INTRODUCTION

Normal pressure hydrocephalus (NPH) was first described by Adams and Hakim in 1965 (1). A rare disease of the central nervous system, it presents with ventricular enlargement, gait disturbance, dementia, and urinary incontinence secondary to cerebrospinal fluid flow disturbance without elevated intracranial pressure. Ventricular enlargement results in hypoperfusion of especially the frontal lobe, basal ganglia, and thalamus (2). NPH was divided into two terms of etiologic factors: 1) primary (idiopathic) NPH and 2) secondary NPH with underlying trauma, infection, or hemorrhage. A study conducted in Norway reported the incidence of NPH as 5.5/100000 and the prevalence as 22/100000 (3).

NPH may manifest with various neuropsychiatric findings. In a study of 64 idiopathic NPH patients, at least one neuropsychiatric finding was detected in 73.4% of the cases (4). The three most common
findings in that study were apathy (70.3%), anxiety (25.0%), and agitation (17.2%). In another study involving 35 cases, the frequency of psychiatric signs was 71% (5). The most common findings in this study were anxiety, depressive mood, and psychotic signs. Neither of these two studies reported findings of obsessive compulsive disorder (OCD).

Other information on the association between NPH and psychiatric findings is based on case reports. Personality changes, anxiety, depression, psychotic syndromes, delusional disorder, agitation, mania, kleptomania, and OCD were observed in case reports published so far (6-11).

We aimed to present a chronic NPH case who developed obsessive compulsive (OC) symptoms during disease course, with further approach to this patient. It is thought that this is the first case of OC symptoms associated with chronic NPH, published in our country.

**CASE**

The case was a 43-year-old male patient. He was married and had a child. Retired due to physical disability as a workman, he left secondary school at the past and lives in Aliaga. There are no previously known psychiatric complaints and admission. Approximately 7 years ago, he left consciousness after falling into a pit while walking; and was admitted to the emergency room with subdural and epidural hematoma, and further followed up in the intensive care unit. Urinary incontinence and ataxia developed and lasted so far. Six years ago, the patient developed generalized tonic-clonic (GTC) seizures, which were 5-6 times per week; and was reevaluated by cranial computed tomography (CT) and magnetic resonance imaging (MRI), where ventricular enlargement was detected. The results of lumber puncture (LP) performed at that time could not be achieved. The case was diagnosed with NPH. Electroencephalography (EEG) showed additional diagnosis of epilepsy, where the patient was prescribed phenytoin 300mg/day. However, upon failure to achieve a seizure control, the patient was interned to the neurology department of Atatürk Training and Research Hospital in Izmir Katip Celebi University, where phenytoin was discontinued and carbamazepine 800mg/day with levetiracetam 3000mg/day was started. Despite being indicated by neurosurgery department, ventriculoperitoneal shunt could not be performed as the patient and his relatives did not give consent. The patient benefited from anticonvulsant therapy and did not have an epileptic seizure within the next 5 years. His relatives stated that he had a 2-month depressive episode, featuring insomnia, anxiety, anhedonia, thoughts of worthlessness, and eventually attempt of suicide (jumping off) 5 years ago and soon after the discharge. They further stated that he did not seek psychiatric advice and the condition was resolved by family support.

Seven years ago, there was a gradual increase in the rate of decline in short-term memory starting after the accident. Three years ago, the condition deteriorated such that he started to confuse the name of his relatives and not be able find the way to home. Two years ago, irritability and attempts to harm his relatives were added, for which he did not visit any psychiatrist. The follow-up for epilepsy continued neurology department of Atatürk Training and Research Hospital in Izmir Katip Celebi University.

Cleaning compulsions of the case were first observed one year ago. He washed the clean glasses in the kitchen 3-4 times, wiping the benches and tables again and again. Even the smallest garbage in the house was disposed. A few months later, he developed a behavior of hand washing for 5 minutes each time about 20 times a day, and of taking shower lasting about 1 hour. He swore to himself that he did not believe in God, and then swore he believed it a few times.

The patient developed a seizure one month ago due to non-compliance to anticonvulsant treatment. Upon developing four episodes of the GTC seizures, he was hospitalized to Atatürk Training and Research Hospital of Izmir Katip Celebi University for adjusting anti-epileptic treatment and improving seizure control.

As the patient was observed to usually spend his time in the bathroom, wash his hands frequently and
want to take a bath, and become agitated whenever he notices a garbage in his room; a psychiatry consultation was requested.

The mental status of the patient was consistent with his age and socioeconomic status. He had diminished self-care and was conscious, with an indifferent time and complete place and person orientation. An irritable affection was observed and an irritable mood was identified. Reaction time was prolonged, and associations were relevant. No psychotic finding was observed. Some ruminative thoughts related with contamination and religion were determined: the environment at home and the hospital was so dirty that he might have caught infection if he had not washed his hands frequently; or he lost faith in God. Ruminative thoughts were considered as obsessions and compensatory behaviors as compulsions on the basis that the counselor stated that these symptoms caused significant distress in himself, affecting his daily life negatively, for which he wanted to get rid of them. There was no suicidal and homicidal thought. Spontaneous and voluntary attention was decreased. A decrease in the recent and remote memory was observed. His abstraction was impaired. He was not aware of the current events, he was able to do simple operations, and he knew partly of the money and statesmen. Psychophysiological function alterations included decreased libido, insomnia, and anhedonia. Increased psychomotor activity and cleaning compulsions were observed as expressed behaviors. Severe deterioration in social and occupational functioning was identified.

The patient’s mini mental test result was 20/30 (orientation: 7, recording memory: 1, attention and calculation: 4, recall: 1, language: 7). The Montreal Cognitive Assessment Scale was detected as 13/30 (visual spatial: 0, nomenclature: 2, attention: 3, language: 1, abstraction: 1, delayed recall: 2, orientation: 4). The Yale Brown Obsessive Compulsive Scale (Y-BOCS) was applied, upon which the patient got a total of 30 points: 15 points from each of obsession and compulsion. The patient was diagnosed with major neurocognitive disorder due to other general medical condition.

Routine complete blood count, biochemistry, urinalysis, cranial imaging, and EEG were performed by the neurology department during the hospital admission. All routine laboratory examinations were normal except for urinary tract infection. No lumbar puncture could be made as the patient failed to cooperate. Cranial CT showed supratentorial obstructive hydrocephalus. Cranial MRI reported advanced non-communicating hydrocephalic enlargement and an arrested hydrocephalus in supratentorial areas of ventricular system. Arrest form was reported as a hydrocephalus. The EEG showed diffuse basal rhythm disorder that was predominant in anterior regions of the hemispheres and paroxysmal epileptiform abnormality in anterior regions of the hemispheres.

The department of neurosurgery was consulted for the management of the underlying organic condition. The neurosurgery council discussed the case and concluded that hydrocephalus had become chronic with cortical atrophy, and that shunt could not be performed due to the risk of cortical bleeding and nerve damage. We re-evaluated the patient and suggested sertraline 50mg/day for OCD and quetiapine 50mg/day extended release for insomnia and irritability. During the stay in neurology department of Atatürk Training and Research Hospital of Izmir Katip Celebi University, irritability and insomnia was observed to regress. Furthermore, seizure control was achieved upon adjusting antiepileptic drug regimen to a previously used dose (carbamazepine 800mg/day, levetiracetam 3000mg/day). All these led to discharge of the patient to be further monitored in an outpatient setting by neurology and psychiatry departments.

Patient’s irritability and hostile behavior disappeared after one month’s follow-up. No side effects of any drugs and epileptic seizures were identified. His relatives stated continence of cleaning compulsions, but in a less severe form. Y-BOCS score was 23 points in total; 12 points from obsession and 11 points from compulsions. Y-BOCS symptom checklist indicated dirtiness and religious obsessions with cleaning compulsions and repetitive ritual
behaviors. It was observed that the patient did not give the expected response to the treatment. However, current treatment was decided to be maintained considering his epilepsy diagnosis and drug-drug interactions.

Significant improvement in the OC symptoms was detected in the second visit one month later. Y-BOCS score was 15 in total; 8 points from obsession and 7 points from compulsion. No agitation was observed, 75% reduction in irritability was found. Forgetfulness and episodes of being lost due to dementia continued. No epileptic seizure was identified. The patient was maintained on sertraline 50mg/day and quetiapine 50mg/day sustained release form.

The outpatient clinic follow-up was ongoing.

DISCUSSION

The first case of NPH and OCD co-occurrence was reported in 1994 by Abbruzzese et al. (10). A 20-year-old patient with dirtiness obsession and cleaning compulsion did not respond to either behavioral therapy and clomipramine 250mg/day or subsequent therapy with fluvoxamine 300mg/day. MRI showed idiopathic NPH, upon which a ventriculoperitoneal shunt was performed with a dramatic response with a complete alleviation of OCB findings (10).

Another case was reported by Kaufman et al. (11). In a 80-year-old female patient who applied with the classical triad, contamination obsessions and cleaning compulsions developed two months after the diagnosis of idiopathic NPH. There was no significant decrease in OCD symptoms of the patient four days after the shunt, and no information was given about the subsequent prognosis (11).

Sharma et al. (12) reported a 29-year-old male patient. Two years after the diagnosis of epilepsy, he presented with obsessions of being unsure and compulsions of controlling. NPH was found on MRI, for which he underwent ventriculoperitoneal shunt with thorough resolution of OCD symptoms (12).

Many studies have been carried out on underlying neurochemical and anatomical structures for the formation of OCD. Despite no consistent results, volumetric abnormalities were detected in basal ganglia in a group of cases (13,14). In single photon emission tomography (SPECT) studies, basal ganglia showed decreased perfusion, whereas bilateral orbitofrontal cortex, left parietal cortex, and bilateral parietal cortex had increased perfusion (15-17). Positron emission tomography (PET) studies reported conflicting findings; while some studies indicated an increase in metabolism especially in the orbitofrontal cortex and basal ganglia, a study showed decreased metabolism in all brain regions (18,19). In two trials comparing OCD and healthy controls, the ventricle:brain tissue ratio was reported as significantly higher in OCD patients (20,21).

Ventricular enlargement is a possible cause of neuropsychiatric findings in patients with NPH. The cerebral cortex, basal ganglia, and periventricular gray matter are the areas that affected by ventriculomegaly. Typically, cortical atrophy and decreased total brain volume is observed. Basal ganglia, which are thought to play a key role in OCD pathophysiology, are also inevitably affected. Furthermore, cerebral hypoperfusion was demonstrated in patients with NPH (22). The previously mentioned cases were acute cases and the authors attributed the sudden onset of OCD symptoms to the sudden hypoperfusion of the basal ganglia. Restoration of perfusion in basal ganglia after shunt operation might explain the dramatic improvement in the findings (11).

Unfortunately, we could not perform shunt operation because of chronicity of the disease and risks of intracranial hemorrhage. Unlike others, our case responded psychopharmacological treatment significantly. Usually, high-dose serotonergic antidepressants are needed for treatment in classical OCD patients. However, it is pleasing that the patient benefited from the use of low-dose antidepressants, avoiding from lowering of the epilepsy threshold and drug-drug interactions. The patient responded to the sustained release quetiapine treatment, which was initiated for insomnia and behavioral problems. Quetiapine was selected because of its low drug-drug interaction potential and its relatively better tolerability in epilepsy patients (23). Quetiapine was administered with a low-dose regimen since the side effects,
particularly of the extrapyramidal system, are observed more frequently in NPH, as in other organic pathologies (24). While its effect on the control of the emotional problems and impulse control problems is inarguable, it is thought that low dose use alone cannot explain the improvement in OC symptoms.

Patients with moderate to advanced stages of dementia may exhibit stereotypic behaviors, intrusive-ruminative thoughts, and OC symptoms (25,26). In particular, OC complaints and collecting behavior may be seen in frontotemporal dementia (27). Non-specific effects of neurodegenerative process and frontal hypoperfusion is thought to be responsible for the development of OC symptoms (28). In our case, dementia developed secondary to NPH. The ego dystonic character of his ruminative behaviors and intrinsic thoughts is important for evaluating these complaints as OC symptoms and for distinguishing them from stereotypic or impulsive behaviors. OC findings might have developed secondary to chronic cerebral hypoperfusion and atrophy. Therapeutic options for behavioral disorders in dementia include selective serotonin reuptake inhibitors and low-dose antipsychotics (26). In addition to that for obsessions, sertraline and low doses of quetiapine may be effective in the treatment of irritability and impulse control problems.

Up to 10-22% of patients with epilepsy have concurrent OCD diagnosis (29). In epileptic patients, stereotypic or compulsive behaviors may occur in the inter-ictal period (29). A study by Ertekin et al. in our country reported that the most common symptoms of OCD in epileptic patients were contamination and order/symmetry (30). Involuntary intrusive thought and compensatory behaviors in our case are considered as OC symptoms as they elicited marked distress and had ego dystonic character. The importance of development of frontal lobe dysfunction in OCD is well recognized, therefore the detection of epileptic discharges in bilateral anterior regions of the hemispheres might play a role in the development of OC symptoms (29). The therapeutic effect of anticonvulsants on OC symptoms has not been proven, but some case reports suggested potentially positive effects (31). In any case, main goal for the control of pre-ictal and inter-ictal symptoms in concurrent epilepsy and psychiatric disorder is establishing an effective anti-epileptic treatment (32). In fact, establishment of the seizure control in our case may be considered to play an important role in the treatment of OC symptoms.

Another factor that should be considered in the differential diagnosis is the drug-induced OC symptoms. There is only one case report on the association of levetiracetam and OCD; the case was reported to have a generalized developmental disorder; and the OCD signs developed shortly after drug initiation (33). In our case, OCD findings did not develop despite the long-term use of levetiracetam and subsequent re-initiation. Carbamazepine-associated OCD was not reported, rather being reported to be useful in treatment-resistant OCD cases (34,35). These suggest that the drugs used by the patient are unlikely to cause OCD symptoms.

In our case, OC symptoms might have developed secondary to uncontrolled epileptic seizures and neurocognitive disorder characterized by cerebral hypoperfusion and diffuse cortical atrophy due to chronic NPH. A multidisciplinary approach was needed because of the chronic nature of the clinical condition with co-occurrence of multiple neurologic pathologies. We recommend that all areas involved in the pathophysiology be addressed and that a multidisciplinary management approach be taken for similar cases. It is evident that further studies are warranted regarding the association of NPH to OC and other neuropsychiatric findings, also addressing its therapeutic options.

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Informed Consent: Written consent was obtained from the patient.

Peer-review: Externally peer-reviewed.

REFERENCES


5. Oliveira MF, Oliveira JR, Rotta JM, Pinto FC. Psychiatric symptoms are present in most of the patients with idiopathic normal pressure hydrocephalus. Arq Neuropsiquiatr 2014; 72:435-438. [CrossRef]


