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An initial investigation of Abnormal Bodily Phenomena in subjects at Ultra High Risk for Psychosis: their prevalence and Clinical implications

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Abstract

Background: Contemporary phenomenological research has considered abnormal bodily phenomena (ABP) to be a phenotypic trait of subjects with schizophrenia in their first psychotic episode. Yet the prevalence of ABP and their clinical significance in subjects at Ultra High Risk (UHR) of psychosis remains unidentified. This study is an exploratory investigation of ABP in UHR subjects and matched healthy controls (HC) examining their relation to clinical features and basic self-disturbances.

Methods: A sample of 26 UHR and 14 HC subjects from three prodromal and early intervention clinics in South London, West London and Cambridge was assessed with the Abnormal Bodily Phenomena questionnaire (ABPq), Comprehensive Assessment of At-Risk Mental States (CAARMS), the Positive and Negative Syndrome Scale (PANSS), the Social and Occupational Functioning Assessment Scale (SOFAS) and the Examination of Anomalous Self Experiences (EASE) checklist.

Results: In our sample ABP occurred in 73.1% of UHR subjects and prominent ABP (proABP) were referred in 53.8% of them. No HC subject reported ABP. The UHR group with proABP had lower CAARMS total score ($t=-9.265$, $p=0.006$). There were no differences in PANSS total score ($t=-1.235$, $p=0.277$), SOFAS score ($H(2) 22.27$, $p=0.666$) and EASE total scores ($z=8.565$, adjusted $p=0.185$) in the UHR subjects with prominent ABP versus those that did not.

Discussion: This initial investigation suggests that ABP could be a prevalent phenotypic feature of UHR subjects.

Keywords

Phenomenology, psychopathology, Ultra High Risk, psychosis, abnormal bodily phenomena, anomalous self-experiences

Abbreviations

Matched healthy control (HC); Ultra High Risk (UHR); Abnormal bodily phenomena (ABP); Abnormal Bodily Phenomena questionnaire (ABPq); Anomalous Self-Experiences (ASE); Examination of Anomalous Self Experiences (EASE); Prominent Abnormal Bodily Phenomena (proABP); UHR subjects with prominent ABP (UHRproABP+); UHR subjects with no prominent ABP (UHRproABP-); Comprehensive Assessment of At-Risk Mental States (CAARMS); Positive and Negative Syndrome Scale (PANSS); Social and Occupational Functioning Assessment Scale (SOFAS);

1. Introduction

Abnormal bodily phenomena (ABP) have been examined in patients with psychosis and particularly schizophrenia even in their seminal representations [1]. ABP are considered to include several different types of symptoms: disturbed *coenesthesia* (an assortment of uncanny bodily feelings with or without delusional interpretation), kinesthetic hallucinations and disruptions of body structure and boundaries. As a group, ABP are subjective phenomena that may be quasi-ineffable in nature and until recently they have been absent from diagnostic textbooks as we lack sensible, specific and reliable tools to assess them.

Following increasing evidence of the limitations of standardized criteria as well as fully structured interviews [2] [3], the development of phenomenologically-based, semi-structured interviews has allowed a better insight into subjective phenomena [4]. This is the case of the EASE interview [5] which focuses on anomalous subjective self experiences (ASE), the use of which has become increasingly relevant in research on psychosis [6] and schizophrenia [7] [5]. The concept of “self” has brought about major interdisciplinary interaction aiming to find common conceptual ground for research. One of the present conceptions takes the self as a pyramidal structure with three elements: the pre-reflexive (the “basic” experiences which implicitly compose one’s way of being in the world), reflexive (the cognitive appraisal of oneself and the world) and narrative (the autobiography which one narrates when portraying oneself). The three elements are interlinked in such a way that a disturbance in pre-reflexive experience of the self could lead to disturbances of other levels of self experience. There are major inputs to this conception from various authors including Joseph Parnas, Louis Sass, Dan Zahavi and Shaun Gallagher [7-12]. The “Anomalous self-experiences” selected by the EASE are an heterogeneous group of phenomena which involve symptoms such as: subjectively experiencing a disturbed way of thinking, subjectively experiencing a disturbed way of being in the world, subjectively experiencing disturbances to one’s own body, disturbances in interpersonal relations and disturbances in principles and attitudes to the world. The EASE interview has shown a good internal and external validity and its use in subjects at UHR [13-16] and in prodromal phases of schizophrenia [17,18] has shown that ASE are present in these populations and suggested they could be used as an early marker.

A similar research track has now explored the prevalence and relevance of ABP in patients with first psychotic episode and, in particular, in subjects with diagnosis of schizophrenia [19] [20]. The “Abnormal Bodily Phenomena Questionnaire” (ABPq) [20,21] allows a systematized enquiry into these experiences. Studies using ABPq have shown an assortment of bodily phenomena occurring in schizophrenia [20] and the in the first episode psychosis [19]. These include uncanny feelings of numbness, loss of body vitality, the sense of disappearance of parts, change in form (shrinking or enlargement), dimension unusual heaviness or lightness) or even “movement of internal parts” of the body [19,22,23]. Examples of these phenomena are presented in the Box 1.

Box 1. Example of Abnormal Bodily Phenomena (our sample)

Experience of violation “when he stretched his hand I felt an energy entering my body. That energy was pounding my right side until it break in”

Experience of externalization “I felt that my body had become as a boiling soup and I was no longer in control of my energy that flows around me. Then my body became an ingredient in the soup parts of me are the vapors and rest of the soup ”

Morbid objectivization “I felt my brain freezing. Then thoughts were moving so slowly I could feel them drive from ear to ear”

Devitalization “The loss of the animal part in me, I lost it! I feel that I act as I was programmed, even eating and I feel like a robot, even my arms feel artificially attached to me”

Experience of internal dynamization “I felt my brain starting to rotate, only slightly, but to a point where I was worried it would detach from my neck and it was going to fall to the floor”

Experience of transformation “I felt that my hands were shifting as I moved them towards my face – bigger than smaller. Such a scary sense that I had to look away”

Dysmorphophobia “I always felt my left part of my body was not ok. I even wondered that it might not be mine. There is a sense of evil from the left side. I feel that it’s evil”

Disesthetic paroxysm “blurring feeling on my cheek and neck when someone speaks to me. I cannot describe it but its sufficiently awkward for me to be scared of being too close”

Recurrent pain-like experience “the pain just comes and goes... normally when I’m breathing I can end up feeling needles in the lung. I know wrong kind of pain as its so different. it doesn’t help taking anything”

Various studies suggest that ABP occur as trait features before the first psychotic episode in patients with schizophrenia [24-29]. The Comprehensive Assessment of At-Risk Mental State scale

[31], one of the instruments relevant to the UHR profile, also selects disturbances of bodily phenomena. Yet no study has independently regarded their presence and clinical significance in subjects at high-risk of psychosis. Therefore their occurrence and phenotypic profile is unknown, as is their relation with other presenting symptoms.

This study used the ABPq to appraise ABP in a sample of subjects at High Risk of Psychosis (HR) and assess their occurrence in this population. Our first goal was to measure the prevalence of ABP in UHR group and matched healthy controls (HC). Our second goal was to see how the presence of ABP was related to other clinical features in this population including (i) functional features (SOFAS), (ii) UHR symptoms (PANSS and CAARMS) and (iii) disturbance of basic self-experiences (EASE).

2. Methods

2.1. Setting and sample

Our sample included subjects at High Clinical Risk for psychosis from OASIS (prodromal clinic, SLAM NHS Foundation Trust, London [30]), West London Early Intervention and CAMEO (“Cambridge Early Onset”, Cambridge University, Cambridge, UK) teams. These clinics are responsible for early intervention in young adults where they undertake a detailed multidisciplinary assessment including combined review of clinical judgment and screening instruments [30]. We added the ABP and EASE semi-structured clinical interviews. Their UHR group is defined by High-Risk criteria derived from Comprehensive Assessment of At Risk Mental State (CAARMS) [31] and the Social and Occupation Functioning Assessment Scale (SOFAS) [32]. Some features of our sample included: subjects were between 18 and 35 years of age, 65% of our subjects had never used drugs while 12.5% had experimented and 15% had moderate to severe use 22.5% were taking antidepressants while 7.5% were taking antipsychotics. After an average of 28 months of follow-up (November 2015) two of the UHR subjects transitioned to a psychotic episode. Enrollment process for controls included advertisement for those matched for age and gender subjects and that had no present or past personal or family psychiatric history. This study had ethical approval and all the subjects participated after signing a written

voluntary informed consent form.

2.2.Procedure:

In this study, detailed semi-structured interviews were performed by two psychiatrists (LM and IB) blind to clinical diagnosis. They have strong training in Psychopathology and a course in using the EASE (semi-structured interview similar to the Abnormal Bodily Phenomena questionnaire). Joint scoring of audio-recorded interviews ensured inter-raters reliability. The final scores were decided through consensus discussion.

2.3.Sociodemographics

In line with previous OASIS studies, socio-demographic data was collected from the subjects' clinical file. These included age, gender, education, personal or family history of psychiatric disorder and present or past psychiatric treatment, country of birth and employment (full time students were considered employed). [30]. Healthy controls were asked the same questions during the interviews. The sample was recruited between August 2013 and November 2013.

2.4.Clinical Measures

Anomalous Bodily Phenomena Questionnaire (ABPq) [21]

The prevalence of ABP was assessed with the ABPq, a nine items semi-structured interview split into five sections: Demarcation (experience of violation and externalisation), Vitality (morbid objectivisation and devitalisation), Coherence (experience of internal dynamisation), Identity (experience of transformation and dysmorphophobia) and Activity (disesthetic paroxysm and recurrent pain-like experiences). Each item is scored for its frequency, intensity, impairment and coping in 7 point assessment scale. The developers of ABPq proposed that we should acknowledge the overall score as the sum of all scores and the presence of “prominent ABP” (proABP) in subjects with scores over 3 in any of the domains. Selected examples of patients' answers are present in the introduction textbox.

Positive and Negative Syndrome Scale (PANSS) [33]:

A seven-point assessment of 30 items split into three sections: 7 positive, 7 negative and 16 general psychopathology items. It was part of the assessment at admission in

the OASIS and CAMEO teams. In our study it allowed the assessment of general psychopathology and positive and negative symptoms and their relation to ABP.

Comprehensive Assessment of At-Risk Mental State (CAARMS) [31]

A semi-structured clinical interview for attenuated psychotic symptoms (thought disturbances and perceptual abnormalities), which is part of the assessment of OASIS and CAMEO teams. It has 26 items divided across 7 subscales: 4 Positive Symptom items, 2 Cognitive and 3 Emotional Disturbances items, 3 Negative Symptoms items, 4 Behavioral Change items, 4 Motor/Physical Changes items, and 8 General Psychopathology items. These are rated on severity and frequency of the symptom in a 6-point assessment (from 0 absent/never to 6 psychotic and severe/continuous).

Social and Occupation Functioning Assessment Scale [32]

This scale is a modified version of the Global Assessment of Functioning (GAF) scale separating the measures of social and occupational functioning from the measures of symptoms and psychological functioning. Its scores range from 0 to 100. Scoring is according to information obtained in the psychiatric interview.

Examination of Anomalous Self Experiences checklist (EASE) [34]

It is a semi-structured interview for anomalous subjective experience that includes 57 items (88 if sub-items) and a Cronbach α of 0,87 [6]. It is divided into five subscales a) 28 sub-items cognition and stream of consciousness; b) 36 sub-items in self-awareness and presence; c) 16 sub-items in bodily experiences; d) 6 sub-items in demarcation/transitivity; e) 8 sub-items in existential reorientation. Our score was an overall dichotomous score in accordance to presence (1) or absence (0) of items. It is a reliable instrument to enquire anomalous experiences of the “pre-reflexive” sense of first-person perspective or basic self [6]. They have been empirically substantiated in a) early psychosis [5], b) prodromal phases of psychosis [13,35] and c) in the schizotypic and those with schizotaxia [36]. The abnormalities of self-awareness have been shown as promising in the conceptualization of those at risk of psychosis [37] and in schizophrenia-prone individuals [16,17].

2.5. Statistical Analysis

Descriptive statistics including ANOVA and t-tests for continuous variables and Fisher exact test for categorical variables were used to compare between group

differences. Non-parametric tests were used as appropriate when the assumptions for parametric null hypothesis tests were violated. We tested inter-rater reliability by independent reevaluation of the tape recording of the first 6 interviews (three interviews performed by IB and three by LM which were subsequently evaluated by the other interviewer).

Our first aim was to measure the prevalence of ABP and proABP in UHR subjects and matched HC. Our second aim was to investigate if the presence of proABP affected clinical features of UHR including a) UHR symptoms (CAARMS and PANSS scores), b) functional status (SOFAS score) and c) disturbance of basic self-experiences (EASE score). For CAARMS and PANSS scores, as normality assumption was maintained, we used t-tests to compare UHRproABP- or UHRproABP+ groups. For SOFAS and EASE scores, as the assumption of normality and homogeneity of variance were violated (Shapiro-Wilk test significant in at least one group for each variable, $p < 0.05$; Levene test significant in every variable but EASE Existential Reorientation subscale, $p < 0.05$) we decided to perform Kruskal-Wallis non-parametric test to compare HC, UHRproABP- and UHRproABP+. For post-hoc analysis we used Dunn-Bonferroni's approach [38] for multiple comparisons in non-parametric test. When not otherwise specified, two-side $p < 0.05$ was considered significant. Given the exploratory nature of this study and the small sample size, we decided not to control for familywise error rate. Our decision was made to avoid loss of statistical power. However, we performed a Bonferroni correction in the post-hoc analysis given the large increase in the number of null hypothesis formulated in this procedure. The adjusted p-value ($p * \text{number of pairwise comparisons}$) has been reported. All the analyses were performed under SPSS IBM 22.

3. Results

3.1. Sociodemographic characteristics

Interviews took on average 58 min (SD=10) in HC and 134 min (SD=40) in UHR subjects. The inter-rater correlation was 0.90 ($p < 0.001$) overall. The sample comprised 26 UHR subjects with mean age of 23.73 years (SD 4.35) and of which 57% were male. The matching sample of HC participants presented no differences on baseline demographics, accounted in **Table 1**, but the employment rate was higher in HC than UHR (Fisher's exact test 6.983, $p = 0.029$).

3.2. Prevalence of ABP and proABP and average ABPq score

In our sample, ABP were absent in all subjects in HC group and were found in 19 (73.1%) UHR subjects where its overall score (mean, SD) was 35.43 ± 35.80 . These represent subjects with several ABP, which they evaluated in their frequency, intensity and impairment and to which they discussed the coping strategies. Regarding the individual domain scores, our UHR sample subjects reported (mean, SD) a disturbance in ego demarcation score of 4.84 ± 9.76 , a disturbance in ego vitality score of 9.0 ± 10.47 , a disturbance in ego coherence score of 3.00 ± 6.26 , a disturbance in ego Identity score of 8.05 ± 12.86 and a disturbance in ego activity score of 10.53 ± 9.36 . The scoring system allowed us to identify 14 subjects (53.8%) with prominent ABP and hereinafter the acronym UHRproABP+ represents these subjects while UHRproABP- represents those that had no ABP or that considered them mild.

3.3. EASE, SOFAS, CAARMS and PANSS scores across HC, UHRproABP+ and UHRproABP- groups

There were statistically significant differences in EASE total and specific scores across the three groups of HC, UHRproABP- and UHRproABP+. **Table 2** portrays EASE and SOFAS scores across HC, UHRproABP- and UHRproABP+ groups. The post-hoc analysis between UHRproABP- and UHRproABP+ groups showed no differences between their SOFAS score ($z = -1.371$, $p = 1.00$) and their EASE total score ($z = 8.565$, adjusted $p = 0.185$). **Figure 1** displays sample distribution of EASE scores across HC, UHRproABP- and UHRproABP+ groups.

In our sample the UHRproABP+ vs UHRproABP- subjects had on average lower CAARMS total score (UHRproABP- 41.42 ± 12.32 , UHRproABP+ 36.36 ± 24.23 , $t = -9.265$ (24), $p = 0.006$). There were no statistically significant differences in PANSS total score between those groups (UHRproABP- 51.33 ± 12.72 , UHRproABP+ 54.07 ± 17.04 , $t = -1.235$ (24), $p = 0.277$). **Table 3** details the differences between PANSS and CAARMS specific domain scores across UHRproABP+ and UHRproABP- groups.

4. Discussion

This is a preliminary study exploring the prevalence and the impact of ABP on other clinical features in UHR subjects. In our sample, ABP were specific and highly prevalent in UHR subjects - more than half of the UHR subjects showed prominent ABP. There were significant differences in EASE scores across HC, UHRproABP- and UHRproABP+. Yet subjects with prominent ABP (versus other UHR subjects) only differ in their CAARMS overall score (which was lower) while they did not in their EASE total, PANSS and SOFAs scores.

Our first aim was to determine if ABP were present in the UHR subjects. We found that 73% of our sample subjects experienced ABP and ~53% experienced prominent ABP. This feature contrasted with HC group that reported no ABP. This allowed us to speculate that ABP might be a phenotypic marker of psychosis vulnerability, which only further studies with larger samples and with clinical control group can show. Still, as much as 30% of our sample had no ABP and 50% had no prominent ABP. This seems attuned to the fact that the UHR is heterogeneous, presents various comorbidities and has diverse diagnostic and functional outcomes [13].

Our second aim was to determine if ABP impacted on clinical features of the UHR population. We found that the presence of proABP status did not differentiate the UHR sample across PANSS and SOFAS scores and that proABP subjects had lower CAARMS overall score. Again due to the small sample size in our study, the ABP value as a clinical feature remains unknown. Yet the lower CAARMS scores made us raise the possibility that ABPq might assess phenomena unlike other criteria defining UHR for psychosis (including bodily phenomena considered in the CAARMS) as 50% of our UHR sample had prominent ABP and lower CAARMS score was found.

Our third aim was to test the relation between ABP and anomalous subjective self-experiences. Our UHR group had higher EASE scores compared to controls. This is in line with previous evidence showing the presence of ASE in the UHR group [13,14] and the possibility that they segregate the UHR subjects at risk of schizophrenia [16]. In our sample we found that subjects referring prominent ABP did not have significantly higher EASE scores. This may be due to weak statistical power due to small group size in each group and further studies with larger samples are needed to clarify this issue. It should not be understated that the relation between ABPq and EASE interview also occurs at phenomenological level and it is important to elucidate

if they are overlapping constructs or if one is a sub-construct of the other. There are studies suggesting that ASE lead to ABP [12] while others advocate that ABP antecede the occurrence of ASE [39,40]. It seems important to stress that the bodily phenomena assessed by the ABPq and the EASE interview are different, for instance, the “internal dynamism” phenomenon only appears in the first and the “mirror-related” phenomenon is only materialized in the second. An overall better conceptualization of the mutual implications between ABP and ASE is needed.

4.1 Limitations

First, due to our small sample size, our study should be considered as an exploratory study. We cannot also exclude that our results could be false negatives or false positives. The generalization of our results might be limited by the specifics of our sample as only two UHR clinics were used. Along these limitations, which are specific to our study, it must be considered that the UHR construct has shown conceptual and empirical heterogeneity further limiting the relevance of our results to other UHR populations [41]. The difference in employment rates across our sample groups seemed to be an artefact of the process of selection of the UHR group (the use of SOFAS for functioning). A clinical control group (e.g. affective, anxious or personality disorders) and follow-up data are necessary to better define the clinical relevance of ABP. Lastly, prescribed drugs and illicit substances were appraised and yet the small sample size did not allow the determination of their effect on our scores.

5. Conclusions

This is the first study to explore the prevalence and implications of Abnormal Bodily Phenomena in subjects considered at UHR. In our sample ABP were highly prevalent (while absent from matched HC) and in 50% of subjects we found prominent ABP. We raised the possibility that ABP could be a phenotypic component of UHR psychopathology and particularly of a subgroup of these subjects. Future prospective and with larger samples studies are necessary before endorsing our considerations and particularly, to understand if ABP predicts clinical outcomes or treatment response in UHR subjects.

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Tables and Figures

Table 1: Socio-demographic and clinical characterization of sample

	Total sample (N=40) mean±SD or n (%)	HC (N=14) mean±SD or n (%)	UHR (n = 26)		F or Fisher's	p
			UHRproAB P- (N=13) mean±SD or n (%)	UHRproAB P+ (N=14) mean±SD or n (%)		
Age at inclusion	23.9±3.9	24.21±3.22	23.25±4.070	24.14±4.605	0.227	p=0.798
Country of Birth					1.107	p=1.000
United Kingdom	38 (95)	13 (92.9)	12 (100)	13 (92.9)		
Other	2 (5)	1 (7.1)	-	1(7.1)		
Gender					3.048	p=0.236
Male	19 (47.5)	4 (28.6)	7 (41.7)	8 (57.1)		
Female	21 (52.5)	10 (71.4)	5 (23.8)	6 (42.9)		
Ethnicity					1.014	p=0.707
White British	24 (60)	10 (71.4)	7 (58.3)	7 (53.8)		
Other	15 (37.5)	4 (28.6)	5 (41.7)	6 (46.2)		
Missing	1 (2.5)	-	-	1 (7.7)		
Employed/ Studying					6.983	p=0.029
Yes	27 (67.5)	13 (92.9)	8 (66.7)	6 (46.2)		
No	12 (30)	1 (7.1)	4 (33.3)	7 (58.3)		
Missing	1 (2.5)	-	-	1 (7.6)		
Mean n° years of Education (years)	14.78±2.91	16.14±3.44	15.08±1.83	13.04±2.33	4.565	p=0.017

number of subjects or Mean ± SD. Percentages under parenthesis. p-values refer to ANOVA and Fisher's Exact Test between HC, UHRproABP- and UHRproABP+ group for continuous and categorical values respectively

Table 2: Between groups differences in EASE and SOFAS scores

	HC (N=14) median (range) Mean rank	UHRproABP - (N=12) median (range) Mean rank	UHRproABP + (N=14) median (range) Mean rank	H(2)	p
EASE score	0.50 (0-9) 7.68	20.50 (6-38) 22.79	34.00 (18-83) 31.36	29.570	p<0.001
EASE Cognition and Consciousness score	0.0 (0-5) 7.75	7.00 (2-15) 23.50	13.00 (5-23) 30.68	28.634	p<0.001
EASE Self-awareness and Presence score	0.00 (0-4) 7.68	11.00 (3-19) 24.50	13.50 (6-31) 29.89	28.105	p<0.001
EASE Bodily experiences score	0.00 (0-1) 9.82	1.50 (0-4) 20.83	3.50 (1-15) 30.89	24.854	p<0.001
EASE Demarcation/ transitivism score	0.00 (0-0) 13.50	0.00 (0-1) 21.21	1.00 (0-6) 26.89	13.121	p=0.001
EASE Existential Reorientation score	0.00 (0-5) 12.79	0.50 (0-7) 19.33	4.00 (1-8) 29.21	15.065	p=0.001
SOFAS score	91.00 (90-100) 30.00	60.00 (40-75) 11.70	59.00 (43-70) 13.07	22.270	p<0.001

Three groups were compared and considering the non-normal distribution of data and heterogeneity of variance, we adopted the Kruskal–Wallis non-parametric test. Median (with range) and mean rank for each group are reported. P values correspond to differences between the three groups. Abbreviations: HC: Healthy controls; UHRproABP+: UHR subjects with prominent ABP; UHRproABP-: UHR subjects with no prominent ABP

Table 3: Impact of proABP on the PANSS and CAARMS scores

	UHRproABP - (N=12) mean±SD	UHRproABP + (N=14) mean±SD	t (24)	p value
PANSS Positive Symptoms	12.17±3.90	13.29±4.14	-0.705	p=0.487
PANSS Negative Symptoms	12.25±4.84	12.07±4.95	0.093	p=0.927
PANSS General Psychopathology	26.83±7.15	28.81±9.89	-0.547	p=0.589
CAARMS Positive Symptoms	8.83±4.46	7.86±3.48	0.306	p=0.762
CAARMS Cognitive Disturbances	3.50±1.93	1.93±1.82	2.111	p=0.045*
CAARMS Emotional Disturbances	3.08±2.93	2.93±2.98	0.129	p=0.899
CAARMS Negative symptoms	6.25±3.36	4.14±3.35	1.597	p=0.123
CAARMS Behavioral Changes	5.58±3.75	5.93±4.92	-0.198	p=0.844
CAARMS Motor/Physical Changes	1.17±2.29	2.29±3.99	-0.857	p=0.400
CAARMS General Psychopathology	13.50±5.99	11.29±7.80	0.802	p=0.430

* Significant difference (2-sided p<0.05)

Figure 1: Examination of Anomalous Self Experiences Scores in HC, UHRproABP- and UHRproABP+

The three groups differed for total EASE score ($N=40$, $H(2)=29.570$, $p<0.001$)

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