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School-age human figure drawings by very preterm infants: validity of the Draw-a-Man test to detect behavioral and cognitive disorders.

Short title: School-age human figure drawings by very preterm infants

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ABSTRACT

Detecting an abnormal developmental trajectory in very preterm infants remains challenging. The objective of this study was to determine the correlation between the Draw-a-Man test (DAMT) and behavioral and cognitive disabilities in very preterm infants. From the school-age follow-up of the Premag study, which evaluated the neuroprotective effect of prenatal magnesium sulfate before 33 weeks of gestation, 281 human figure drawings were assessed (mean age, 11 years). Behavioral and cognitive disabilities were associated with delayed DAMTs but test performance indicators were insufficient to use DAMT as a screening or a diagnostic test.

Key words: very preterm infants; magnesium sulfate; human figure drawings; Draw-a-Man test; behavioral disorder; cognitive disorder.

1 INTRODUCTION

Clinical follow-up of very preterm infants has become a major current and relevant practice in neonatal units. Infants born very preterm are at high risk of developing cognitive and motor impairments, and this risk is inversely correlated with gestational age [1]. Early identification of motor, cognitive, and behavioral impairment is a delicate challenge for pediatricians wishing to initiate an early intervention to reduce further developmental delay and handicap. Screening instruments for the follow-up of infants born very preterm have to be simple to use, quick to execute, inexpensive, and easy to interpret by the developmental pediatrician or pediatric psychologist. Many screening tests are available including the evaluation of human figure drawings (HFDs). Indeed, HFDs become more complex and differentiated with increasing age, from the first rough drawings at 2 years to the sophisticated drawings of teenagers. HFDs could serve as a short screening tool to explore cognitive problems in childhood. Among HFD tests, The Draw-a-Man test (DAMT) was originally developed by Goodenough to estimate children's intellectual capacities [2].

In the context of a school-age follow-up of the French PREMAG randomized controlled trial, which evaluated the neuroprotective effect of prenatal magnesium sulfate in very preterm infants born before 33 weeks of gestation (WG), children were asked to draw an HFD [3]. HFDs made by the PREMAG children were analyzed using the DAMT methodology. In light of the results of the school-age outcomes, we first hypothesized that DAMT may be relevant to detect cognitive and behavioral impairments. Therefore, we preliminarily compared the DAMT of the placebo and magnesium sulfate groups to ensure of the homogeneity of the cohort; then we assessed a correlation between DAMT and behavioral and cognitive impairments in the overall PREMAG cohort.

2 OBSERVATIONS

2.1 Methods 2.1.1 Initial study PREMAG was a prospective multicenter randomized controlled trial that took place in 13 French perinatal centers. Between July 1997 and July 2003, women pregnant at a gestational age younger than 33 WG were eligible if birth was expected or planned within 24 h. Women received 4 g magnesium sulfate solution or isotonic 0.9% saline. The study included 564 women who gave birth to 688 infants. A parent of each child provided written informed consent. The Clinical Trials.gov Identifier was NCT00120588 [4,5].

2.1.2 School-age study

Between December 2009 and April 2012, we contacted the families of all surviving children. A mail questionnaire was sent to the parents, who remained blinded to the treatment group in the perinatal period. This second protocol was approved by the Comité consultatif sur le traitement de l'information en matière de recherche dans la Santé (CCTIRS) of the French Ministry of Health (17 December 2009) and the Commission Nationale de l'Informatique et des Libertés (CNIL) (19 April 2010). All questionnaires were collected and analyzed at the Neonatal Intensive Care Unit at Rouen University Hospital, with secure computers used for data entry and storage [3]. Parents gave informed consent to the research and to publication of the results.

2.1.3 Outcome measures

The main outcome was the DAMT of very preterm infants of the PREMAG cohort at school age. The objective of this study was to assess the correlation between DAMT and behavioral and/or cognitive disabilities.

2.1.4 Procedures

Parents completed a 48-item questionnaire about their child that used several neuropsychomotor development items (including the long-term follow-up EPIPAGE questionnaire [6], and the French version of the Strengths and Difficulties Questionnaire (SDQ) [7]), as well as questions about the child's general health, motor, cognitive and sensory

deficits, behavioral disorders, schooling, and family situation. We developed outcome indicators from the questionnaire to assess motor deficits, cognitive deficits/learning disabilities, psychiatric disorders, and overall deficiencies [3], and the SDQ evaluated behavioral skills. Children were asked to draw a human figure on a white page included in the parental questionnaire.

2.1.5 The Draw-a-Man test

Children were asked to draw an HFD on a piece of paper. The rating scale contains 51 items, and each item is evaluated as normal, delayed, or in advance. Two experimenters (CC, AJ) coded the drawings independently, blinded to the treatment group. Each drawing yielded a score ranging from 0 to 51 based on the number of details in the drawing and the accuracy of the placement of each body part. The average of the two scores was retained. If a difference between the two experimenters' scores was greater than 20%, a third analysis was performed by them together. The score was converted into a theoretical age according to the guidelines set out in the DAMT manual [2]. The score was considered (1) normal if predicted age was within 1 year of chronologic age; (2) delayed if predicted age was less than chronologic age by at least 2 years; and (3) advanced if predicted age was greater than chronologic age by at least 2 years.

2.1.6 Statistical analysis

All statistical analyses were undertaken on an intention-to-treat basis. Comparison between the magnesium sulfate and placebo groups was based on a logistic model employing generalized estimating equations to account for correlations between twins or triplets. Comparisons of primary and secondary outcomes were adjusted for gestational age, singleton/multiple pregnancy, socioeconomic variables (mother's educational level, both parents' occupations), gender, and birthweight residual, which had been previously identified as significant predictors of the primary outcomes [3]. The birthweight residual was estimated from the linear regression of birthweight on gestational age in order to eliminate the colinearity between the two variables. The association between DAMT and behavioral and/or cognitive disabilities were analyzed on the entire cohort. We calculated sensitivity, specificity, positive and negative predictive values, positive and negative likelihood ratios, and pre-test and post-test probabilities. For test performance indexes, delayed DAMT was considered a positive test and normal or advanced DAMT was considered a negative test. Odds ratios (OR) and corresponding 95% confidence intervals were estimated. Statistical significance was defined as p<0.05. These analyses were performed using SAS software, version 9.2 (SAS Institute, Cary, NC, USA).

2.2 Results

2.2.1 Study participants

At school age, 185 children were lost to follow-up (26.9%) and 431 children were assessed. The mean assessment age was 11 years (range, 7–14 years). Long-term outcomes were available for 503 children (including 72 who had died), 252 in the magnesium sulfate group and 251 in the placebo group. The overall characteristics of children and mothers were broadly similar in the two groups. Nevertheless, the magnesium sulfate group included more boys and had a lower mean birth weight than the placebo group (Table 1). The main characteristics of the participants and families lost to follow-up were also similar (gender, gestational age, single or multiple pregnancy, reason for preterm birth, prenatal magnesium sulfate exposure, cerebral palsy at 2 years), except for a greater prenatal corticosteroid exposure for the participants (95.6% versus 88.6%, p=0.001). No children died between the ages of 2 and school age. Overall, 301 children drew a human figure, 152 in the magnesium sulfate group and 149 in the placebo group. Seventy-two questionnaires were collected by phone and 58 children did not answer the HFD question. The experimenters did not include 20 HFDs because they did not match the DAMT criteria. Finally, 281 HFDs (64.5% of the school-age cohort) were eligible for analysis (Figure 1). Between respondents and nonrespondents to the HFD question, no differences were identified for the main characteristics (age, sex, gestational age, single or multiple pregnancy, magnesium sulfate exposure), except for behavioral problem and prosocial scales of SDQ, and for cognitive deficits, which were more frequently abnormal among the children who did not respond (data not shown).

2.2.2 Preliminary analysis: does prenatal exposure to magnesium sulfate have an impact on DAMT results?

One hundred and forty-four children who drew a human figure were in the magnesium sulfate group, and 137 were in the placebo group. DAMT results were similar in the magnesium

sulfate group and in the placebo group; respectively, 29.8% vs. 35.0% of the HFDs were delayed and 70.2% vs. 65.0% were normal or advanced (adjusted odds ratio (aOR)=0.81, 95% CI: 0.48-1.39, p=0.45).

Considering the absence of an impact of the magnesium sulfate treatment, the entire cohort was considered for the subsequent analyses.

2.2.3 Association between DAMT and behavioral and cognitive disabilities

Among the 281 children who drew a HFD, 140 children (49.8%) were classified in the normal group, 91 (32.4%) in the delayed group, and 50 (17.8%) in the advanced group (Figure 2). Regarding the SDQ, the abnormal overall score was significantly associated with a delayed DAMT score with or without adjustment (aOR=1.94, 95% CI, 1.12–3.36, *p*=0.02) (Table 2). Based on the sensitivity and specificity analyses, we expected that 43% (95% CI, 33–55) of the very preterm children with an abnormal SDQ score would also have a delayed DAMT score while 72% (95% CI, 65–78) of the very preterm children with normal SDQ score would also have a normal DMAT result. The positive predictive value was 35% (95% CI, 26–45) and the negative predictive value was 78% (95% CI, 72–83). Behavioral problems, hyperactivity, and peer problems were significantly associated with a delayed DAMT score without adjustment, but these associations did not remain after adjustment.

Cognitive deficits and learning disabilities were also associated with a delayed DAMT score (aOR=3.43, 95% CI, 2.10–5.62, p<0.0001) (Table 2). The test sensitivity was 42% (95% CI, 35–50), specificity 82% (95% CI, 74–89), positive predictive value 78% (95% CI, 68–86), and negative predictive value 48% (95% CI, 41–56).

These tests mean that among children with an abnormal SDQ or cognitive deficits and learning disabilities, 43% and 42% had a delayed DAMT, respectively (true positives). It means otherwise that among children with a normal SDQ or without cognitive deficits and

learning disabilities, 28% and 18% had a delayed DAMT, respectively (false positives). Diagnostic test performance indicators are reported in Table 3.

3 DISCUSSION

We assessed the school-age DAMT of the children included in the randomized controlled trial PREMAG. First of all, there was no impact of magnesium sulfate for DAMT results. Considering the entire school-age cohort, a significant association was identified between delayed HFDs and behavioral and/or cognitive disabilities; however, performance indicators were insufficient to use DAMT as a screening or a diagnostic tool.

The proportion of delayed HFDs (32.4%) in our cohort is close to the rate of cognitive deficiencies of the very preterm children participating in the EPIPAGE study at school age (31%) [1]. In addition, these results are close to those of a French study that showed a correlation between scores of the Goodenough test and the K-ABC of very preterm infants at 5 years, but behavioral and cognitive disabilities were not explored and performance indicators were not provided [8].

DAMT results were statistically associated with behavioral and cognitive deficit but other deficits could affect the DAMT score such as motor dysfunction (even minor), visual deficit, attention disability, dysexecutive syndrome, dyspraxia, or dysgraphia. Other factors such as creativity, drawing experience, and personal interest for drawing may interfere with DAMT [9,10].

DAMT scores are based on the number of details in the drawing and the accuracy of the placement of each body part. These criteria may not be appropriate for assessing child development. In addition, DAMT may not be relevant for detecting behavioral and/or cognitive disabilities. Recently, Tükel et al. categorized the developmental stages of HFD (1, scribble; 2, pre-tadpole; 3, tadpole; 4, transitional; 5, conventional-I; and 6, conventional-II), analyzed the drawing order (1, top-down; 2, center-periphery; and 3, bottom-up), and objective and quantitative criteria (size, symmetry, proportion, and circularity) using an image-processing computer program (ImageJ) [11]. They demonstrated that immature HFD is

a moderate-to-good indicator of visuospatial intellectual difficulties and that fine motor control is crucial in the developmental stages of HFD.

Analysis of DAMT performance indicators showed low sensitivity for behavioral disorder (43%) and cognitive deficits/learning disabilities (42%) with quite good specificity (respectively, 72% and 82%). On the basis of the positive likelihood ratio, a patient with a delayed DAMT will be 1.52 times more likely to have a behavioral disorder compared with a patient with the same characteristics but with a normal DAMT. Finally, there was a small difference between pre- and post-test probability, 0.26 and 0.35, respectively, which suggests an absence of clinical relevance. A similar trend was observed for cognitive deficits and learning disabilities.

A number of limitations in this study should be mentioned. Firstly, the DAMT is a historical tool, which has been revised and improved several times. However, it seems appropriate for a postal questionnaire because it needs only one HFD and the instructions are very simple, while other newer tests, such as the Goodenough-Harris test or the Draw-A-Person: a Quantitative Scoring System, needs three drawings on separate pieces of paper. The child is asked to draw a man, a woman and him- or herself. This procedure may be more complex to produce by children in a context of a postal questionnaire. Secondly, 130 (30.1%) of the 431 children in the school-age cohort did not make an HFD. Among them, 72 questionnaires were collected by phone. However, the overall characteristics of respondents and non-respondents to the DAMT question were broadly similar, except for behavioral problems and the prosocial scale of the SDQ, and for cognitive deficits, which were more abnormal for those who did not respond, meaning that we cannot exclude a non-response bias in this study.

4 CONCLUSION

School-age DAMTs of very preterm infants were not significantly different in case of prenatal exposure to magnesium sulfate. HFD is a simple, feasible, and effective tool that requires a short execution time and is appreciated by children. Despite a statistically significant association between the DAMT results and behavioral and cognitive/learning disorders, performance indicators demonstrated that DAMT is not effective as a screening or diagnostic test of developmental problems frequently noted in preterm children. Future research should aim to develop other tests using the HFD as a child developmental delay screening tool, for example by using a computer program for image processing.

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Ethics approval This protocol was approved by the Comité Consultatif sur le Traitement de l'Information en Matière de Recherche dans la Santé of the French Ministry of Health.

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Table 1: Characteristics of the children and their families^a

Characteristics	MgSO ₄ group ($n=218$)	Placebo group (<i>n</i> =213)	P- value	
Children				
Gestational age, mean (SD), weeks	29.9 (2.0)	30.1 (1.9)	.31	
Less than 27	21 (9.6)	16 (7.5)		
From 27 through 29	70 (32.1)	68 (31.9) .73		
From 30 through 32	127 (58.3)	129 (60.6)		
Singleton birth	139 (63.8)	143 (67.1)	.46	
Birth weight, mean (SD), g	1397 (24)	1476 (25)	.02	
Male sex	117 (53.7)	82 (38.5)	.02	
Exposure to prenatal corticosteroids	208 (95.4)	204 (95.8)	.85	
Reasons for preterm birth ^b				
Isolated preterm labor	39 (17.9)	61 (28.6)		
PPROM	82 (37.6)	67 (31.5)		
Chorioamnionitis	58 (26.6)	58 (27.2)	.06	
Prepartum hemorrhage	28 (12.9)	22 (10.3)		
Other	11 (5.0)	5 (2.4)		
Child's age at long-term follow-up, mean (SD), mo	134.0 (17.1)	134.4 (16.6)	.76	
3 rd percentile	100.5	103.1		
97 th percentile	166	163.6		
Families Maternal age at birth, mean (SD), y	29.5 (5.3)	29.5 (4.9)	.94	
Maternal educational level				
Did not finish high school or equivalent	3 (1.4)	2 (0.9)		
Completed high school or equivalent	59 (27.1)	80 (37.6)		
Some college or university	48 (22.0)	51 (23.9)	.08	
University graduate	103 (47.2)	75 (35.2)		
Unknown	5 (2.3)	5 (2.4)		
Family arrangement				
Single parent	46 (21.1)	46 (21.6)		
Two-parent family	162 (74.3)	151 (70.9)		
Shared custody	9 (4.1)	9 (4.3)	.22	
Foster care	1 (0.5)	5 (2.3)		
Other or unknown	0 (0)	2 (0.9		

Values are n (%) unless stated otherwise.

Abbreviations: PPROM: preterm premature rupture of membranes, SD: standard deviation ^aThese data are for the 431 children with adequate information upon reception of the questionnaire. ^b Women with a pregnancy-associated vascular disease (i.e., pre-eclampsia, growth restriction, hemolysis, elevated liver-function test results, low-platelet syndrome, retroplacental hematoma) were not eligible. ⁷

Table 2 Behavioral and cognitive deficits

Values are n (%) unless stated otherwise.

		Delayed DAMT (n=91)	Normal or advanced DAMT (n=190)	Unadjusted OR (95% CI)	OR adjusted for patients' characteristics (95% CI) ^a	P -value ^a
	Emotional Symptoms Scale					
	Normal	52 (57.1)	122 (64.2)	1.46 (0.91–2.34)	1.32 (0.80–2.18)	.28
	Borderline + abnormal	39(42.9)	68 (35.8)	1.40 (0.91–2.54)		
R	Conduct Problem Scale					
iire (SD(Normal	68 (74.7)	150 (78.9)	1 (4 (1 022 (1)	1.40 (0.832–.36)	.20
	Borderline + abnormal	23 (25.3)	40 (21.1)	1.64 (1.032–.61)		
onna	Hyperactivity Scale					
Strengths and Difficulties Questionnaire (SDQ)	Normal	63 (69.2)	151 (79.5)	1.00 (1.152	1.57 (0.902–.74)	.11
	Borderline + abnormal	28 (30.8)	39 (20.5)	1.90 (1.153–.13)		
	Peer Problems Scale					
	Normal	60 (65.9)	143 (75.3)	1 (7 (1 012 75)	1.58 (0.922–.71)	.10
	Borderline + abnormal	31 (34.1)	47(24.7)	1.67 (1.012–.75)		
	Prosocial Scale					
	Normal	87 (95.6)	181 (95.3)	1.01 (0.292 (6)	0.92 (0.302–.87)	.89
	Borderline + abnormal	4 (4.4)	9(4.7)	1.01 (0.382–.66)		
	Total Difficulties Score					
	Normal	59 (64.9)	148 (77.9)	2.21 (1.353–.62)	1.94 (1.123–.36)	.02
	Borderline + abnormal	32 (35.1)	42 (22.1)			
(Cognitive deficits and learning disabilities ^b	71 (78.0)	98 (51.6)	3.50 (2.205–.57)	3.43 (2.105–.62)	< 0.0001

Abbreviations: DAMT, Draw-a-Man test; OR, odd ratio; CI, confidence interval

^aThe odds ratio was adjusted for gestational age, singleton/multiple pregnancy, socioeconomic variables, gender, and birthweight (using the residual of the linear regression of birthweight on gestational age).

^bCognitive deficits and learning disabilities were defined in Chollat et al., J Pediatr 2014 [3]

Table 3 DAMT performance evaluation

	Abnormal SDQ	Cognitive deficits and learning disabilities		
Sensitivity	43% (95% CI, 335–5)	42% (95% CI, 35–50)		
Specificity	72% (95% CI, 65–78)	82% (95% CI, 74–89)		
Positive predictive value	35% (95% CI, 26–45)	78% (95% CI, 68–86)		
Negative predictive value	78% (95% CI, 72–83)	48 % (95% CI, 41–56)		
Positive likelihood ratio	1.52 (95% CI, 1.08–2.13)	2.35 (95% CI, 1.52–3.63)		
Negative likelihood ratio	0.79 (95% CI, 0.64–0.99)	0.71 (95% Cl, 0.61–0.82)		
Pre-test probability prevalence	0.26	0.6		
Post-test probability prevalence	0.35	0.78		

DAMT, Draw-a-Man test; CI, confidence interval; SDQ, Strengths and Difficulties Questionnaire

Figures:

Figure 1: Flow-chart of the study DAMT, Draw-A-Man test; HFD, human figure drawings

Figure 2: HFD (human figure drawings) examples

A, Chronologic age: 12 years / predicted age: 12 years. B, Chronologic age: 11 years / predicted age: 7 years. C, chronologic age: 10 years / predicted age: 12 years



