

## A Multi-Method Assessment in Adolescent and Adult Females with Giant Congenital Melanocytic Naevus and their Families: Body Image and Psychological Adjustment

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### Abstract

**Background:** Giant Congenital Melanocytic Naevus (GCMN) is a morphological skin alteration present from birth, involving up to 80% of the body surface. GCMN could have a detrimental effect on body perception due to several factors including its aspect, extension and the potential exposure to significant number of surgical interventions.

**Objective:** This pilot study assessed quality of Body Image (BI) and psychological adjustment in subjects with GCMN and their parents.

**Methods:** Subjects and parents underwent a multi-method assessment including a semi-structured interview, a self-administered rating scale to assess BI (Body Uneasiness Test, BUT) and two personality tests: A self-report (MMPI-2RF/A) and a performance test (Rorschach, R-PAS method).

**Results:** Ten families were enrolled in the study. GCMN subjects were all females with high average surgical interventions (median=13). In GCMNs a substantial impairment of BI was detected by the BUT (global severity index=2.34 ± 0.81; Body Image Concern=3.25 ± 0.95), MMPI-2RF/A presented normal ranges and R-PAS showed elevations for the quality of human representations (PHR/GPHR: 119.1 ± 8.1). Mothers showed a trend for health concerns at MMPI-2RF (Malaise: 64.2 ± 9.5), fathers showed under-reporting in almost all tests.

**Conclusion:** This set of GCMN females with relevant surgical history shows significant BI impairment with several aspects of non-integrated body identity at unconscious level, that may result in inability to envision the self and relations with others in adaptive way. Parents show different profiles, including conscious health concerns in mothers and denial in fathers. Families with a GCMN subject could benefit from integrated approaches including medical advice, psychological support and social integration projects.

**Keywords:** Giant congenital melanocytic nevus; Body image; Multi-method assessment

### Introduction

Congenital Melanocytic Naevus (CMN) is a morphological alteration caused by an abnormal concentration of melanocytic cells during embryonal and fetal development [1] which presents from birth and is undetected by routine pre-birth diagnoses. It looks like a dark-colored part of the skin that can look flat, mamillated and sometimes hypertrichotic and may involve up to 80% of the body surface at one or more locations [2]. Giant congenital melanocytic nevi (GCMN) are those CMNs measuring 20 cm or more in greatest diameter [3]. Due to multiple aspects including the low incidence of GCMN (<1:20.000 newborns [4]), controversial data on pathological outcomes [5] and the absence of an international consensus on treatment, a family with a newborn affected by GCMN usually faces several issues and uncertainties that relate to lack of information and support, expectations or interventional options. Although repeated pediatric interventions including dermatologic and plastic surgery are often pursued starting from the earliest months of life in GNMC subjects, a complete excision of deep melanocytic cells is very unlikely to occur, and no data exists in favor of surgical intervention to improve health or reduce risk of malignant melanoma [6]. Whatever the choice of each parents or individual with GNMC is on treatment, such a pervasive morphological alteration may play an important role in the development of the subject's identity processes. It is known that an impaired perception of the body implies a distressing and maladaptive condition that has been proven to be associated with negative psychological functioning, such as low

self-esteem, depression, anxiety and eating disorders [7,8]. Although an extensive skin disease such as GNMC could have a detrimental effect on its owner's body perception, there is currently no data that assess the quality of Body Image (BI) neither in subjects with GCMN who undergo surgical treatment, nor in their parents (who intervene actively in the early treatment process of the newborns). The present study has been conducted to address this gap and to explore potential opportunities to support GNMC patients and their families.

### Materials and Methods

#### Design

This is a pilot study involving subjects with CGMN and their parents. Primary objective is to assess quality of BI in CGMN individuals alone and compared with their parents by means of a Multi-Method assessment procedure including a semi-structured interview and three psychometric tests. Secondary objectives include assessment

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of psychological adjustment in the enrolled individuals through the examination of reality testing resulting from the psychometric tests and subjective perception of CGMN dimensions. Participants were selected from a population in the Italian territory diagnosed at birth with GCMN and who were members of the national GCMN association (Nevus Italia ONLUS), a non-profit organization that promotes the information on this condition and provides support to families. The study inclusion criteria consisted in the selection of GCMN subjects of at least 12 years resident in Italy and with at least one of the parents agreeing to participate. Exclusion criteria were: NMCG subjects with other physical impairments or malformations, mental retardation or an existing clinical diagnosis of mental illness. A Multi-Method assessment procedure included a semi-structured interview, the Body Uneasiness Test [9], the Minnesota Multiphasic Personality Inventory-2RF [10] (MMPI2-RF) for adults, the Minnesota Multiphasic Personality Inventory-2A (MMPI-A) for adolescents [11] and the Rorschach Performance Assessment System (R-PAS) [12]. The semi-structured interview was done with the patients and their parents and included questions involving GCMN medical aspects, its clinical and surgical history, subjective perception of Naevus dimension and psychosocial aspects of their specific experiences. The Body Uneasiness Test (BUT), a 71-item self-administered rating scale [9], was used to directly explore areas specifically related with BI. Additionally, MMPI-2RF/MMPI-2A and R-PAS were used to collect information on both explicit and implicit aspects of the personality for each participant [13]. For the specific purpose of this study, in addition to the analyses to assess validity and the general profile of the subjects, we selected specific scales associated with both explicit and implicit aspects BI and self-perception: Global Severity Index (GSI), Body Image Concern (BIC) and Avoidance (A) from the BUT; Infrequent somatic responses (Fs), Somatic Complaints (RC1) and Malaise (MLS) from MMPI2-RF; Hypochondriasis (Hs), Health Concerns (A-Hea) for the MMPI2-A; Anatomic Content (An), Morbid responses (MOR), percentage of Non-Pure Human responses (NPH%) and Poor Human representation proportion (PHR/GPHR) from the R-PAS.

Considering the exploratory nature of the study and the limited sample, no statistical hypotheses were formulated for this study. The data analysis included test scorings and descriptive analysis with appropriate aggregate indices (mean and standard deviations or median and quartiles, depending from distribution) with comparisons between

variables performed with Mann-Whitney U test. The R package version 3.5.1 was used for statistical analysis.

## Procedure

For each family participating in the study, the assessment was completed during a single visit done by a maximum of three assessors. All participants were informed of the aims of the study and provided informed consent prior to the assessment. Parents of participants under 18 years old provided consent for their daughters. The procedures followed in this study were in accordance with the Declaration of Helsinki. The interview preceded the tests and the self-reports preceded the Rorschach test administration, to establish the best cooperation with the subjects, due to the higher emotional involvement of the Rorschach. Each assessor was specifically dedicated either to interview, self-reports or Rorschach. In order to reduce assessor variability, the Rorschach test was administered by the same researcher to all the members within a single family, with a maximum of two colleagues for the whole study. All collected MMPI-2RF/A and R-PAS resulted valid except for the R-PAS of a GCMN participant who did not complete the test due to the emotional overwhelm during the administration.

## Results

### Participants

Ten families were enrolled in the study between January and September 2017. The sample included 10 GCMN female subjects, 9 mothers and 8 fathers for a total of 7 families with both parents and 3 families with only one parent enrolled. One father was deceased, one mother and one father refused to participate. Researchers used a convenience sampling opening the possibility to participate in the study to the 45 families of the national GCMN association (Nevus Italia Onlus) fitting the age inclusion criteria. A total of 22% of families consented to be included in the study. Although the percentage of female gender in the source population was 40%, all GCMN subjects consenting to this study were females. Subjects and parents' demographic data are presented in Table 1. History of surgical interventions is summarized in Table 2. All subjects but one underwent at least one surgical intervention for GCMN treatment, including 6 subjects with at least one dermatologic surgery (laser dermal abrasion) and 7 subjects with plastic surgery (including skin expanders or skin transplant). The

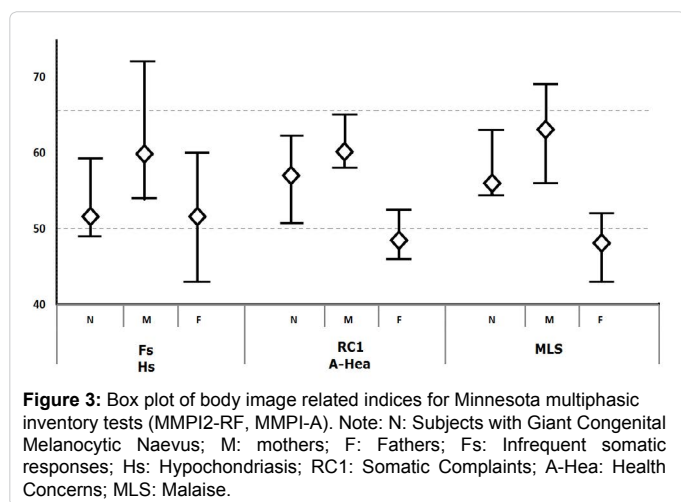
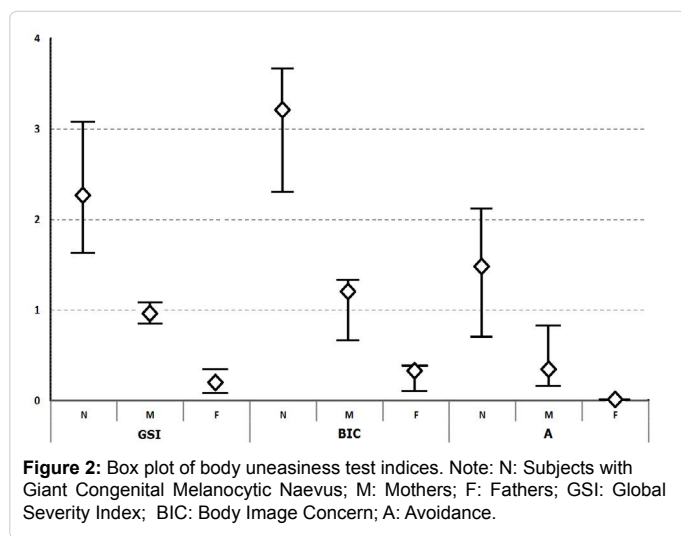
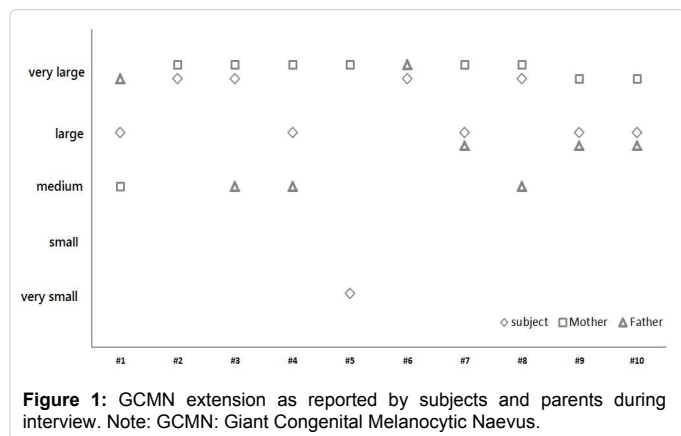
Demography	CGMN subjects	Mothers	Fathers
Participants (N)	10	9	8
Age (yrs, mean ± std)	23 ± 11	55 ± 13	54 ± 10
Female gender (N, %)	10 (100%)	9 (100%)	0 (0%)
Education (yrs, mean ± std)	13 ± 4	14 ± 5	11 ± 3
<b>Marital status</b>			
Single (N, %)	7 (70%)	0 (0%)	0 (0%)
Married (N, %)	3 (30%)	8 (89%)	8 (87%)
Divorced/widow (N, %)	0 (0%)	1 (11%)	1 (13%)

**Table 1:** CGMN Subjects and parents' demographics.

Interventions	Percentage
Any surgery	9 (90%)
Dermatologic surgery (N, %)	6 (60%)
Plastic surgery (N, %)	7 (70%)
Both (N, %)	4 (40%)
Number of interventions (N, mean ± std)	20 ± 18
Age of first intervention (yrs)	<1 y
Age of last intervention (yrs)	16 ± 4

**Table 2:** History of surgical intervention in subjects with CGMN.

median number of interventions per subject was 13 (IQR: 11-25, range: 0 to 30) with all interventions starting at neonatal age and last intervention at median age of 15 years (IQR: 13-18). Eight subjects out of 9 who underwent surgery had a total number of interventions above 10. The dimensions of the GCMN, as reported by subjects and individual parents on a Likert scale ranging from “very small” to “very large” (Figure 1) showed no unanimous agreements on subjective



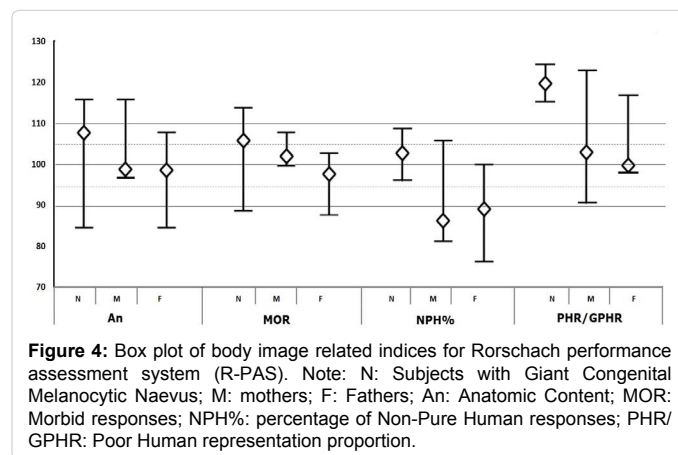
Naevus extension in all the families with maximum disagreement occurring between mother and father pairs ( $p=0.04$ ).

### Body image

All the selected indices from BUT, both absolute values (Figure 2) and compared with age-corrected reference ranges for healthy subjects (9), confirmed presence of an impaired BI in all GCMN subjects with significantly higher values than their parents (Global severity index-CGMN vs. mothers:  $2.34 \pm 0.81$  vs.  $1.05 \pm 0.91$ ,  $p=0.008$ ; CGMN vs. fathers:  $2.34 \pm 0.81$  vs.  $0.25 \pm 0.20$ ,  $p<0.001$ ). When analyzing indices associated with explicit and implicit aspects of BI and self-perception (Figures 3 and 4) results from MMPI-2RF/A presented normal ranges for most of the NMCGs, a higher degree of variability in mothers and a tendency to under-reporting in fathers. Unlike the MMPI-2RF/A, R-PAS variables showed elevations in GCMNs, particularly in the realm of the quality of human representations (PHR/GPHR:  $119.1 \pm 8.1$ ). Despite the limited sample size, the wide age range of GCMNs and medical treatment history, the study data and show consistent and uniform results in the group of GCMNs. A substantial impairment of BI is detected by the BUT in all GCMNs. BI impairment shows to be more associated with “concerns for their aspect” (BIC) rather than a worry for their health status (as also appears from MMPI-2RF/2A and R-PAS). From the set of indices considered to assess BI, the most striking result is that subjects with GCMN have strongly impaired internalized scheme of self and the others, representing the inability to envision the self and relations with others in adaptive way, as indicated by a high PHR/GPHR value in R-PAS, above the upper limit for all subjects where the parameter is computable. Mothers showed in general a higher degree of variability: one mother only shows significant body-related concerns at BUT and 5 out of 9 showed conscious concerns for their own health at MMPI-2RF (Malaise:  $64.2 \pm 9.5$ ). Rorschach data have higher variability and does not allow general considerations. Data related with fathers highlight a general pattern of denial and underestimation of the problem, at MMPI-2RF where almost all indexes show under-reporting. During the interviews some of the fathers seem to confine the GCMN topic mainly as a medical issue.

### Psychological adjustment

Reality testing values of MMPI-2RF/A scales for CGMN were in the normal range (TDH:  $56.5 \pm 6.7$ ; Rc8:  $53.7 \pm 5.3$ ; Rc6:  $52.5 \pm 6.6$ ) but the R-PAS variables connected to a fragile self and to issues in thinking and perception clarity under emotional stress showed very high values in most GCMN subjects (Ego Impairment Index:  $125.1 \pm 13.3$ ; Thought and Perception Composite:  $121.3 \pm 7.6$ ). The group of fathers also show



a moderate impairment testing (Thought and Perception Composite:  $114.5 \pm 9.4$ ). For the GCMN subjects there is a notable discrepancy for the indicators of reality testing between MMPI-2RF/2A vs. Rorschach. While MMPI-2RF/A shows a good quality of adjustment (lack of elevations for Infrequent Psychopathology Responses, Thought Dysfunction, Ideas of Persecution, Aberrant Experiences, Psychopathic Deviate scales in all GCMN subjects but one), R-PAS detects the presence of problems in the reality testing area with either ego impairment index or the thought and perception composite index above the upper limit in all subjects.

## Discussion

The perception of our own body and the gaze of others upon us defines the quality of our BI and may affect our identity and psychological functioning in several directions [14,15]. Previous research has shown that both a positive or a negative quality of BI correlates with several effects, in particular optimism, self-esteem, social support, adaptive coping on one hand [16,17] (positive BI) and distressing and maladaptive conditions such as depression or eating disturbances on the other [7-19] (negative BI). While a positive BI reflects into behaviors of love and respect for the body and leads to internalization of positive information an individual with negative BI perceives himself or herself with defects that are not harmoniously integrated [20]. Among those conditions that negatively affect BI, skin diseases play an important role and have been shown to be associated with negative BI, dysphoria, anxiety and low self-esteem in numerous studies, for psoriasis and vitiligo [19].

The present study is the first investigation to determine quality BI in a small set of GCMN females with a relevant average history of surgical interventions and their parents and showed a substantial impairment of BI in the GCMN subjects. GCMN may relate to a BI impairment for several factors. First, it is a permanent morphological condition that exists from birth, it could be so extensive and pervasive to involve large amount of the body surface (including face) and persists during the entire lifespan of the individual. Moreover, factors associated with surgical treatment may play a role in affecting BI in GCMN subjects. This includes the high number of invasive surgical interventions starting from the early stages of life and the phenomena of post-surgical re-pigmentation or the presence of extensive scars over the body, that could have a major influence on BI as the GCMN itself. All these factors are well represented on average in this sample, considering that 80% of the subjects underwent 10 or more interventions, all starting at neonatal age and often including plastic surgery.

This study also included both parents, who underwent the same tests as GCMN. This choice was done because both parents intervene actively in the decision process at support of GCMN children and because quality of BI have been demonstrated to depend from parents and peers [20]. As parents showed to be more comfortable with their bodies, we could argue that BI impairment in the GCMN subjects was not influenced by a negative BI perception in the other family members.

The study used different means of assessment, including two personality tests: a self-report (MMPI2) and a performance (Rorschach) test. Although self-reports are currently the most popular methods in psychological assessment, the additional use of a performance test was included to capture implicit aspects of the personality and fill potential gaps, if a discrepancy existed between the self-reported characteristics and the actual behavior [13]. Accordingly, even though a poor BI was detected *via* a self-administered rating scale (BUT), the addition of the personality tests helped to better identify the critical areas of impairment.

The results of this study, although limited in size and confined to a sample of females could be of help in understanding the psychological and psychosocial factors that involve GCMN subjects and their families and could have relevance not only for individuals but also for the management of different choices and decision (medical, familial and social) that GCMN and their families could face. Accordingly, it seems legitimate to conclude that GCMN subjects and their families should benefit from an integrated approach that would include medical advice, psychological support and social integration projects.

## Study limitations

This study included a very limited sample of subjects, mainly due to the rare occurrence of the GCMN and the consent process from the family members to participate. Accordingly, we could not test a specific hypothesis or detect other potentially meaningful differences that would have required a higher sample size. This also prevented to analyze dependence from different variables such as medical intervention history or age, that has high variability with half of the sample being adolescents and half adults up to the age of 45. Given the complexity of the scenarios associated with the medical treatment history in these subjects, we cannot determine if a negative BI is due to the GCMN or a combination of the Naevus, scars and surgical history, because almost all subjects considered for this study underwent several interventions during their lives. Notably, although most published studies on GCMN include a slightly higher proportion of females, with ratios ranging from 1.17:1 to 1.46:1 [4], this study, who opened enrollment to GCMN of both genders, succeeded in enrolling families with only female subjects. Although this selection potentially suggests major concerns for BI aspects in this gender, this research could not specifically address this topic.

## Conclusion

From the analysis of the study data, we may conclude that this set of GCMN females do not perceive themselves as ill or suffering because of their morphological alteration, however they show several aspects of non-integrated body identity at unconscious level. Therefore, they may present with difficulties both in self-understanding and understanding others, with consequent relational problems. Parents show different profiles, including conscious health concerns in mothers and a consistent denial outline in fathers, but none of them substantially presents with a negative BI. This supports that an impaired BI in the GCMN subjects of this sample is not caused by a shared family pattern but, most likely, belongs to their morphological condition.

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## Disclosures

None of the authors have conflicts of interest to declare.

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