surgery in Heart Institute, Cluj Napoca, Romania, between September 1st, 2001 and July 1st, 2006. All patients were surgically corrected per prim am, without prior palliations, with 2 deaths (2.6% mortality). 4 older patients at repair (14, 18, 21, 32 years) were excluded from the initial group, trying to reduce the influence on statistical data. After the exclusion of older patients and mortality group, we obtained a homogeneous group of 71 patients. The 71 patients were split into 2 groups, depending to operation age: group 1 (25 patients), corrected below age of 1 year, and group 2 (46 patients), corrected above age of 1 year (*Table 1*). We thereby divided the study group considering a lot of studies emphasizing postoperative issues in patients with late TOF repair (Katz *et al*., 1982, Lillehei *et al*., 1986, Kavey, Blackman, & Sondheimer, 1982, Niwa *et al*., 2002, Marie *et al*., 1992, Garson *et al*., 1985, Dean & Lab, 1989, Kirklin *et al*., 1989). The surgical corrections employed in the total group of 71 patients were: transannular patch (46 patients), infundibular patch ± PA patch (17 patients), transatrial and transpulmonary repair (8 patients). 58 patients were followed-up for 82.66±17.26 months. Among these 58 patients, 21 were in group 1 (surgery below age of 1 year), and 37 were in group 2 (surgery above age of 1 year). 13 patients were not followed-up due to: 1 patient with Down syndrome, 2 patients with postoperative stroke, 4 patients with preoperative neurologic injury (stroke or other issues), 2 institutionalized patients, and 4 patients lost at follow-up.

The parents of patients in the followed-up group completed a CBCL test (Achenbach *et al*., 1983). The CBCL test evaluated affective, behavioral and temperamental dimensions. A prior psychological evaluation (3 years earlier)

was performed on 58 patients, but the results of that examination were the aim of another study (Chira *et al.,* 2017: 78-88). After the completion of the CBCL test by the parents, we analyzed comparatively the values of affectivity, anxiety, somatic, ADHD, opposing behavior and conduct problems over the 2 age groups and correlation between these values and preoperative hematocrit values. The normal, subclinical and clinical levels of CBCL dimensions were studied comparatively on the 2 age groups. The statistical analysis was performed using IBM SPSS v.19. When we compared means, we used T test for independent samples, with Monte Carlo test for p value computation. The correlation between CBCL values and hematocrit values was performed using regressions.

Results

Absolute values of affectivity, anxiety, somatic complaints, ADHD, opposing behavior and conduct problems were studied comparatively on the 2 age groups.

Table 1: Mean values and standard deviation of CBCL test variables

CBCL VARIABLE	Age at surgery	No.	Mean	Standard deviation
Affectivity problems	< 1 year	21	2.81	2.294
	>1 year	37	6.68	5.164
Anxiety problems	< 1 year	21	2.48	2.522
	>1 year	37	4.92	3.086
Somatic complaints	< 1 year	21	1.33	1.683
	>1 year	37	1.59	1.363
ADHD problems	< 1 year	21	3.24	3.673
	>1 year	37	6.05	3.383
Opposing behavior	< 1 year	21	1.76	1.338
	>1 year	37	3.46	2.049
Conduct problems	< 1 year	21	1.29	1.146
	>1 year	37	2.73	2.156

Difference between mean values of affectivity problems is statistically significant ($p<0.05$).

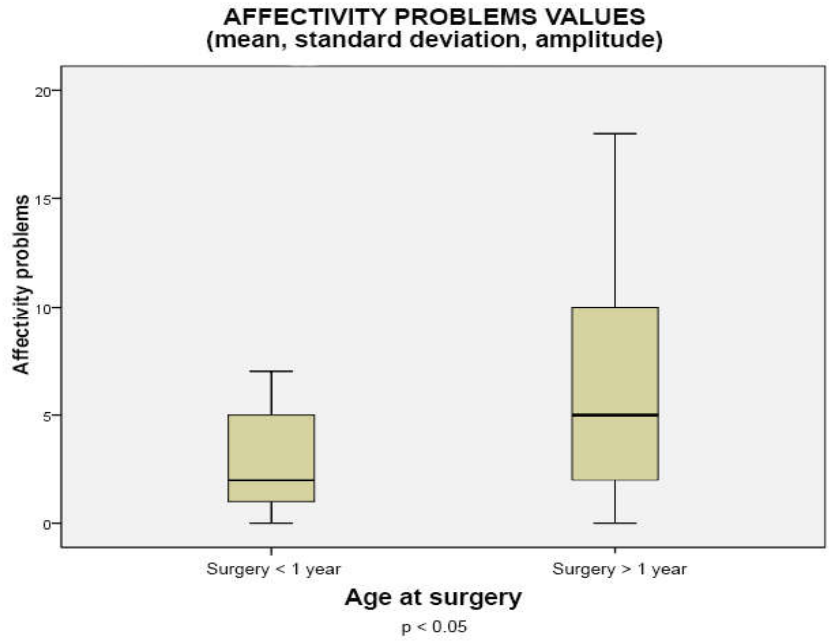

Figure 1: Affectivity problems values in the 2 age groups

Difference between mean values of anxiety problems is statistically significant (p<0.05).

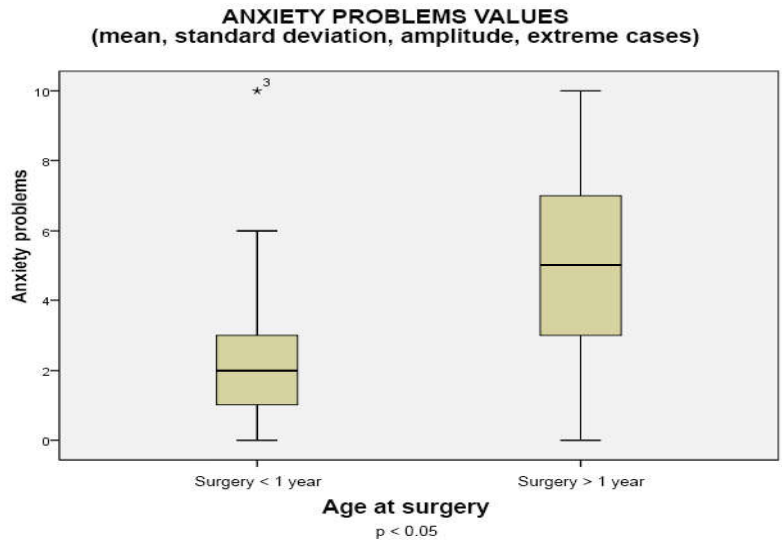

Figure 2: Anxiety problems values in the 2 age groups

Difference between mean values of somatic complaints is not statistically significant. Regarding ADHD, opposing behavior and conduct problem we found statistically significant differences ($p<0.05$) between mean values amongst both age groups, in all three elements of CBCL test. When postoperative affectivity problems and preoperative hematocrit values were analyzed in regression, we obtained a statistically significant positive correlation (affectivity problems corresponded to high hematocrit values).

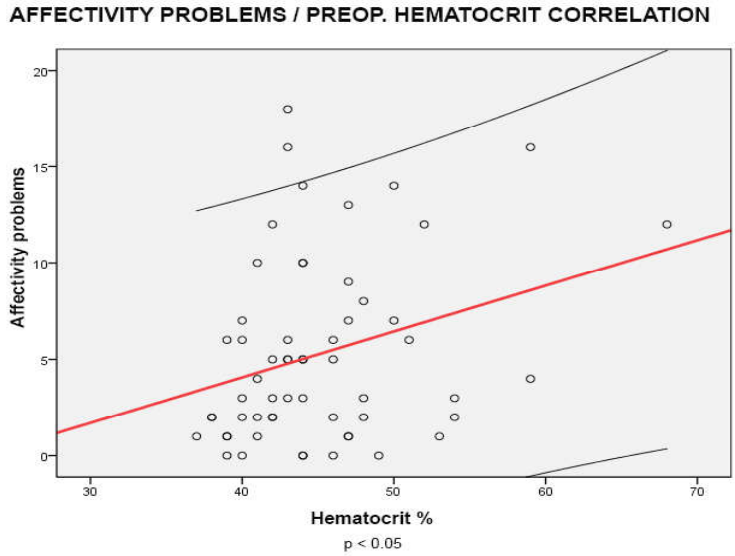

Figure 3: Correlation Affectivity problems/Preoperative hematocrit (severity of hypoxia)

Anxiety and opposing behavior problems were correlated in regression with preoperative hematocrit values, obtaining a statistically significant positive correlation (anxiety and opposing behavior problems corresponded to high hematocrit values) ($p<0.05$). In respect to postoperative somatic complaints, ADHD and conduct problems, no statistically significant correlation were obtained. The variables studied in CBCL test can be analyzed as nominal values or ordinal values (normal, subclinical, clinical).

Table 2: Normal, subclinical and clinical levels of CBCL variables

CBCL VARIABLE	Age at surgery	Normal level %	Subclinical level %	Clinical level %
Affectivity problems	<1 year	81	19	0
	>1 year	48.6	16.2	*35.1*
Anxiety problems	<1 year	81	14.3	4.8
	>1 year	40.5	24.3	*35.1*
Somatic complaints	<1 year	71.4	14.3	14.3
	>1 year	56.8	37.8	5.4
ADHD problems	<1 year	85.7	4.8	9.5
	>1 year	67.6	24.3	8.1
Opposing behavior	<1 year	95.2	4.8	0
	>1 year	67.6	*29.7*	2.7
Conduct problems	<1 year	100	0	0
	>1 year	91.9	8.1	0

Analyzing affectivity problems amongst the two age groups we found no clinical affectivity problems in group 1, as opposed to a high level in group 2 (35.1%). The subclinical level was comparable in the two groups. The normal level was notably elevated in group 1 (81%). Differences between affectivity disorders levels in the two age groups were statistically significant ($p<0.05$).

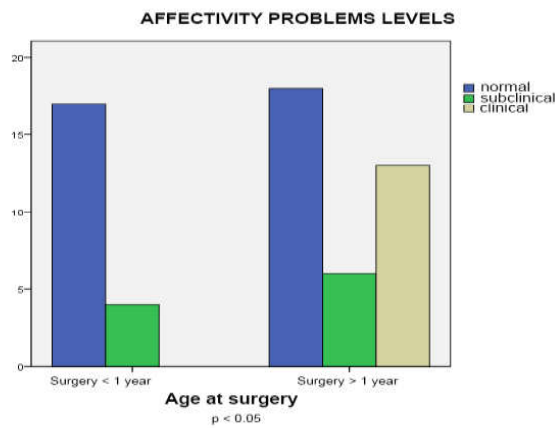

Figure 4: Levels of affectivity problems on the 2 age groups

Anxiety problems levels also differ in the two age groups. Normal level is higher in group 1 (81%), subclinical level is higher in group 2 (24.3% as opposed to 14.3%). Clinical level is raised in group 2 (35.1% compared to 4.8%). Differences between anxiety problems levels in the two age groups were statistically significant ($p<0.05$).

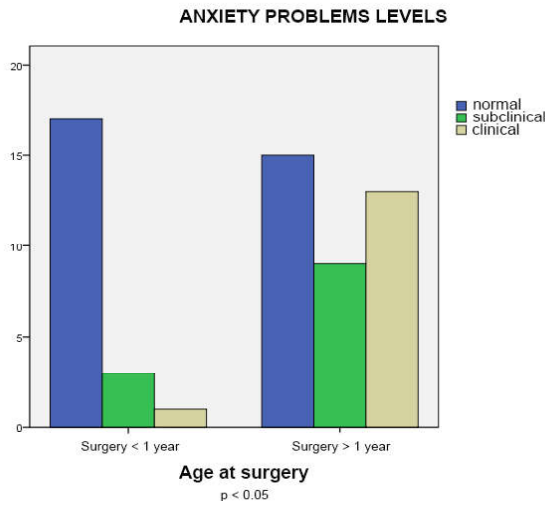

Figure 5: Levels of anxiety problems on the 2 age groups

When it comes to somatic complaints, the differences between the two age groups were minor and not statistically significant. Generally, we observed a slight prevalence of the normal level in group 1, minor predominance of the subclinical level in group 2 as well as a mild dominance of the clinical level in group 1. The differences are, once again, not statistically significant. ADHD problems levels are not significantly different in the two age groups. The clinical level is roughly equal, a minor prevalence of the subclinical level in group 2 could be observed (24,3% compared to 4,8%)and, regarding the normal level differences, they were relatively small. The differences are not statistically significant.

Opposing behavior is different between the two age groups. The normal level is predominant in group 1 (95.2%) while the subclinical level in group 2 (29.7% compared to 4.8). The clinical level could be observed in only one case belonging to group 2. Differences between opposing behavior levels in the two age groups were statistically significant ($p<0.05$). The levels of conduct problems are not statistically different among the two groups. We did not find any clinical level case. However, we encountered 3 subclinical level patients belonging to group 2.

Discussions

Many studies have revealed the involvement of general and cerebral hypoxia in postoperative cognitive and behavioral development or patients (Oates *et al*., 1995; Wray & Sensky, 2001). The first element is the *severity of hypoxia*. From this point of view, our study has a relatively homogenous constitution ensured by exclusion of emergency cases with unacceptable hypoxia. Thus, O_2 saturations range between 71% and 91% for group 1 with mean $m_1 = 81.43\%$ and 70%-90% for group 2 with mean $m_2 = 84.41\%$, difference between m_1 and m_2 being statistically insignificant.

Another important element related to psychological development is *duration of hypoxia*. This aspect is essential in our study since it analyzes differences between two groups of patients undergoing surgery at different ages, considering that preoperative hypoxia extension can affect cerebral function and subsequently patient's psychological development (Nyakas, Buwalda, & Luiten, 1996; Miatton *et al*., 2007). Hypoxia span influences the preoperative hematocrit by increasing its value in the context of chronic oxygen deprivation. Therefore, our study analyzes the effect of preoperative hypoxia duration on postoperative psychological development by correlating the hematocrit with the assessed psychological dimensions.

We conducted a descriptive analytic statistic of the data obtained by quantitative assessment of the following dimensions: affectivity problems, anxiety problems, somatic complaints, ADHD problems, opposing behavior and conduct problems for the two age groups. We compared the two groups via the T test for independent samples. Regression correlation was performed for expository purpose (Reiss, & Judd, 2009), between hematocrit (independent variable) and psychological dimensions (dependent variable). We seek any relationship linking medical factors and psychological development, this type of correlation being suitable for statistical control of other factors' influence on dependent variables.

By descriptive statistics analysis of the data obtained from CBCL and inters group comparison, we displayed differences in development of affective, behavioral and social functioning levels increasingly higher as corrective surgery is postponed. Regression correlations of medical and psychological dimensions concluded that psychological development is associated with the duration of preoperative hypoxia. Studying the distribution of development disturbances levels with CBCL test we concluded there are major differences between the two age groups when speaking about affectivity and anxiety problems. The distribution of these values suggests the effect of preoperative hypoxia upon affectivity and anxiety in postoperative period. Contributing to these elements is a series of factors such as family, parents' educational level and their concern about the development of a child with congenital cyanotic heart disease. Parental overprotection is studied in numerous international articles, factor which could negatively influence affectivity

and anxiety problems of the child with Tetralogy of Fallot (Wray & Sensky, 1999; Landtman *et al*., 1960; Linde *et al*., 1966; Offord *et al*., 1972).

Behavioral alterations of the parents can be secondary to the difficulty of the clinical condition of the child (Gudermuth, 1975; D'Antonio, 1976; Shor, 1978), or repetitive hospital admissions prior to correction (especially between 6 months and 4 years of age) (Wray & Sensky, 1999; Prugh *et al*., 1953; Vernon, Schulman, & Foley, 1966). Conduct problems fluctuate less, according to the scores we obtained and the distribution of the values in the two age groups. These behavioral elements could be affected by hypoxia to a smaller extent; however they are likely influenced by parents' behavior and the comprehension of the cardiac disease (Landtman *et al*., 1960; Linde *et al*., 1966; Offord *et al*., 1972; DeMaso *et al*., 1990).

Somatic complaints and ADHD problems vary less amongst the two age groups. Regarding somatic complaints, we could say they are conditioned on the physiology of the individual. On the other hand, when it comes to ADHD, our study does not analyze enough elements to enable any correlation between ADHD and preoperative physiology or family environment. Inspection of opposing behavior levels reveals differences between the two age groups. We did not encounter any clinical level case.

The results of the CBCL test were correlated with preoperative hematocrit to determine which behavioral element is mostly affected by long term preoperative hypoxia. Affectivity, anxiety problems and opposing behavior have a positive correlation to preoperative hematocrit with statistical significance, meaning the higher the preoperative hematocrit, the bigger the problems. We observed a positive correlation between the other CBCL test elements (somatic complaints, ADHD and conduct problems) and preoperative hematocrit, but not of statistical significance.

Conclusion

Mean value of affectivity, anxiety, opposing behavior, ADHD and conduct problems have a statistically significant difference between the two age groups, with smaller values belonging to patients undergoing corrective surgery under the age of 1. Levels of affectivity and anxiety problems, as well as opposing behavior, have a positive correlation with the value of preoperative hematocrit. Normal, subclinical and clinical levels of affectivity and anxiety problems and opposing behavior are significantly different between the two age groups, with subclinical and clinical levels being higher in patients undergoing correction after the first year of age.

Study limitations

This study's limits reside in the relatively short distance between the surgical correction and the moment performing the CBCL test. These patients have

been observed during infancy and adolescence and additional adult follow up is necessary for extensive deductions of further development.

Acknowledgements

Another paper entitled Early Repair Benefits in Cognitive Development of Patients with Tetralogy Of Fallot was published in *Revista de Cercetare si Interventie Sociala,* 2017, 57: 78-88 based on the same 77 group of patients. The aim of that study was cognitive development. The aim of study in this paper is behavioral development.

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