Cotard's: A controlled single case study of putative perceptual, cognitive and psychological risk factors.

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Abstract

In this controlled case study, we report the psychological and neuropsychological features in a 55-year-old female (D.R.) with a history of Cotard's. Her nihilistic delusion was marked by the belief that her torso had dissolved, and food passed directly from her mouth into her legs which were hollow. Her delusion developed in the context of major depressive disorder (with psychotic features), which was diagnosed by a certified psychiatrist (F.O.).

During the prodromal phase, D.R. experienced distortion of body parts and abnormal somatosensory sensations which exhibited a spatial correlation with the somatosensory homunculus and areas of heightened sensory innervation. During remission, low-level visual perceptual impairments in identifying degraded letters, object silhouettes and matching unfamiliar faces viewed under different lighting conditions and or different angles, that strikingly dissociated from an otherwise intact neuropsychological profile. Additionally, subtle manifestations of internally generated bodily signals, such as distortion, peculiar tastes, and feelings of uncleanness, were noted, along with an external attribution style. Collectively, these features are posited to represent stable or mediating vulnerability factors rather than transient indicators of dysfunction. This observation aligns with the notion that organic or functional involvement in Cotard's manifests when information processing systems are subjected to high-stress levels during the prodromal and active phases of functional disorders. Inherent weaknesses within information processing systems and high-level attribution biases may be artificially exposed under direct measurement by challenging the processing systems with specific stimuli and tasks that are particularly challenging.

The findings are discussed in reference to models of delusion formation.

Keywords: Cotard's, Sensory homunculus, Prodromal, Remission, Neuropsychology, Attribution bias, Visual perceptual impairment, Body distortion.

Introduction

The Cotard delusion is a rare condition that affects a person's sense of self. In the first recorded case, Cotard (1884) reported on a woman who believed she was only skin and bone, with no brain, nerves, chest, or internal organs. She also thought she was immortal and that neither God nor the devil existed. In another case, a man with nihilistic beliefs had difficulty remembering his children's faces. Cotard interpreted this as meaning that the abnormal nihilistic belief is characterized by a disruption in an individual's ability to visualize their body parts mentally. It is important to understand the disruptions in information processing and the stages at which they occur to help identify putative risk factors and episode indicators of Cotard's.

In this controlled case study, we report a comprehensive psychological and cognitive neuropsychological assessment while our patient (D.R.) was in full remission. Additionally, D.R., who is a physician, provided unique insights into the changes in her sense of self that occurred during the prodromal phase. This is the first such account of potential episode indicators or mediating risk factors in Cotard’s as far as the authors are aware. Our planned 'step-wise' approach complements the recent case study of Mr A, a 19-year-old male who developed Cotard syndrome in the context of autoimmune encephalitis with depressive symptoms (Restrepo-Martínez et al., 2019). His nihilistic delusion was characterised by the belief that his tendons had dried up, his organs, particularly his heart, were getting bigger, and he could no longer feel the blood flowing through his veins. Of particular interest is the presence of depersonalization at the time of admission, but after six sessions of electroconvulsive therapy, both his Cotard's delusion and depersonalization were remitted. However, abnormal bodily experiences persisted; for example, he felt that one of his kidneys was "not working properly, perhaps because it had been damaged as a result of an infection”. This inquiry is predicated on the well-established dual-factor model of delusion formation, which encompasses disturbances in both 'low-level' perceptual and 'high-level' cognitive information processing (Helmsley & Garety, 1986; Lyon et al., 1994). Billion (2016) posited that depersonalization, denoting a sense of detachment from one's body or body parts (Sierra & David, 2011), functions as the initial low-level perceptual factor in the genesis of Cotard’s syndrome. Bolstering this assertion is the recent model suggesting that depersonalization emanates from a deficiency in the attenuation of somatosensory information processing, thereby engendering abnormal perceptions and beliefs about the self-model (Ciaunica et al., 2022). Somatosensory attenuation pertains to the notion that self-generated touches are perceived as less intense (owing to their predicability) than external touches of equivalent intensity, which carry greater uncertainty and therefore novelty and heightened sensation (Blakemore et al., 2000). Gerrans (2019, 2020) argues that depersonalization, although often associated with Cotard's syndrome, is not a necessary aspect of it. Instead of a two-factor view, Gerrans proposes a hierarchical model in which a sense of ownership of one's body arises from the integration of basic interoceptive experiences, awareness of the internal physiological state of the body (Garfinkel & Critchley, 2013 as referenced in Garfinkel et al., 2014) with an affective sense of the body. This integration infuses the body, and its parts, with emotional significance, leading to qualitative changes in the experience of the material self. Accordingly, depersonalisation results from a failure to fully integrate the material self with the affective self, whereas Cotard's reflects a catastrophic breakdown of the hierarchical modelling of the self.

Our investigation of D.R. was guided by three objectives. First, we aimed to provide an account of the prodrome of the nihilistic delusion by utilizing D.R.'s unique insight gained from her medical training. Second, we sought to examine D.R.'s psychological and neuropsychological profile during remission to identify potential risk factors for delusional beliefs related to the body and self. Lastly, we were particularly interested in visual-perceptual processing, drawing inspiration from Cotard's description of his second patient who had difficulty recalling his children's faces.

Case Study

Psychiatric History

D.R. was a practicing physician. She was first presented to psychiatric services in 1981 at the age of 28 years, shortly after the birth of her first child. At the time of presentation, the complaint was of rapidly fluctuating mood that was predominantly low mood. Over the following eight years, D.R. had six episodes of depression, three requiring inpatient treatment. Her treatment included courses of electroconvulsive therapy. She also had one episode of hypomania characterised by motor restlessness, over-talkativeness, euphoria, lack of sleep and pathological over-spending. D.R. remained stable until the age of 51, when she was referred for a recurrence of a major depressive disorder. She had four hospital admissions for a major depressive episode during the following 12 months. These episodes of severe depression were typified by persistently low mood, a feeling that her life was ebbing away, severe feelings of hopelessness and compelling thoughts of suicide that she did not act upon. She also experienced poor sleep, loss of appetite, low energy, anhedonia, weight loss and significant psychomotor retardation, such as slow movement and slow speech. It was over this period that she was given a clinical diagnosis of bipolar disorder with episodes of depression with psychotic features. Her depressive phases were characterised by the presence of Cotard syndrome. She was managed by a consultant psychiatrist (FO). D.R. held the nihilistic belief that her 'insides had dissolved' so that when she swallowed food dropped from her mouth into her legs. D.R. responded to a combination of lamotrigine 200 mg bd and olanzapine 2.5 mg daily.

She has no family history of psychiatric disorders. She is married with two grown-up children. Since then, she has continued to have transient but definite hypomanic episodes lasting no more than a few days at a time. The reported study occurred four years later when D.R. was aged 55 years. D.R. gave informed consent to participate in the study, which was conducted in accordance with Good Clinical Practice guidelines. The study was approved by South Staffordshire Local Research Ethics Committee (REC reference 08/H1203/46).

Neuroimaging

D.R. underwent a structural MRI five years after developing Cotard syndrome to rule out an organic aetiology to her nihilistic syndrome. The brain images, shown in Figure 1 reveal no structural abnormalities in the region of the insula.

(FIGURE 1 HERE)

The Prodromal Phase (objective 1)

D.R. vividly recalled the bodily sensations that preceded the development of her nihilistic delusion. Numbness originated in her upper torso, extending laterally her arms upwards into her head and legs. The numbness was replaced by a tingling sensation when it reached her toes, fingertips, and face. The tingling felt especially pronounced around both eyes, extending down either side of her nose in an inverted U shape. She also reported that parts of her body felt distorted: her mouth was disproportionately large, her head and face felt as if they were miles above and unconnected to 'her body', her legs felt 'wooden' and did 'not work properly'.

We mapped the spatial and temporal progression of D.R.s sensations using Penfield's topographic representation of the sensory distribution of the body. Figure 1. shows a schematic representation of the progression of D.R.'s disordered body perception.

(FIGURE 2 ABOUT HERE)

Self-Awareness and Causal Beliefs during remission (objective 2)

Next, we wanted to explore if disturbances in self-awareness and causal beliefs were present during full remission of D.R.'s nihilistic delusion. Self-report feelings of unreality and altered body perception were assessed by The Cambridge Depersonalization Scale (Sierra & Berrios, 2000) and the Body Distortion Questionnaireand causality and high-order causal beliefs were assessed by The Magical Ideation Scale (Eckblad & Chapman, 1983) and the The Belief in Divine Intervention Scale (Degelman & Lynn, 1995). These assessments were limited to D.R., and did not form part of the controlled case study.

The Cambridge Depersonalization Scale (Sierra & Berrios, 2000) is a 29-item scale that captures the frequency and duration of depersonalization symptoms over the 'last 6 months'. Some of the statements on the scale are "I experience disconnection from parts of my body", "I suddenly feel detached from reality or isolated from the world" and "I have lost some physical sensations like hunger and thirst, so eating and drinking feels like a mechanical routine". The evaluation of each item involves two distinct Likert scales. One assesses the frequency of experiences within the past six months using a 7-point scale that ranges from "never" to "more than a week." The other scale measures the duration of the experience using a 4-point scale that ranges from "a few seconds" to "about a day." A total score is calculated by adding all item scores. The maximum score is 319, with higher scores indicating increasing intensity.

The 81 item Body Distortion Questionnaire (Garner & Garfinkel, 1997) uses a three-point scale (Yes, No, Undecided) to explore physical and functional attributes of one's body. Illustrative statements include 'My body feels unusually heavy', 'My mouth feels like it is changing in size', 'My skin feels less ticklish than usual', 'My toes feel dirty'.

Performance on both measures is presented in Table 1.

(TABLE 1 ABOUT HERE)

Experiences of depersonalization were rare in occurrence, of short duration and limited in scope – centered on feelings that her body didn't belong to her. There was no evidence of derealization, emotional numbing or anomalies in subjective recall.

On the other hand, body distortion was more pervasive. For example, she endorsed statements about her body felt unusually heavy, her hips felt large, her arms felt long, and her nose felt big. A theme of ‘dirtiness’ or odour was also present: her armpits and toes felt dirty, she had a ‘dirty’ taste in her mouth, and the odour of her breath was unpleasant.

The Magical Ideation Scale (Eckblad & Chapman, 1983) contains 24 items scored in a true-false format. Sample items include “If reincarnation were true, it would explain some unusual experiences I have had” and “Numbers like 13 and 7 have no special powers”. The maximum score is 30, with higher scores indicating a belief that specific words, thoughts, emotions or rituals can influence the external world.

The Belief in Divine Intervention Scale (Degelman & Lynn, 1995) contains six items rated on a binary agree/disagree scale. Four items endorse divine intervention, such as ‘God sometimes directly intervenes to heal individuals of diseases like cancer’, and two refute divine intervention e.g., ‘God does not intervene in our daily lives’.

(TABLE 2 ABOUT HERE)

D.R. showed no evidence of trait superstitious thinking or strong causal beliefs.

Neuropsychological Profile (objective 3) The second part of our study comprised a controlled neuropsychological investigation of D.R. We wanted to explore if Cotard’s original observation of disordered ‘mental vision’ was manifest in a neuropsychological investigation of visual cognition, and to compare her performance with verbal cognition within a hierarchy of information processing demand.

Control Groups Eight females of superior or very superior IQ (IQ range 120-129, and IQ 130 and above, respectively) provided baseline data for the neuropsychological assessment (The National Adult Reading Test, NART, Nelson & Willison, 1991). Four females had a diagnosis of major depressive disorder (without psychotic features) formed the psychiatric control group and four females without a psychiatric or neurological history formed the normal control group. The NART comprises 50 written words in British English with irregular spellings (e.g. "aisle"). Previous research shows a strong correlation between the number of errors made on the NART with full-scale scores on the Wechsler Adult Intelligence Scales (Bright et al., 2018).

Table 3 shows the age and premorbid estimate of IQ for D.R. and both control groups.

(TABLE 3 ABOUT HERE)

A two standard deviation range either side of the control mean indicated the cut-off for superior or inferior performance.

Symptoms of depression and attribution styles were examined with the self-report 9 item Hamilton Depression Inventory (HDI, Reynolds & Kobak, 1995) and the Attributional Style Questionnaire (ASQ, Peterson et al., 1982), respectively.

The HDI asks respondents to consider how each statement best describes their behaviour or how they were feeling over the past two weeks, using a likert scale of Not at all (0) to Almost all the time (4). Statements relate to low mood, somatic and anxiety symptoms, suicidal ideation, negative beliefs, and maladaptive thinking styles. The ASQ is a self-report instrument that rates explanatory styles for bad events and for good events. The ASQ presents 12 hypothetical events, six have good outcomes and six have bad outcomes, and the participant is asked to write down the one major cause of each event and then rate the cause along a 7-point continuum according to five causal dimensions. The first dimension asks about whether the event has an internal or external cause i.e., how much respondents believe they themselves are responsible for the event (from 1 = totally due to other people or circumstances to 7=totally due to me). The next question is about whether the event has a stable or unstable cause i.e., how much respondents believe the event's cause is present over time (1=will never again be present to 7=will always be present). The third question is about whether the event has a global or specific cause i.e., how much the respondents believe the cause of the event occurs across varying conditions (1=influences just this situation to 7=influences all situations in my life). The fourth question is about the importance of the situation (from 1 = not at all important to 7=extremely importance. The final question is about the locus of control i.e. extent to which one feels in control of events in your life (1= not at all to 7 = full control).

Neuropsychological Test Battery The battery consisted of assessments that separately assessed visual and verbal information processing under conditions of increasing information processing demands.

Low-level nonverbal processing

The perception of visual objects was assessed with three subtests (Silhouettes, Object decision, Progressive silhouettes subtests) from the Object Perception component of the Visual Object and Space Perception Battery (VOSP, James & Warrington, 1967). This test has been established as a valid measure of assessing ventral and dorsal streams of information processing. The silhouettes subtest required naming 15 animals and 15 inanimate silhouettes presented from unusual angles. The object decision subtest involved naming a real object juxtaposed by 3 nonsense objects. In the progressive silhouettes subtest, cards are presented that have shadows of 1 or 2 objects, each presentation reveals more recognizable elements of each of the objects.

Face processing was assessed by the long version of The Benton Facial Recognition Test (BFRT, Benton et al., 1994). On each item, subjects are presented with a target photo, and they are asked to choose the target individual from six faces presented simultaneously with the target photo. There are three parts to the BFRT: (1) matching a frontal view of the target with an identical photograph, (2) matching a frontal view of the target individual with three photos of the target taken from different angles, and (3) matching a frontal view of the target individual with three photos of the target taken under different lighting conditions. No time limits are placed on the BFRT, and scores are classified as normal when 41 or above. The black and white photos used in the BFRT consist of unfamiliar male and female faces with their hair and clothing shaded out so that subjects must rely on the face.

Visuospatial processing was examined with the Dot counting, Position discrimination, and Number location subtests of the Visual Object and Space Perception Battery(VOSP). The dot counting subtest required counting random assortments of five to nine dots presented on a card. In the position discrimination subtest, 2 cards are simultaneously presented, one of which has a centrally located dot, the other one an off centred dot. The centrally located dot has to be pointed out. The number location subset involves indicating the position of a dot on one card depending on a sequence of spatially distributed numbers (between 1-9) displayed on a second (simultaneously viewed) card.

Mid-level nonverbal processing

The self-report Vividness of Visual Imagery Questionnaire (Marks, 1995, VVIQ) interrogated the perceptual richness of semantic representations. The VVIQ comprises 16 scenarios such as ‘Visualize the sun rising above the horizon into a hazy sky …to carefully consider the picture that comes before your mind’s eye’ and to rate the vividness of the representation from ‘No image at all’ (1) Clear and vivid as real seeing’ (5). Each scenario is repeated, once with the participant’s eyes open and once with the eyes closed.

Short-term retention and manipulation of spatial representation were examined by spatial span forward subtest (Wechsler Memory Scales, 3rd edition, WMS-3, Wechsler, 1997) and a three-dimensional object rotation task (Cube analysis, VOSP). In the Cube analysis subtest, subjects must count stacked blocks, including any hidden ones, presented as drawings on a card.

High-level nonverbal processing

Spatial working memory and nonverbal reasoning provided estimates of executive functioning. The reverse spatial span subtest (WMS-3) has two broad components, a primary visuospatial storage demand and an active secondary processing demand that reverses the order of a spatial sequence of tapped blocks. The spatial working memory task comprises nine blocks presented in a 3X3x3 arrangement (with each block slightly offset relative to its neighbour blocks. The examiner taps out a sequence which the participant must reproduce in the reserve order. The maximum score on the reverse digit span test is 16. A common feature of working memory tests is the incremental increase in the digit or block span over successive trials. In each case, two trials are presented of the same span length. At least one of the two trials must be correct to progress to the next increment. The test is stopped when both trials of the same span length are failed.

The Brixton Test of Spatial Anticipation examines nonverbal reasoning (Burgess & Shallice, 1997). Participants are presented with a 56-page Brixton Spatial Anticipation Test stimulus booklet. Each page contained an array of 10 circles (2 rows of 5 circles), numbered from 1 to 10. On each page, one circle was blue. The position of the blue circle changed from one page to the next, and the changes are governed by a series of simple rules that changed without warning. Participants are presented with one page at a time and were asked to point to where they thought the blue circle would be on the next page based on the rule inferred from the previous pages. Responses are considered correct if they followed the present rule, and on trials where the rule changed, a response was correct if it followed the previous rule. The total number of errors across 55 trials was used as an outcome measure; higher scores thus reflect worse performance (maximum number of errors = 55).

Verbal processing

Perceptual recognition was examined with degraded letters (Incomplete letters subtest, VOSP); verbal working memory with a digit span task (WMS-3); and executive function by cognitive control (The Hayling Tests of sentence initiation and sentence suppression, Burgess & Shallice, 1997). The Hayling tests consist of two sets of 15 sentences, each missing the last word. In the first section, the examiner reads each sentence aloud, and the participant must complete the sentences. The second part of the Hayling requires participants to complete a sentence with a nonsense ending word (and suppress a sensible one).

Results

The estimate of D.R.'s negative affect and attributional style at the time the neuropsychological assessment took place are presented in Table 4.

(TABLE 4 HERE)

D.R. showed no evidence of negative affect. Her score was more than 2 standard deviations below her psychiatric control group and was at the low end of the normal control group range. On the other hand, she showed maladaptive attribution styles for events with good outcomes and with bad events. The causes of good events were characterised as being unstable, global (her scores fell below the cutoff for both control groups) and over which she had limited control (as compared to the normal control group cut off only). At the same time, she attributed the causes of negative events to internal causes, over which she felt had very little control (her scores fell below the cutoff for both control groups).

The results of the neuropsychological test battery assessing visual cognition are presented in tables 5-7. Of note, D.R. was impaired in recognising ambiguous stimuli across two unrelated paradigms compared to both control groups. The first required naming object silhouettes presented from noncanonical view points, and matching unfamiliar faces viewed from different angles and under different lighting conditions. Conversely, D.R. had no problem completing the low level assessments of visuospatial processing. No other impairments were evident in higher-level visual cognition. Her visual imagery and spatial imagery performance were either within range (visual scene and spatial imagery) or exceeded the top end of the range (spatial span) of both control groups. Finally, high level executive functions dependent on visual cognition were also spared (spatial working memory and spatial anticipation).

(TABLES 5-7 HERE)

D.R.'s verbal cognition profile was strikingly similar to her visual cognition. She evidenced an impairment in naming ambiguous stimuli (degraded letters). Otherwise, verbal short-term memory, verbal working memory, and cognitive control over verbal responses were preserved

Summary of key findings

D.R.'s psychological and neuropsychological profile was marked by a series of striking dissociations. She showed cross domain interoceptive sensibility and body distortion, but there was no evidence of depersonalisation. There was no tendency towards magical thinking or abnormal causal beliefs, but her attribution style was maladaptive despite an absence of negative affect. Her neuropsychological profile was intact apart from a cross-domain impairment in accessing stored information from ambiguous stimuli (naming degraded letters, naming object silhouettes presented from unusual viewpoints, and matching unfamiliar faces viewed under different lighting conditions and or different angles.

Discussion

In this controlled case study, we present the findings of a psychological and neuropsychological investigation of a patient (D.R) in remission from major depressive disorder and Cotard's. The case is noteworthy because as a medically trained doctor, D.R. brought a high level of insight and was able to reflect on her psychotic episode that had occurred four years previously.

During her account of the prodromal phase, several observations are of interest. First, is the synergy between the numbness in her torso and its subsequent elaboration into the delusion belief that 'her insides had dissolved, and food dropped directly from her gullet into her hollow thighs and legs'. A second point of interest is the association between the tingling sensation and distorted body parts during the prodromal phase, and a related third point is their subsequent conspicuous absence from her nihilistic delusion. Another observation of note is the similarity between the spatial representation of the body in Penfield's sensory homunculus, and the sequence of D.R.'s abnormal body sensations. In particular, the tingling sensation felt in two proximal parts of the sensory homunculus (face and fingertips) is reminiscent of an earlier account of cortical remapping of the face and hand area of the sensory homunculus in a group of eight individuals with an upper limb amputation (Ramachandran & Rogers-Ramachandran, 2000) – a point picked up later in the discussion.

The next set of findings document the persistence of trait interoceptive sensibility. D.R. remained affected by body distortion, specifically with regards to their limbs and facial features. D.R. did not mention experiencing any unpleasant odours or feelings of "dirtiness" during the prodromal phase of her delusion or as a feature of her Cotard's However, cross-domain sensory and perceptual accounts are consistent with previous studies on insular stimulation conducted by Penfield.

Of particular note is Penfield's study of 36 epileptic patients with EEG evidence of abnormal electrical activity located over the area of the insula. During awake surgery, electric stimulation of discrete points of the insula patients reported several phenomenological experiences that interest the current case. First, visceral sensations and somatic sensory sensations were elicited. The visceral sensations were experienced in the region of the lower abdomen, in the region between the xiphoid process and the epigastrium or the umbilicus, and described variously as feeling "something funny", " or simply as a nonspecific "sensation". Occasionally, there were associated phenomena such as an unspecified taste or odour. The somatic sensory sensations were experienced as "tingling," "warmth," "numbness," and "vibration" in the lips, tongue, mouth, throat, the face, the head, and contralateral arm, hands, leg, and toes. Oral and upper extremity responses were most frequent and often discrete, limited to the tongue, lips, or fingers. When sensations involve more than one discrete area of the body surface because of a single stimulation, they usually spread to immediately contiguous areas of the sensory homunculus as "for example, from hand to arm, face to hand, toes to the leg, or trunk to arm" (Penfield & Faulk, 1955, p. 456).

As previously mentioned, abnormal perceptual experiences arising after limb amputation links anomalous perceptual experiences and neural plasticity. Ramachandran and Ramachandran (2000) conducted a study on 18 patients who had either arm amputation or brachial avulsion. The study found that 8 patients experienced sensation in their phantom limb which was referred from their face. This was observed in patients who had a map of their hand on their lower face region and the referred sensations were specific to the type of sensation felt. For instance, a patient would feel hot, cold, vibration, rubbing, metal or massage at precisely localized points on their phantom limb. Points on other body parts were generally ineffective in eliciting these sensations in the phantom limb, but there was often a second topographically organized map proximal to the amputation stump. This arrangement of points is precisely what one would expect if the afferents from the upper arm skin and face skin were to invade the hand territory from either side, since the hand area in the Penfield map is flanked on one side by the upper arm and the other side by the face. Anomalous perceptual experiences of the shape and perceived size of body parts have also been reported after regional anaesthesia. For example, Gandevia and Phegan (1999) showed that subcutaneous infiltration of the thumb with lidocaine induced an increase in the perceived size of both the anaesthetized thumb and the unanaesthetised lips in normal subjects. A later study by Paqueron et al. (2003) extended these findings during the course of upper limb, lower limb or spinal anaesthetic blocks in patients (n

= 36) undergoing orthopaedic surgery. They regularly tested sensory and motor function and analysed the relationship between body image distortions and impairment progression. Most patients experienced perceptual changes in the shape and size of the deafferented limb, and these changes differed from postural illusions. Nearly half of the patients felt their unanaesthetised lips and/or mouth swelling during upper limb block, indicating dynamic interactions between cortical representations. The authors concluded that the perception of body shape and awareness of its postural variations are built from different models, and peripheral afferent activity maintains a unified body image. Although this conclusion is based on a different phenomenology to Cotards, but features overlap with D.R.'s sensations of tingling and swelling of facial features during the prodromal stage of her delusion. Importantly, this study shows that changes in interoception sensibility can occur during the normal population, are reversible, and involve the integration of (aberrant) afferent information with cortical body representation.

D.R.'s neuropsychological profile revealed a disturbance of low-level perceptual processing, which stands in stark contrast to her spared cognitive and executive abilities. D.R's problems matching unfamiliar faces is consistent with Cotard's earlier observations and other case studies of patients with Cotard's delusion, such as the case reports of K.H (Wright et al., 1993) and R.B. (Leafhead & Kopelman, 1997). In both instances, formal neuropsychological testing revealed impairments in the forced choice recognition of unfamiliar make faces and labelling of emotional facial expressions. A study conducted by Darke et al. (2021) examined face processing in 86 psychiatric inpatients with schizophrenia and bipolar disorder, as well as in 20 healthy controls. The study found that abnormalities in face processing were not dependent on diagnosis, and suggested that factors relevant to the task, such as attention, could be responsible for the findings rather than problems in face processing itself. If we assume that major depressive disorder (with psychotic features) is also included in this transdiagnostic conclusion, then D.R. should have exhibited impairments on attention-demanding tasks, particularly spatial working memory, which as already stated, was intact (Fabry, 2020; Song et al., 2021).

An alternative perspective is to consider D.R.'s ability to perceive 'degraded visual percepts', which becomes significant during the early phase of an illness. In these circumstances, the balance of sensory signals generated internally and those perceived externally shifts dynamically, favouring the internal signals. As a result, the conflicting signals are not integrated and are pushed away from each other (Kirsch & Kunde, 2023). This could explain why D.R. experienced distorted features and the disappearance of her torso during the early phase of the illness, without being influenced by external sensory signals from vision, such as looking at her reflection in the mirror or through touch

In this report, we document a case of Cotard's characterized by the presence of bodily distortions and abnormal somatosensory sensations during the prodromal phase, which exhibited a spatial correlation with the somatosensory homunculus and areas of heightened sensory innervation. During the remission phase, the patient displayed signs of visual perceptual impairments dissociated from an otherwise intact neuropsychological profile. Additionally, subtle manifestations of internally generated bodily signals, such as distortion, peculiar tastes, and feelings of uncleanness, were noted, along with an external attribution style. Collectively, these features are posited to represent stable or mediating vulnerability factors rather than transient indicators of dysfunction. This observation aligns with the notion that organic or functional involvement in Cotard's syndrome manifests when information processing systems are subjected to high-stress levels during the prodromal and active phases of functional disorders. Inherent weaknesses within information processing systems and high-level attribution biases may be artificially exposed under direct measurement by challenging the processing systems with specific stimuli and tasks that are particularly challenging. It is conceivable that a breakdown in multimodal sensory integration, in conjunction with affective processing and impaired reasoning, operates in a complex interaction to give rise to Cotard's syndrome.

Declarations

Ethics approval and consent to participate: The research protocol was in line with the Declaration of Helsinki and was approved by the South Staffordshire Local Research Ethics Committee (REC reference 08/H1203/46). Patient D.R. gave informed consent to participate in the study, which was conducted in accordance with Good Clinical Practice guidelines.

Consent for publication: Written informed consent for the publication was obtained from the patient.

Data Availability Statement: The data that support the findings of this study are available from the corresponding author, NMJE, upon reasonable request.

Competing interests: The aauthors declare that they have no competing interests.

Author's contribution: conception and design of the work: NMJE, FO; data collection and analysis: NMJE & EDR; data interpretation and manuscript preparation: NMJE, EDR, SW, FO. The authors have read and approved the manuscript.

Funding: No funding was obtained for this study.

Acknowledgements: The authors would like to thank patient D.R. sincerely.

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TABLES AND FIGURES

Figure Legend

Fig. 1. Whole brain structural magnetic resonance images showing sagittal, coronal and horizontal views are presented in the upper panel. The following brain structures are referenced: The Red nucleus (1); The Mammillary bodies (2); The anterior portion of the hippocampus (3); The fronto-parietal operculum (4); The Insular cortex (6); The Caudolenticular grey bridges (7). The lower panel shows a coronal section through the insular showing a: Circular sulcus of insula; b: short gyri of insular; c: Central sulcus of insular; d: long gyrus of insular. Mapped according to Nieuwenhuys, Voogd & van Huijzen (2007), The Human Central Nervous System, 4th edition.

Fig. 2.

A schematic representation of the progression of patient D.R.'s disordered body perception.

Table Legend

Table 1. D.R's Scores on The Cambridge Depersonalization Scale and The Body Distortion Questionnaire

Table 2. D. R's scores on The Magical Ideation Scale and The Belief in Divine Intervention Scale

Table 3. Age and premorbid estimate of IQ for D.R. and both control groups.

Abbreviations: IQ, Intelligence Quotient; MDD control group, control group with a diagnosis of Major Depressive Disorder.

Table 4. Measures of negative affect and attribution style for D.R., the psychiatric and normal control groups.

Notes and Abbreviations: The Attribution Style Questionnaire's performance evaluation has two panels - upper and lower. The upper panel displays the attributional style and locus of control for events with good outcomes, whilst the lower panel shows the same for events with bad outcomes. D.R.'s scores which fall outside the control 2 standard deviation cutoff are highlighted. SD, Standard deviation.

Table 5. Measures of low level visual object perception and visuospatial processing for D.R., the psychiatric and normal control groups.

Notes and abbreviations: D.R.'s scores which fall outside the control 2 standard deviation cutoff are highlighted. VOSP, Visual Object and Space Perception Battery.

Table 6. Measures of visual scene imagery and spatial imagery for D.R., the psychiatric and normal control groups.

Notes and abbreviations: D.R.'s scores which fall outside the control 2 standard deviation cutoff are highlighted. VVIQ, Vividness of Visual Imagery; VOSP, Visual Object and Space Perception Battery; WMS-3, Wechsler Memory Scales, 3rd edition.

Table 7. Measures of spatial working memory and spatial fluid reasoning for D.R., the psychiatric and normal control groups.

Notes and abbreviations: D.R.'s scores which fall outside the control 2 standard deviation cutoff are highlighted. WMS-3, Wechsler Memory Scales, 3rd edition

Table 8. Performance on measures of verbal processing for D.R, the psychiatric and normal control group

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Table 1. D.R's Scores on The Cambridge Depersonalization Scale and The Body Distortion Questionnaire

Depersonalisation		**Body Distortion**	
Never	26	No	73
Rarely	2	Yes	5
often	1	Undecided	3

Table 2. D. R's scores on The Magical Ideation Scale and The Belief in Divine Intervention Scale

Magical Ideation		**Divine Intervention**	
TRUE	7	Agree	2
FALSE	23	Disagree	4

Table 3. Age and premorbid estimate of IQ for D.R. and both control groups.

	Age	**IQ**
D.R.	55	124
Psychiatric control group		
Mean	55.5	123.75
1 SD	0.7	4.03
Normal control group		
Mean	66.3	119.25
1SD	7.8	0.50

1

Table 4. Measures of negative affect and attribution style for D.R., the psychiatric and normal control groups.

	The Hamilton Depression Inventory	The Attributional Style Questionnaire: **events with good outcomes**				
	Negative affect	**Cause: external - internal**	**Stability: temporary-permanent**	**Pervasiveness : specific-global**	**Importance: not- very**	**Control : none-full**
D.R.	**2.5**	5.67	**3.83**	**3.33**	6.83	3.5
Psychiatric control group						
Mean	15.70	4.46	4.88	4.71	5.79	4.38
1 SD	6.38	0.70	0.52	0.57	0.32	1.46
2 SD range	**2.95**-28.45	3.06-5.86	**3.84**-5.91	**3.57**-5.84	5.16- **6.42**	1.45-7.00
Normal control group						
Mean	4.83	5.46	5.29	5.58	6.54	5.84
1SD	2.00	0.80	0.69	0.29	0.46	0.80
2 SD range	0.83-8.83	3.87-7.05	**3.92** -6.67	**5.00**-6.16	5.62-7.46	**4.23-**7.44
		The Attributional Style Questionnaire: **events with bad outcomes**				
D.R.		**7.00**	5.17	4.17	6.33	**2.83**
Psychiatric control group						
Mean		4.58	5.00	4.67	6.17	4.01
1 SD		0.93	0.49	0.38	0.24	0.49
2 SD range		2.73-**6.44**	4.02-5.98	3.90-5.44	5.70-6.64	**3.02-**4.98
Normal control group						
Mean		5.29	3.42	2.79	6.38	5.25
1SD		0.69	1.23	0.79	0.44	0.91
2 SD range		3.92-**6.67**	0.95-5.89	1.20-4.38	5.50-7.25	**3.44-**7.06

Notes and Abbreviations: The Attribution Style Questionnaire's performance evaluation has two panels - upper and lower. The upper panel displays the attributional style and locus of control for events with good outcomes, whilst the lower panel shows the same for events with bad outcomes. D.R.'s scores which fall outside the control 2 standard deviation cutoff are highlighted. SD, Standard deviation.

Table 5. Measures of low level visual object perception and visuospatial processing for D.R., the psychiatric and normal control groups.

	Visual object perception tests (VOSP)			Benton Test of Unfamiliar Face Matching	Visuospatial tests (VOSP)		
	Object naming	**Object decision**	**Progressive silhouettes**	**Unfamiliar male faces**	**Dot Location**	**Position Discrimination**	**Number Location**
D.R	**22**	14	**13**	**39**	10	20	9
Psychiatric control group							
Mean	29.75	19.25	9.50	47.25	9.75	20.00	9.25
1SD	0.50	1.50	1.29	3.59	0.50	0.00	1.50
2 SD range	**28.75-** 30.00	16.25-20.00	6.92-**12.08**	**40.06**-54.44	8.75-10.00	20.00-20.00	6.25-10.00
Normal control group							
Mean	25.75	18.25	7.75	47.25	9.25	19.50	9.25
1 SD	3.86	0.96	2.06	2.60	1.50	1.00	0.96
2 SD range	18.03-33.47	16.34-20.00	3.63-**11.87**	**42.05**-52.45	6.25-10.00	17.50-20.00	7.34-10.00

Notes and abbreviations: D.R.'s scores which fall outside the control 2 standard deviation cutoff are highlighted. VOSP, Visual Object and Space Perception Battery.

Table 6. Measures of visual scene imagery and spatial imagery for D.R., the psychiatric and normal control groups.

	Visual imagery-eyes open (VVIQ)	Visual imagery - eyes closed (VVIQ)	Spatial Imagery (Cube analysis, VOSP)	Spatial Span WMS-3)
D.R.	2.56	4.38	9	9
Psychiatric control group				
Mean	2.73	3.72	9.75	7.25
1SD	1.56	0.95	0.50	0.50
2 SD range	0.00-5.85	1.81-5.63	8.75-10.00	6.25-**8.25**
Normal control group				
Mean	2.38	2.75	9.50	5.25
1 SD	0.77	1.51	0.58	0.96
2 SD range	1.00-3.92	0.00-5.77	8.35-10.00	3.34-**7.16**

Notes and abbreviations: D.R.'s scores which fall outside the control 2 standard deviation cutoff are highlighted. VVIQ, Vividness of Visual Imagery; VOSP, Visual Object and Space Perception Battery; WMS-3, Wechsler Memory Scales, 3rd edition.

Table 7. Measures of spatial working memory and spatial fluid reasoning for D.R., the psychiatric and normal control groups.

	Spatial Working Memory (WMS-3)	Brixton Test of Spatial Anticipation
D.R.	8	5
Psychiatric control group		
Mean	8.25	8.25
1SD	2.06	2.06
2 SD range	4.13-12.37	4.13-12.37
Normal control group		
Mean	6.00	4.50
1 SD	0.82	2.38
2 SD range	4.37-7.63	0.00-9.26

Notes and abbreviations: D.R.'s scores which fall outside the control 2 standard deviation cutoff are highlighted. WMS-3, Wechsler Memory Scales, 3rd edition

Table 8. Performance on measures of verbal processing for D.R, the psychiatric and normal control group

	Incomplete Letters (VOSP)	Verbal Span (rehearsal, WMS-3)	Verbal Working Memory (WMS-3)	Hayling Tests of sentence Initiation and sentence suppression
D.R.	**18**	14	8	6
Psychiatric control group				
Mean	20.00	12.25	9.00	7.00
1SD	0.00	2.87	2.94	1.83
2 SD range	20.00-20.00	6.51-17.99	3.11-14.89	3.35-10.65
Normal control group				
Mean	19.50	10.50	9.00	5.50
1 SD	0.60	3.42	4.16	1.00
2 SD range	18.30-20.00	3.67-17.33	0.67-17.33	3.50-7.50

Notes and abbreviations: D.R.'s scores which fall outside the control 2 standard deviation cutoff are highlighted. VOSP, Visual Object and Space Perception Battery; WMS-3, Wechsler Memory Scales, 3rd edition.

Figure 1. D.R's Whole brain structural magnetic resonance images showing sagittal, coronal and horizontal views are presented in the upper panel. The lower panel shows a coronal section through the insular. Mapped according to Nieuwenhuys, Voogd & van Huijzen (2007), The Human Central Nervous System, 4th edition.

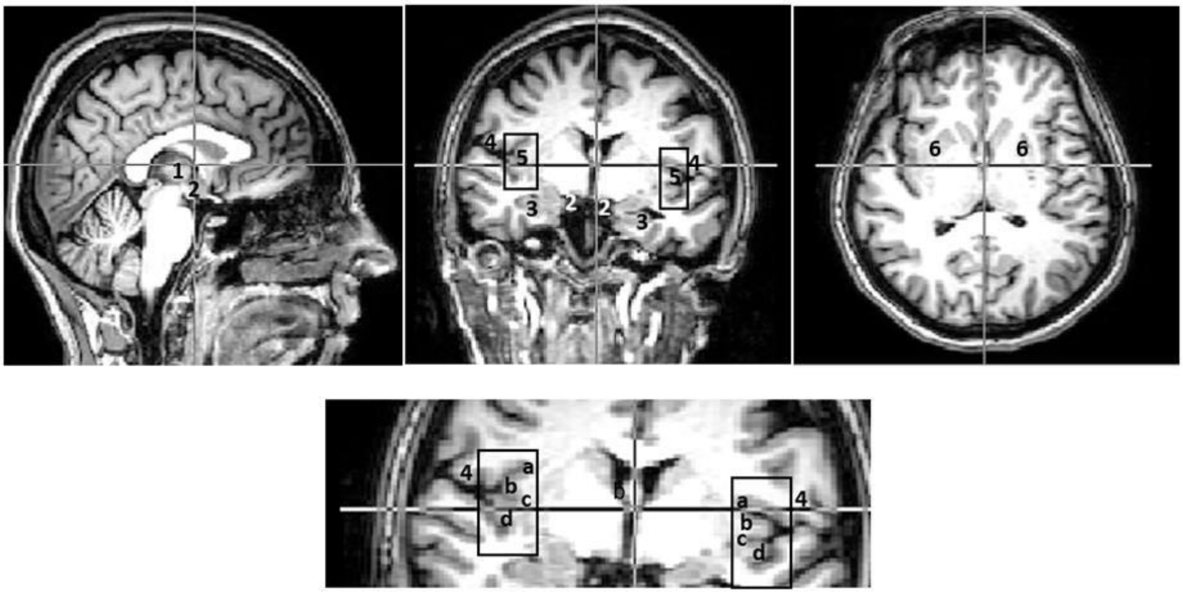

Figure 2. A schematic representation of the progression of D.R.'s disordered body perception.