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Case Report

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Ogilvie's syndrome presented with delirium in an older lady with corticobasal syndrome

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Abstract

Objective. Corticobasal syndrome (CBS) is one of an atypical parkinsonian syndromes characterized by extrapyramidal features as well as cortical involvement signs. A variety of factors may lead to delirium in older adults with chronic progressive life-limiting neurological illnesses like CBS. Ogilvie's syndrome (OS) is an acute colonic pseudo-obstruction in which abdominal distension, nausea, vomiting, and constipation can be seen. We report a case of OS identified as the underlying possible cause of delirium in an 80-year-old woman with CBS. We also discuss the importance of holistic approach which is essential to manage the underlying cause and to preserve the quality of life in particular for the frail geriatric population who potentially needs palliative care or already benefits from palliative care. Method. An older patient with CBS presented with symptoms similar to that of acute colonic

obstruction and subsequently developed delirium. The patient was found to have colonic pseudo-obstruction (OS).

Result. Neostigmin infusion was therefore given to treat it and delirium was resolved. Significance of results. To the best of our knowledge, clinical manifestation of delirium as OS in a patient with CBS has not been previously reported. OS may be superimposed to CBS in older patients, and OS in such patients may play a role as a precipitating factor for the development of delirium. Given the fact that CBS is progressive and rare neurodegenerative disease and almost all of these patients need palliative care, eventually, health-care professionals, especially in palliative care, should be aware of distinctive challenges of life-limiting chronic neurological illnesses, such as conditions that may lead to the development of acute colonic pseudo-obstruction because the rapid treatment of them prevents the use of potentially harmful drugs, surgical procedures, or inappropriate interventions.

Introduction

Acute colonic pseudo-obstruction (Ogilvie's syndrome, OS) is a disease characterized by acute enlargement of the colon in the absence of a mechanical cause preventing the flow of small or large intestinal contents (Vanek and Al-Salti, 1986). The development of acute colonic pseudo-obstruction has no definite causes; however, many clinical conditions which are common in older adults (Conner et al., 2020) and at increased risk for it, such as advanced age, comorbidities, electrolyte imbalance, polypharmacy, and immobility, have been identified (Ates Bulut et al., 2018). The main clinical feature is abdominal distension which is reported in 80% of the patients. Additionally, nausea-vomiting and constipation are also reported in 60% and 50% of the patients, respectively (Vanek and Al-Salti, 1986; Jetmore et al., 1992; Ates Bulut et al., 2018; Conner et al., 2020).

On the other hand, corticobasal syndrome (CBS) is a chronic progressive neurodegenerative disorder characterized by the involvement of various combinations of basal ganglia dysfunction signs such as akinesia, asymmetric rigidity, dystonia, and as well as cortical ones such as apraxia, myoclonus, alien-limb phenomena, and cognitive impairment. Constipation, one of the signs of autonomic failure, may also be observed in patients with atypical parkinsonian syndromes (Deutschländer et al., 2018), and constipation causing fecal impaction and accompanying a variety of conditions may cause delirium in older adults with chronic progressive neurological diseases. These diseases like CBS are one of the most common predisposing conditions for delirium. Besides, now, CBS and other atypical parkinsonism syndromes have been reported not only to carry a heavy symptom burden equivalent to that of patients with advanced cancer but also have a higher burden of multimorbidity and risk of becoming wheelchair-dependent or bedridden. Additionally patients with CBS need palliative or hospice care, eventually (Bükki et al., 2016; Wiblin et al., 2017) and intestinal motility problems, such as constipation, colonic pseudo-obstruction, as well as delirum, may be common in these patients receiving palliative care and can cause extreme suffering and discomfort.

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Fig. 1. Plain abdominal radiographs showed an enlargement of the colon loops and air-fluid levels (before treatment).

Therefore, identifying and managing the possible underlying cause is crucial for patient outcomes, particularly, in those who potentially need palliative care or already have benefited from a palliative care service. To the best of our knowledge, clinical manifestation of delirium presented as OS in a patient with CBS has not been previously reported.

Case

An 80-year-old female patient was admitted to the emergency room with constipation for the last five days and change in consciousness for the last two days. She has been treated with levodopa-benserazide (625 mg/day), amantadine (150 mg/day), and levothyroxine (50 $\mu g/day$) due to CBS and hypothyroidism for 3 years in our unit. There was no history of new surgery or medication.

In the physical examination performed in the emergency department, her blood pressure was 110/70 mmHg, pulse rate was 112 per minute; and altered mental status, disorientation, fluctuations in consciousness, distraction, asymmetric limb, and axial rigidity, abdominal distention were determined and there were no bowel sounds. The blood test results performed at the hospital admission were as follows: white blood cell count 7000/L, hemoglobin 12.1 g/dL, glucose 116 mg/dL, urea 24 mg/dL, creatinine 0.64 mg/dL, sodium 142 mM/L, potassium 3.8 mM/L, total calcium 8.9 mg/dL, magnesium 0.87 mg/dL, serum TSH 1.39 m(IU)/L, free T4 1.19 ng/dL, all of them were within normal limits. Plain abdominal radiographs showed enlargement of the colon loops and air-fluid levels (Figure 1). After the mechanical cause of obstruction was ruled out, she was transferred to our geriatric clinic because of delirium and OS superimposed to CBD.

In our clinic, conservative therapy was initiated with fluids, lactulose, and enema. In the clinical observation of the case, despite the conservative and supportive treatment, colonic discharge was not provided and delirium continued. However, she has required no additional treatment due to delirium. During the follow-up, she was not able to defecate despite repeated enema application. Therefore, 48 h after the hospitalization, neostigmine (1 mg/30 min) infusion was started and repeated two times. After the second dose of neostigmine administration, we observed an increase in intestinal movement and then stool discharge was started. After stool discharge, an improvement was observed in the patient's delirium clinic. Abdominal distension

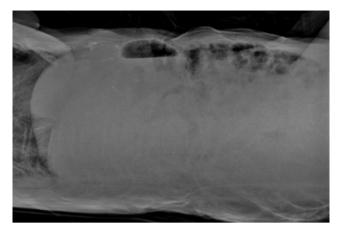


Fig. 2. Decrease in the width of the colon loops and loss of air-fluid levels on abdominal radiography (after treatment).

completely regressed on the 5th day of hospitalization (Figure 2). The patient was discharged with fiber and lactulose treatment.

Discussion

Herein, we report a case of OS presented with delirium in an older lady with CBD. In this case, we consider that CBD is a predisposing factor in delirium development, and OS is a precipitating factor. The treatment of the etiological causes of delirium is very important because it allows the rapid treatment of the cases by preventing the use of potentially harmful drugs or inappropriate interventions.

OS is common in patients over the age of 60 years (Vanek and Al-Salti, 1986), and the pathophysiology is still unknown. It may be caused by an increased sympathetic intestinal innervation or a decreased parasympathetic tone, or both, which results in functional obstruction of the colon (Jain and Vargas, 2012). On the other hand, CBD is a rare neurodegenerative disease, in which the signs of autonomic failure can be seen like other parkinsonian disorders. For this reason, in patients with CBD, it should be kept in mind that accompanying comorbid conditions, related drugs, and age-related changes may lead to the development of OS as well as autonomic dysfunction such as deterioration in bowel and bladder control (Isik et al., 2013). Although OS was firstly described in 1948 (Ogilvie, 1948), this is the first CBD case that presented with OS, as far as we are concerned. Pharmacological treatment of OS aims to counteract intestinal sympatheticparasympathetic dysregulation. Neostigmine therapy turned out to be effective for acute colonic pseudo-obstruction that does not respond to conservative treatment (Turégano-Fuentes et al., 1997; Ponec et al., 1999). Therefore, in patients with OS who do not benefit from the supportive and symptomatic treatment despite potential side effects of neostigmine, it is very important to apply it by taking the necessary precautions for possible complications of the drug. We suggest an initial conservative treatment and drug therapy approach after excluding the causes of mechanical obstruction in patients with a diagnosis of CBD.

Health-care professionals including palliative care physicians should be skilled in a holistic approach addressing many distinctive challenges of life-limiting chronic neurological illnesses in these frail older adults, such that OS may cause acute colonic pseudo-obstruction and should be reviewed from this point of view before the surgical procedure.

Finally, OS may be superimposed to CBD in older patients, and OS in such patients may play a role as a precipitating factor for the development of delirium. Before surgical management, supportive care is crucial for the management of OS in CBD patients and neostigmine treatment should not be delayed in patients who do not benefit from supportive treatment. In addition, the elimination of delirium with the removal of colonic pseudo-obstruction in our case is an indicator of the importance of the etiological approach in delirium treatment.

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Conflicts of interest. The authors of this manuscript declare that there is no conflict of interest.

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