"Need to Know" or the Strong Urge to Find Names of Unique Entities in Acquired Obsessive-Compulsive Disorder

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Abstract: The two forms of obsessive-compulsive disorder (OCD), idiopathic and acquired, have been linked to abnormalities in the fronto-striato-thalamo-cortical circuitry, involving the orbitofrontal cortex, anterior cingulate cortex, thalamus, and striatum. Accumulating evidence indicates that damage to other brain regions (ie, temporal lobes) is also implicated in the pathogenesis of both types of OCD. In addition, some discrete OCD symptoms have received less attention because of their presumed low occurrence and difficultly of categorization. Among these, one intriguing and potentially severe type of obsessive thinking is the so-called "need to know" (NtK), which is a strong urge to access certain information, particularly proper names. In some patients, this monosymptomatic presentation may constitute the major feature of OCD. Here we report the cases of two patients who developed NtK obsessions with tenacious time-consuming, answer-seeking compulsions as the only or more disabling symptomatology in association with malignant tumors involving the right temporal lobe and connected fronto-subcortical circuits.

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AC = anterior commissure. **A-OCD** = acquired obsessive-compulsive disorder. **aTR** = anterior thalamic radiations. **IFOF** = inferior fronto-occipital fasciculus. **ILF** = inferior longitudinal fasciculus. **I-OCD** = idiopathic obsessive-compulsive disorder. **NtK** = need to know. **OC** = obsessive-compulsive. **OCD** = obsessive-compulsive disorder. **OFC** = orbitofrontal cortex. **ROI** = region of interest. **UF** = uncinate fasciculus.

Obsessive-compulsive disorder (OCD) is defined by the presence of repetitive, intrusive thoughts and images (obsessions) and senseless, time-consuming, complex rituals (compulsions) (American Psychiatric Association, 2013). In most cases, the obsessions and compulsions cause great distress, are time consuming (> 1 hour a day), and substantially interfere with normal functioning (American Psychiatric Association, 2013). A "poor insight" subtype of OCD was included in the *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition* (American Psychiatric Association, 1994), and the insight level in OCD was specified further in the *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition* (American Psychiatric Association, 2013).

The most widely accepted model of idiopathic obsessive-compulsive disorder (I-OCD) proposes that a dysfunction of fronto-striato-thalamo-cortical circuitry is responsible for the emergence and maintenance of symptoms. There is a growing consensus, however, that OCD is not a homogeneous condition, in either its clinical phenomenology or its pathophysiological mechanisms (Berthier, 2000; Mataix-Cols et al, 2004). In a systematic review concerning widespread structural brain changes in patients with OCD, Piras et al (2015) described a far-reaching network of cerebral dysfunction, including the dorsolateral prefronto-striatal "executive" circuit and the reciprocally connected temporo-parieto-occipital associative areas. These results support the notion that the brain

alterations responsible for OCD are represented at a network level, and that widespread structural abnormalities may contribute to neurobiological vulnerability to develop the disorder (Piras et al, 2015). Several studies have described the existence of different symptom dimensions or clusters of OCD symptoms (ie, symmetry/ordering, harm/checking) (Berthier, 2000; Mataix-Cols et al, 2004; van den Heuvel et al, 2009) and have proposed that these discrete symptom dimensions may have a distinct neural substrate (Pujol et al, 2004; van den Heuvel et al, 2009).

In addition, the understanding of acquired obsessive-compulsive disorder (A-OCD) is increasing (Berthier et al, 1996, 2001; Carmin et al, 2002; Chacko et al, 2000; Coetzer, 2004; Jenike and Brandon, 1988; Kant et al, 1996; Kumar et al, 2009; Salinas et al, 2009). Patients with A-OCD present a precipitous or insidious onset of OCD symptoms associated with focal brain damage or neurodegenerative diseases, such as semantic dementia (Green-Heredia et al, 2009; Snowden et al, 2001). A-OCD has been discovered in patients with structural lesions in brain areas that are traditionally implicated in I-OCD, but it also has been seen in patients with structural lesions in other regions that are not generally involved in I-OCD (Anderson et al, 2005; Carmin et al, 2002; Figee et al, 2013; Rai et al, 2011; Simpson and Baldwin, 1995). Berthier et al (1996) found that six of 13 patients with A-OCD had structural damage in the temporal lobes. Other authors have also described right temporal involvement in patients showing either shopping compulsions, fear of harming (Rai et al, 2011), or intrusive musical tunes (Zungu-Dirwayi et al, 1999). There is a need for further assessment of A-OCD because, even though it shares some neural, behavioral, and psychological correlates with I-OCD, the two disorders have phenomenological and neurologic differences (Berthier et al, 1996; Huey et al, 2008).

Despite progress in identifying homogeneous and clinical meaningful subphenotypes of OCD (Alonso et al, 2008; Pinto et al, 2008), some monosymptomatic presentations of OCD have received less attention owing to their presumed low occurrence and difficulty of categorization. Among these, one intriguing and potentially severe type of obsessive thinking is the so-called "need to know" (NtK). NtK is defined as the obsession of accessing certain types of information associated with compulsions manifested primarily by intensive checking and rechecking of the accuracy of this information (Carmin et al, 2002). This putative OCD behavior is regularly grouped under a "miscellaneous" category together with other a priori unusual symptoms, such as superstitious fears or being bothered by noises (Goodman et al, 2006). In some patients, however, NtK may be the principal monosymptomatic feature of OCD (Carmin et al, 2002; Simpson and Baldwin, 1995; Swoboda and Jenike, 1995). For example, Lemelson (2003) studied 19 patients with OCD in Bali, Indonesia, and found that the culture shaped the expression of OCD symptoms. Specifically, these patients displayed an obsessional NtK about their social network, such as the identity and status of passersby (Lemelson, 2003).

One of the most striking examples of isolated NtK was reported by Swoboda and Jenike (1995), who described an elderly man with late-onset, treatment-resistant OCD in association with a large right posterior frontal infarct. A brief excerpt from that case report follows (Swoboda and Jenike, 1995, p 2121):

During a television program, he recognized an actor in a commercial but was unable to recall his name. He became extremely anxious and went to great lengths to find the answer, calling several acquaintances and ultimately the [television] network.

To date, there have been no reports of the occurrence of NtK behavior resulting specifically from temporal lobe damage. Here, we describe two such patients. Based on our findings, we hypothesized that NtK behavior may be caused by an abnormal interaction of anatomically linked cortical regions (temporal, orbitofrontal cortex [OFC], and prefrontal cortex) due to lesion-induced disconnections of white matter pathways, which negatively influence the activity of subcortical structures.

CASE REPORTS

Two male patients developed an almost monosymptomatic NtK behavior in association with malignant tumors involving the right temporal lobes. The two patients were referred to us by one of the authors (C.G.-H.), who worked as a neuropsychologist in a local hospital. The patients were referred to us due to our research interest in A-OCD and to the exceptional occurrence of the main and most disabling obsessive-compulsive (OC) symptom: NtK. Both patients signed a written informed consent to participate in this study, and the study protocol was approved by the ethical committee of the University of Málaga, Spain.

Patient 1

Patient 1 was a 46-year-old right-handed businessman who first experienced OC symptoms 6 months before referral to our unit. His family and personal history were unremarkable. The patient reported that while visiting London, he was suddenly overwhelmed by the strong urge to know the name of a London neighborhood, and he ruminated throughout the night until he eventually accessed the target name. One week later, on his return to Spain, the patient gradually began to experience dizziness, palpitations, tiredness, decreased motivation, headaches, and parosmia (olfactory hallucination of gas). During a first neurologic evaluation, his EEG showed normal results, but a structural MRI revealed a large right temporal lobe tumor. The patient then underwent a right anterior temporal lobectomy (excision) including lateral and mesial structures. The histopathological diagnosis was a glioblastoma multiforme. The immediate postoperative cerebral MRI confirmed the complete excision of the tumor.

Six months after his discharge from the hospital, the patient became obsessed with knowing the names of actors or actresses, writers, or flamenco singers and with finding

the meaning of acronyms (ie, What does ONU mean?). The thematic content of NtK symptoms emerged in blocks, meaning that one week he would be obsessed with proper names, but another week, it would be acronyms. The patient's searching compulsions lasted several hours and only abated when he accessed the desired name. He reported frequent NtK intrusions, causing him mild to moderate distress. These compulsions also caused significant social impairment, but the patient had poor insight on the excessiveness and unreasonableness of his obsessional pursuits.

An evaluation of the long-term stability of the patient's neurologic status was performed $1^1/_2$ years after surgery. The MRI scan obtained at that time showed a cavity in the right middle cranial fossa, resulting from the temporal lobectomy, and extensive T_2-weighted hyperintense lesions surrounding the right temporo-occipital region, medial thalamic nuclei, and pulvinar (Figure 1A). The right lateral ventricle and the third ventricle were moderately dilated. Several months later, an 11C-methionine PET ruled out the presence of tumor recurrence. An ^{18}F-fluorodeoxiglucose PET showed significant decreased metabolism in the right temporal lobe, OFC, ventral striatum, caudate/putamen, and thalamus, with increments in metabolic activity in the left thalamus and medial insula. The patient was treated with cranial radiotherapy and chemotherapy, but treatment of NtK was not possible because he developed a disseminated gliomatosis and died soon afterward.

Patient 2

Patient 2 was a 35-year-old, right-handed man who first experienced OC symptoms 3 months after excision of a right anterior temporal tumor. The histopathological diagnosis was an astrocytoma type II in the right temporal lobe, with extension into the insula, external capsule, hippocampus, and mammillary bodies. The lesion also showed an extension to the posterior right OFC. A right temporal lobectomy (excision) was performed without complications. Before the operation, the patient had worked as a high school music teacher, but he did not return to his position after the operation. His family and personal history were unremarkable.

In the postsurgical clinical interview, the patient complained about mental and speech slowness, personality changes, irritability-impulsivity, sexual inactivity, and substantial NtK obsessions. The patient reported that one day he was on a bus with his father, passing by a certain neighborhood, when he suddenly became obsessed with knowing in which house his mother had lived during childhood. The urge grew so strong that, while his father was reflecting on the question, he felt compelled to call his mother for the answer. Ultimately, the patient was able to relax.

Since the surgical intervention, the patient had become more meticulously aware of, and scrutinized in greater detail, people, situations, and conversations, showing a tendency to try to solve immediately any doubts that arose in him by questioning his relatives or the person with whom he talked. The patient admitted that he could be searching for information several hours a day (on average between 5 and 7 hours) and that he did not calm down until he found the answer.

In general, the patient was unaware of his time-consuming OC behavior and its negative consequence for his social relations. In fact, he sometimes revealed a strong interest in other people's cultural background and became haunted by asking and learning as much information as possible about their historic background, showing empathic curiosity (McEvoy et al, 2013). The patient's neurologic examination was conducted by one of the authors (M.L.B.), who was originally from Argentina. Upon discovering this fact, the patient immediately asked questions covering an immense amount of information about that country (eg, Do you know Eva Perón? Where was she born? Where and when did she die?).

These peculiar NtK behaviors only manifested when the patient showed an interest in other people. He admitted that only certain individuals ignited his curiosity and the urge to look up information, whereas he neglected and depreciated other people, such as his wife, on the basis of not being interesting or smart. The patient exhibited OCD with poor insight, with no recognition of the deleterious effects of his behavior, and he was reluctant to take the antidepressant medication (escitalopram) prescribed to him and to undergo psychotherapy.

PATIENT ASSESSMENTS

Cognitive and Psychiatric Assessment

We assessed both patients using cognitive tests that tap executive, attention, memory, and language functions as well as psychiatric scales that rate the symptoms, traits, and severity of OCD.

Neuroimaging

MRI Acquisition

The MRI studies of Patient 1 were performed using a 1.5T MRI scanner (Signa Excite, GE Medical Systems). T_2-weighted images of the whole brain were acquired (repetition time: 1002 ms; echo time: 158 ms; inversion time: 2300 ms; matrix size: 512 × 512; field of view: 240 × 240; flip angle: 90 degrees; slice thickness: 5 mm; voxel size: 0.46 mm × 0.46 mm × 5 mm; slice gap: 1.4 mm, 23 slices).

The MRI studies of Patient 2 were performed using a 1.5T MRI scanner (Philips Achieva) equipped with a six-channel Philips SENSE head coil. T_1-weighted images of the whole brain with three-dimensional magnetization-prepared rapid acquisition gradient echo sequence were acquired (repetition time: 7.03 ms; echo time: 3.17 ms; matrix size: 288 × 288; field of view: 266 × 266; flip angle: 8 degrees; slice thickness: 0.9 mm; voxel size: 0.92 mm × 0.92 mm × 1 mm, 200 contiguous slices). In addition, MRI images of 25 healthy controls (mean age: 56 ± 5 years; range: 48–67 years) were acquired in order to optimize the normalization of the PET images. The acquisition parameters of the controls have been reported elsewhere (García-Casares et al, 2014).

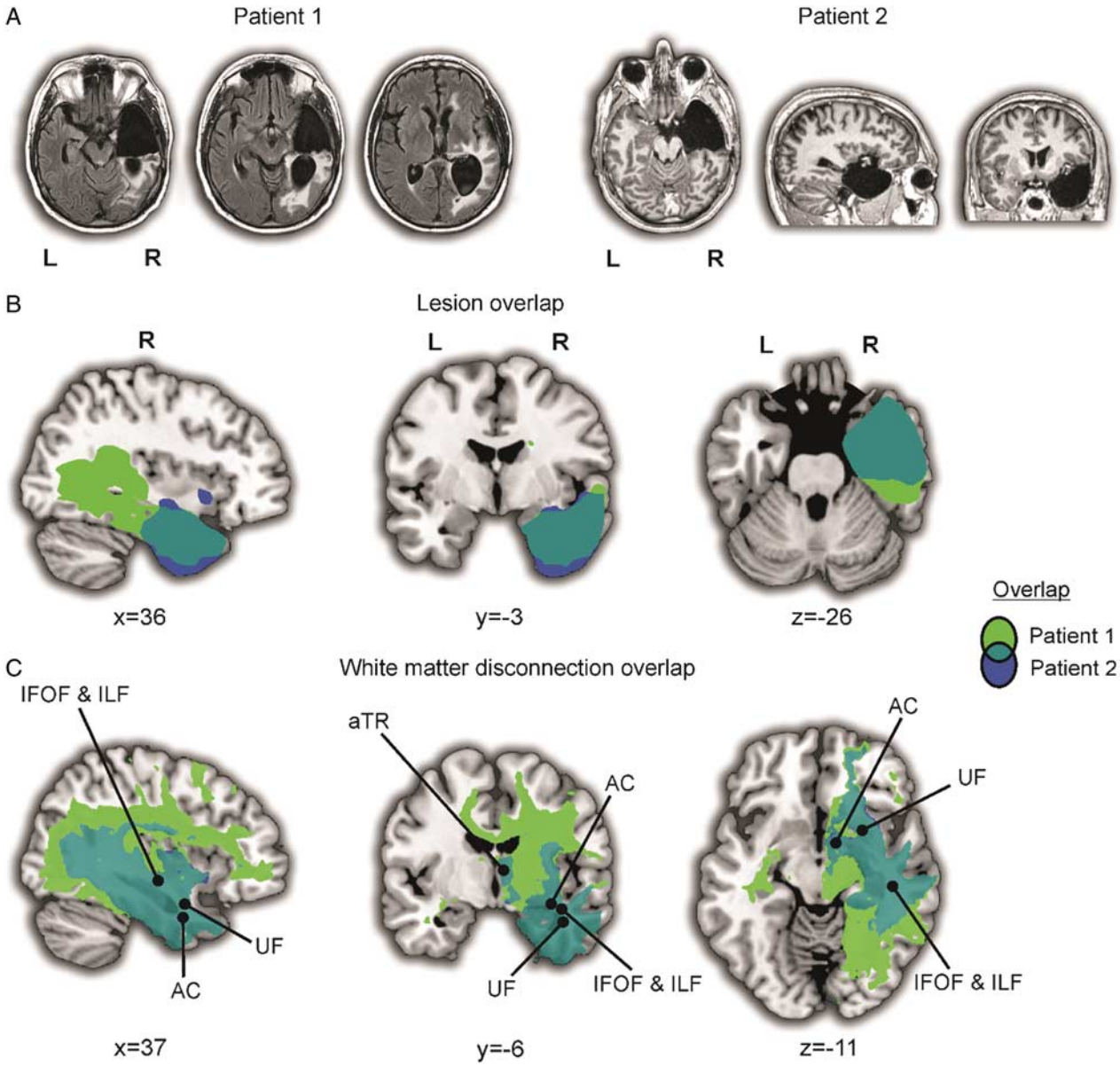

FIGURE 1. Lesions and white matter Disconnectome maps for Patients 1 and 2. **A:** Images of the right-hemispheric structural lesion and resection in native space using T_2-weighed images in axial view (Patient 1) and T_1-weighted images in axial, sagittal, and coronal views (Patient 2). **B:** Lesion overlap across both patients in MNI space. **C:** Disconnectome maps' overlap across patients is depicted in dark green, involving parts of tracts of interest. Neurologic convention is used. Maps are shown using a probability of disconnection greater than 0.9. **AC** = anterior commissure. **aTR** = anterior thalamic radiations. **IFOF** = inferior fronto-occipital fasciculus. **ILF** = inferior longitudinal fasciculus. **L** = left. **R** = right. **UF** = uncinate fasciculus. (Figure 1 can be viewed in color online at www.cogbehavneurol.com.)

Tract-wise Lesion Analysis

We used both Tractotron and Disconnectome Maps software, which are part of the Brain Connectivity and Behaviour toolkit (available at http://www.bcblab.com/BCB/Software.html), to identify (a) the regions of white matter damage that overlapped for the two patients and (b) the proportion of damage (lesion load) of the tracts of interest for each patient.

First, we manually delineated the lesions for Patient 1 and 2 on the T_1-weighted and T_2-weighted images, respectively, in native space using MRIcron software (Rorden and Brett, 2000). Binarized lesion masks were normalized to the MNI152 standard space using Statistical Parametric Mapping (SPM 12) software (Welcome Department of Imaging Neuroscience, University College, http://www.fil.ion.ucl.ac.uk/spm/). Normalized lesion masks were used as input for the Disconnectome Maps software and as seed points to identify which tracks pass through the damaged regions by comparison with a reference tractography data set of 10 healthy controls (Rojkova et al, 2016).

This method allowed us to identify the areas that might be disconnected and the white matter tracts that are likely to be interrupted by the lesion. The Disconnectome map of each patient was thresholded at 0.9, indicating a probability of disconnection greater than 90% due to the lesion, and then the two maps were overlaid on each other in order to explore the regions that were most likely to be disconnected in the two patients.

Next, we evaluated the severity of the disconnection in each patient by measuring the proportion of each tract of interest that was affected by the lesion via Tractotron software. Tractotron computes both the probability of a given tract's being affected by the lesion and its proportion (ie, the number of damaged voxels in the tract divided by the total volume of the tract). The tracts of interest to be explored with Tractotron were selected on the basis of their presumptive relation with OCD and the semantic system: uncinate fasciculus (UF), anterior commissure (AC), forceps minor, anterior thalamic radiations (aTR), inferior fronto-occipital fasciculus (IFOF), and inferior longitudinal fasciculus (ILF).

PET Image Acquisition

^{18}F-fluorodeoxiglucose PET images were acquired in Patient 1 and 25 healthy controls using a PET/CT camera (Discovery ST, General Electric) after an intravenous injection of approximately 2.82 MBq/kg in Patient 1 and 3.3 MBq/kg in the 25 healthy controls. The transversal and axial scanner resolutions were 4.4 mm and 7.2 mm full width at half maximum, respectively. The PET images were reconstructed using the three-dimensional FORE-IR algorithm with CT attenuation correction (matrix size: $128 \times 128 \times 47$; voxel size: $2.34 \times 2.34 \times 3.27$ mm).

PET Analysis

We performed spatial preprocessing and statistical analyses using Statistical Parametric Mapping software, version SPM12 (available at http://www.fil.ion.ucl.ac.uk/spm/), running on MATLAB R2017b (Mathworks Inc). T_1-weighted images and PET images were aligned to anterior-posterior commissure orientation. The reoriented PET images were then co-registered with the structural images. A mask including the lesion and ventricles of Patient 1 was applied to the T_2-weighted image to exclude damaged areas from the normalization process. The PET co-registered volumes of Patient 1 and the healthy controls were then spatially normalized onto the MNI template and smoothed with a full width at half maximum 8-mm Gaussian kernel.

Histogram-based intensity normalization was performed using in-house software. For this purpose, the mean of the smoothed PET images of the healthy controls was calculated. Then, the smoothed images of each participant were voxel-wise divided by this mean image. Histograms of the ratio images were generated, excluding lesioned areas and ventricles for Patient 1. Each smoothed PET study was then divided by the most prevalent value in the ratio image. SPM12 analysis was carried out with the resulting PET studies.

A set of brain regions of interest (ROIs) was preselected to perform voxel-based comparisons with small-volume correction. The ROIs included for analysis were anterior insula, head of the caudate nucleus, OFC, and thalamus, bilaterally. These ROIs were selected from the Wake Forest University PickAtlas Tool (Maldjian et al, 2003), with the exception of the right caudate head. This ROI was hand-drawn over the T_2-weighted image of the centrum semiovale and spatially normalized onto the MNI template using the calculated deformation fields for the T_1-weighted scan of Patient 1. In this analysis, voxels were regarded as significant when falling below a corrected voxel threshold of 0.05 (family-wise error corrected) adjusted for the small volume.

RESULTS

Cognitive and Psychiatric Findings

As depicted in Table 1, both patients showed impairment in tests of executive function, including the Trail-Making Test (Army Individual Test Battery, 1944) (parts A and B in Patient 1) and the Road-Map Test of Direction Sense (Money et al, 1965) (Patient 2). Memory for scenes (ie, the Scenes I subtest from the Wechsler Memory Scale, Third Edition, Spanish version [Wechsler, 2004]) was markedly impaired in both patients, but the patients achieved average scores on the other cognitive tests, except Patient 2 scored below average on Part B of the Trail-Making Test and on the Symbol Digit Modalities Test of the Wechsler Adult Intelligence Scale, Third Edition, Spanish version (Wechsler, 1999). The total Yale-Brown Obsessive Compulsive Scale (Goodman et al, 1989) score indicates that both patients suffered from moderate to severe OCD. Both patients had clinically significant OCD, highlighting the NtK symptomatology as the most prominent and disabling feature (although other less disabling and time-interfering symptoms, such as checking doors, were also present).

Neuroimaging Findings

Tract-wise Lesion

In addition to the high overlap across brain lesions at the level of the right anterior temporal lobe in both patients (Figures 1A and B), the Disconnectome maps and Tractotron software showed high overlap between the areas that were indirectly affected by the lesion (ie, disconnected) in both patients (Figure 1C). We evaluated the severity of the disconnection in each patient by measuring the proportion of each tract of interest that was affected by the lesion (ie, number of voxels of a given tract superimposed with the lesion mask divided by the total number of voxels of the tract), as shown in Table 2. Table 2 also shows the probability of a given tract's being affected by the lesion. As expected, all of the a priori selected pathways (UF, AC, aTR, IFOF, ILF) in the right hemisphere were affected because they travel through the anterior part of the temporal lobe (Figure 1C). The forceps minor, a white matter bundle that connects the lateral and medial surfaces of the frontal lobes, was affected only in Patient 1 (Figure 1C, medial image).

TABLE 1. Results of Two Patients' Neuropsychological Testing and Obsessive-Compulsive Disorder (OCD) Measures

		Patient 1		Patient 2	
Domain	**Test/Subtest**	**Raw Score**	**Range**	**Raw Score**	**Range**
Executive Function	Trail-Making Test A (time in seconds)	151	Impaired	52	Below average
	Trail-Making Test B (time in seconds)	408	Impaired	80	Average
	Road-Map Test of Direction Sense	29/32	Average	23/32	Impaired
	Phonological Fluency[1] ("P," 1 minute)	17	Average	14	Average
Attention	Digit forward	8	Average	6	Average
	Digit backward	6		5	
	Block Tapping Test (total score; WMS–III)	70	Average	30	Average
	Symbol Digit Modalities Test (WAIS–III)	100	Average	29	Below average
Memory	Scenes I (WMS–III)	26/64	Impaired	21/64	Impaired
	Word List (WMS–III)	31/48	Average	31/48	Average
	Faces I (WMS–III)	40/48	Average	35/48	Average
	Rey Osterreith Complex Figure[2]				
	Copy	35/36	Average	NT	NT
	Delayed recall	15/36	Average		
Language	Pyramids and Palm Trees Test	NT	NT	52/52	Average
	Boston Naming Test	57/60	Average	59/60	Average
	Semantic Fluency (Animals, 1 minute)	19	Average	21	Average
OCD Measures	Leyton Obsessional Inventory[3]	Symptoms: 3 Traits: 2		Symptoms: 10 Traits: 9	
	Yale-Brown Obsessive Compulsive Scale				
	Obsessions	13		10	
	Compulsions	11		8	
	Total	24	Clinically significant	18	Clinically significant

[1]Benton et al, 1994.
[2]Rey, 1941.
[3]Cooper, 1970.
NT = Not tested. **WAIS–III** = Wechsler Adult Intelligence Scale, Third Edition, Spanish version. **WMS–III** = Wechsler Memory Scale, Third Edition, Spanish version.

PET Images

Results derived from the ROI analysis revealed significant decreases in the metabolic activity, or hypometabolism, of Patient 1's right OFC, anterior insula, caudate head, and thalamus, whereas increased metabolic activity, or hypermetabolism, was found in his medial insula and left thalamus (Figure 2).

TABLE 2. Proportion and Probability of Damage for Each Tract of Interest

	Proportion (%) of damage*				
	UF	**AC**	**aTR**	**IFOF**	**ILF**
Patient 1	36.7	29.6	3.7	22.4	52.8
Patient 2	38.0	27.5	0.2	4.6	29.9
	Probability of damage (%)†				
	UF	**AC**	**aTR**	**IFOF**	**ILF**
Patient 1	100	88	100	100	100
Patient 2	100	96	100	100	100

*Number of voxels of a given tract superimposed with the lesion mask divided by the total number of voxels of the tract.
†Probability of each tract to be affected by the lesion.
AC = anterior commissure. **aTR** = anterior thalamic radiations. **IFOF** = inferior fronto-occipital fasciculus. **ILF** = inferior longitudinal fasciculus. **UF** = uncinate fasciculus.

DISCUSSION

In these two patients who developed OCD in association with large malignant tumors involving the right temporal lobe, the most consistent OC behavior for both was NtK, manifested by a strong urge to access the accurate name for unique entities when they are not readily available. Inaccessible names were mainly those of famous people and acronyms. To a lesser extent, the names were of places laden with intense emotional valence (eg, the address where the mother of Patient 2 grew up). Patient 2 occasionally described other less disabling OC symptoms (eg, checking the door). Overall, our patients did not show the typical OC symptoms (ie, aggressive thoughts, fear of contamination, washing/cleaning rituals) that are usually described by patients with I-OCD (Mataix-Cols et al, 2004; van den Heuvel et al, 2009).

The NtK symptoms of our two patients were severe (lasting >5 hours per day), appeared on a daily basis, and interfered with their activities of daily living and social functioning. In general, NtK was triggered by both environmental and internally generated stimuli. In neither of the patients, however, was NtK associated with symptoms that are suggestive of epileptic activity. The NtK symptoms in Patient 1

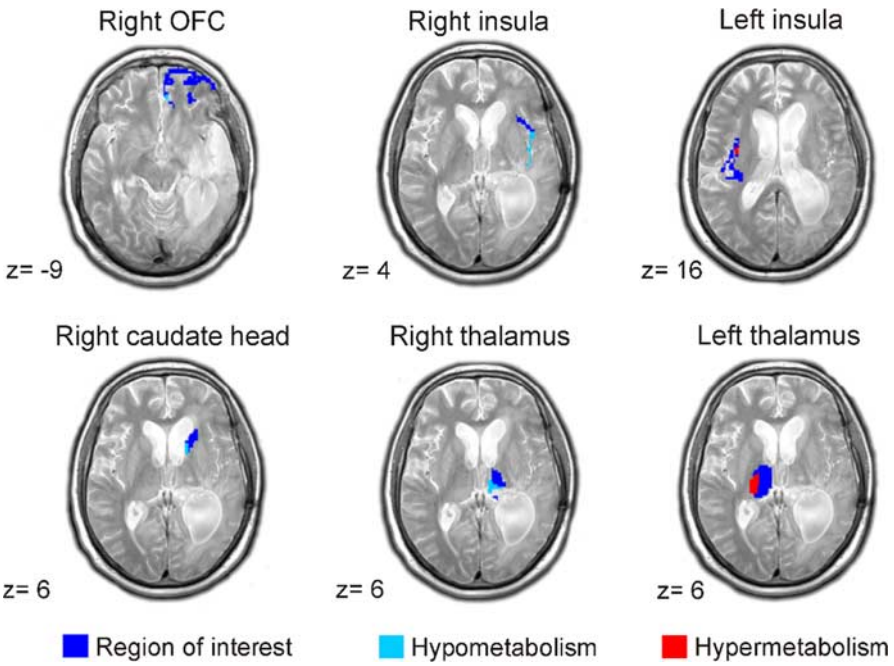

FIGURE 2. Altered metabolic activity in regions belonging to the fronto-striato-thalamo-cortical circuitry related to obsessive-compulsive disorder. Axial view of the ^{18}F-fluorodeoxiglucose PET region of interest analysis in Patient 1. The orbitofrontal cortex (OFC), anterior insula, caudate head, and thalamus in the right hemisphere show reduced metabolic activity, whereas the medial insula and left thalamus show increased metabolic activity in Patient 1 compared to 25 healthy controls. Results are shown at a voxel threshold of 0.05 (family-wise error corrected), adjusted for the small volume. (Figure 2 can be viewed in color online at www.cogbehavneurol.com.)

heralded the presence of a right temporal glioblastoma, disappeared after lobectomy, and recurred several months later. By contrast, the NtK symptoms in Patient 2 emerged only after his lobectomy.

Both patients were intermittently unable to retrieve specific knowledge (ie, the names of famous actors/actresses, singers, streets; meaning of acronyms) and persistently engaged in obtaining missed information through the media (internet). They also compulsively asked relatives and acquaintances for the information. It seems that the impaired ability to access specific information prevented both patients from obtaining the reward of completeness, eventually leading to heightened anxiety and searching compulsions (Huey et al, 2008). When someone provided the correct name, both patients obtained a temporary relief of anxiety until a new obsession arose. The likelihood that these symptoms were attempts to compensate faulty semantic retrieval may be taken into consideration. Nevertheless, both patients indifferently acknowledged having deficits in some cognitive tests tapping executive function and memory for faces only after being informed of such by the examiners. As shown in Table 1, both patients had average performances on the Boston Naming Test (Kaplan et al, 1983) and the Semantic Fluency subtest of the Consortium to Establish a Registry for Alzheimer's Disease battery (Morris et al, 1989), and Patient 1 had average scores on retrieving the names of objects/animals from the Pyramids and Palm Trees Test (Howard and Patterson, 1992).

Moreover, these two patients never saw their NtK as evidence of, or compensation for, cognitive failure, as has been described in other patients with A-OCD (Berthier et al, 2001; Jenike and Brandon, 1988; Salinas et al, 2009). This attitude, or failure to acknowledge the irrationality of their symptoms, was likewise observed during administration of the Yale-Brown Obsessive Compulsive Scale (Goodman et al, 1989), which revealed the presence of OCD with "poor insight" (American Psychiatric Association, 2013). Indeed, this scale revealed that both patients appeared unconcerned about their recurrent intrusive thoughts and searching compulsions despite the resulting high level of interference in functioning. In fact, the two patients did little to resist or control their OC behaviors.

A resting ^{18}F-fluorodeoxiglucose PET scan that was performed on Patient 1 several months after the right temporal lobectomy disclosed significant decrement of metabolic activity in structurally spared regions of the right hemisphere (ie, OFC, anterior insula, caudate head, and thalamus), which have previously been related to A-OCD (Berthier et al, 1996; Salinas et al, 2009). There also were increments of metabolic activity in the medial insula and left thalamus, which may be linked to OCD symptomatology (Spalletta et al, 2014). Importantly, all of these structures (except the insula) are integral components of the fronto-striato-thalamo-cortical circuitry that has been implicated in the pathogenesis of OCD (Huey et al, 2008; Pauls et al, 2014) (Figure 3A). The insula has reciprocal connections with fronto-subcortical circuits, particularly with the OFC, and the posterior insular cortex has connections with the temporal lobes (Song et al, 2011). Hence, it is probable that the insular cortex plays a critical role in the modulation of interoceptive

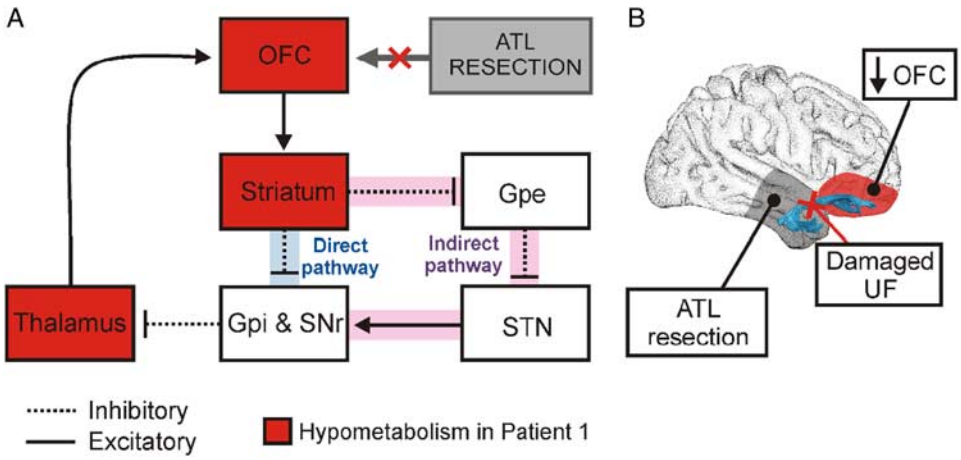

FIGURE 3. The fronto-striato-thalamo-cortical circuitry. **A:** Schematic representation of the fronto-striato-thalamo-cortical circuitry. Notice that the thalamus, the orbitofrontal cortex (OFC), and the striatum show reduced metabolic activity in Patient 1 (colored in red) as revealed by the PET analysis (Figure 2). **B:** Illustration of the anterior temporal lobe (ATL), the uncinate fasciculus (UF), and the OFC. In both patients, the ATL was damaged in the right hemisphere, affecting the inferior fronto-occipital fasciculus, inferior longitudinal fasciculus, and UF, with the consequent interruption of input to the OFC. The right anterior commissure was also damaged. **Gpe** = external globus pallidum. **Gpi** = internal globus pallidum. **SNr** = substantia nigra pars reticulata. **STN** = subthalamic nucleus. (Figure 3 can be viewed in color online at www.cogbehavneurol.com.)

processing among patients with OCD (Alvarez et al, 2015; Gonçalves et al, 2016; Song et al, 2011).

Both of our patients had structural damage with similar topography in the right anterior temporal lobe, which affected the activity of close and remote regions in both hemispheres. In fact, Tractotron and Disconnectome maps allowed us to track streamlines passing through the damaged regions, showing that the lesions affected several white matter tracts connecting the temporal lobe to brain regions that are crucial for the expression of OCD. The right temporal lobectomy affected the IFOF, ILF, AC, and UF. In Patient 1, the forceps minor, a tract recently implicated in the pathogenesis of OCD (He et al, 2018), was also affected. In both patients, damage to the UF may have deleteriously affected the activity of the ipsilateral OFC (see PET results in Figure 2), thus heightening an obsessive pursuit in knowing the name of unique entities. Different sectors of the right temporal lobe that were affected in our two patients, particularly a subtract of the UF linking the anterior temporal lobe with the OFC (Figure 3B), have been implicated in face-name learning (Metoki et al, 2017) and in the retrieval of person-related information (Damasio et al, 2004) and living things (Chan et al, 2004). The cognitive profile of both patients resembled, in part, the cognitive phenotype of I-OCD (especially the executive deficits) resulting from white matter microstructural alterations in temporal lobes (Spalletta et al, 2014) and long-distance tracts (IFOF, ILF, UF, and AC) connecting frontal and temporal cortices with posterior cortices (Garibotto et al, 2010).

Similarly, NtK behaviors probably followed a multiple functional disconnection, cutting the link between social conceptual (right superior anterior temporal lobe) information and social perceptual (right inferior temporal cortex) information (Pobric et al, 2016; Zahn et al, 2007, 2009) with proper names (left anterior temporal cortex via right AC involvement) (Damasio et al, 1996; Gorno-Tempini et al, 1998; Semenza, 2006). The involvement of the IFOF, ILF, AC, and UF, particularly the UF, played a critical role in disrupting connectivity with target cortical areas (OFC and anterior temporal lobe, insula) and other components of the fronto-striato-thalamo-cortical circuitry favoring the emergence of OCD. Moreover, decreased metabolic activity (Patient 1) and structural damage (Patient 2) in the right anterior insular cortex and OFC may explain both patients' poor insight into their NtK behavior (Fan et al, 2017).

In summary, data from the present study concur with previous research on A-OCD (Berthier et al, 1996) and current findings on I-OCD (Figee et al, 2013; Piras et al, 2013, 2015; Pujol et al, 2004; van den Heuvel et al, 2009), which suggest that the temporal lobes participate in the phenomenological expression of both OCD types. Findings from our two patients may help to disentangle the neurobiological underpinnings of a specific subtype of OCD that is acquired after brain damage. Further research on the clinical characteristics, neural correlates, and treatment of monosymptomatic forms of OCD is warranted.

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