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Case of the patient with dyskinesia and hyperpyrexia syndrome

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Introduction

Dyskinesia-hyperpyrexia syndrome (DHS) is characterized by prominent dyskinesia and fever, often complicated by rhabdomyolysis.
This syndrome typically occurs in patients with long-standing PD, particularly who have recently undergone an increase or change in their dopaminergic therapy.

Case

The patient is a 66-year-old male with a medical history of a 2015 diagnosis of PD. His dosage of levodopa/benserazide had been increased one month prior to admission (Table1).

He presented to the Emergency Department with right upper quadrant pain and fever, leading to a diagnosis of acute cholecystitis. He was admitted to the ICU, and he underwent Percutaneous Transhepatic Gallbladder Drainage, Endoscopic Sphincterotomy with stone removal and antibiotic therapy with Piperacillin/Tazobactam.

Post-procedure, the patient remained NPO for three days, during which his PD medications were temporarily discontinued. Upon resuming the medications, he developed newly onset tongue dyskinesia. He was transferred to the Department of Rehabilitation Medicine for proper management of dyskinesia and associated dysphagia. at HD(Hospital Day) 20, CRP decreased from 24.8 to 1.0 mg/dL, WBC from 17,000 to 4,900/μL and follow-up CT showed no signs of infection. However, high fever persisted up to 39.2°C. Laboratory tests confirmed an elevated Creatine Kinase (CK) of 374 U/L at HD 24 (Chart1). It was unclear whether the dyskinesia was due to a lack or an excess of dopaminergic drugs. However, considering that the dyskinesia occured a week after hospitalization and persisted for three weeks despite strict medicine adherence, we suspected DHS. Management included returning dopaminergic drugs to their previous maintenance doses and initiating symptomatic treatment with Baclofen 20mg, and Clonazepam 0.5mg per day (Table1).

by HD 38, the CK level normalized to 101 U/L and the dyskinesia improved. However, around HD 46, the fever recurred and CK rose again to 284 U/L. In response, Baclofen was increased to 40mg, and the Clonazepam was increased to 1.5mg per day. Furthermore, the morning dose of levodopa/benserazide was additionally reduced (Table1).

The fever finally subsided and the patient was transferred to General Surgery for a cholecystectomy and was successfully discharged.

	HD 3	HD 24	HD 48
levodopa	1000	700	600
benserazide	125	25	0
Amantadine		200	200
baclofen		20	40
clonazepam		0.5	1.5

Table1. Changes in medication types and dosages (mg/day)

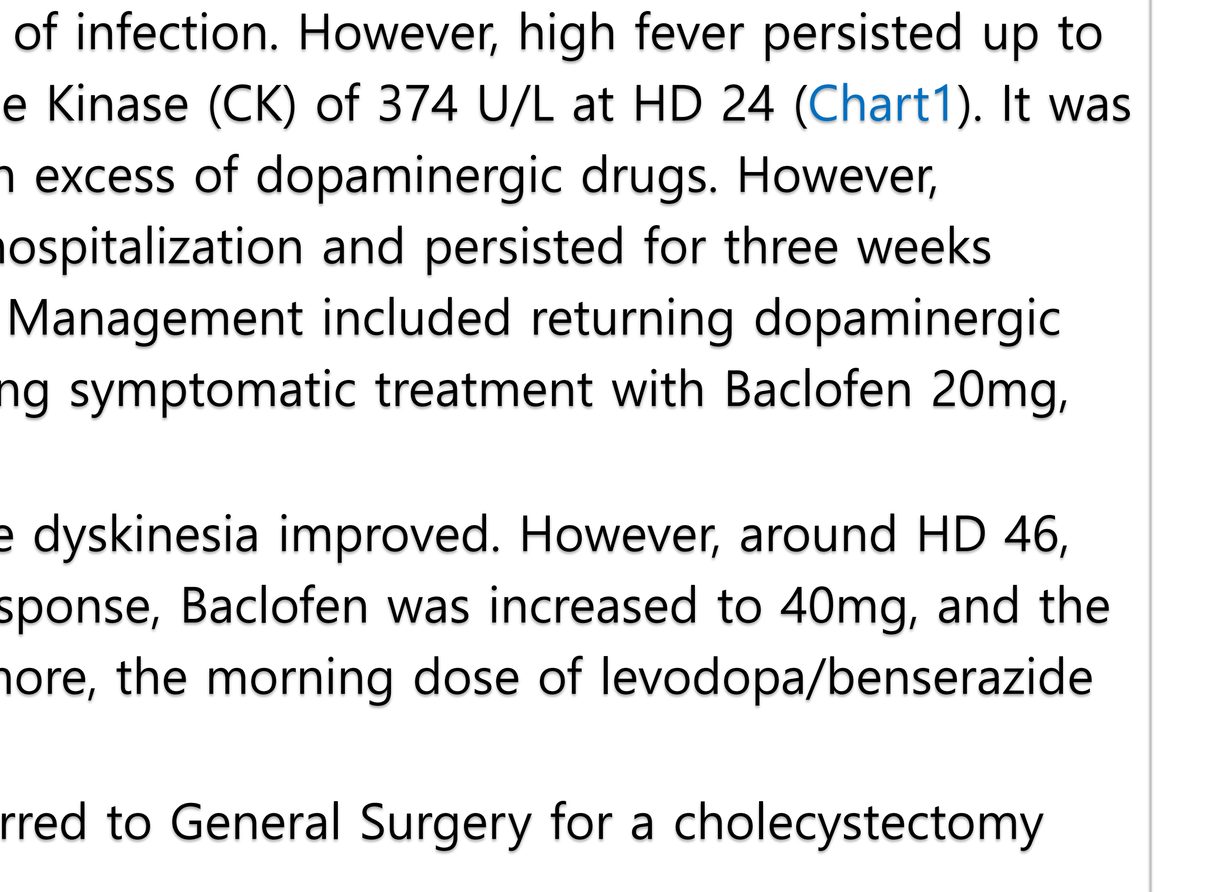

Chart1. Trends of CK (IU/L), and Body Temperature (°C) by length of hospital stay.

Conclusion

If dyskinesia is accompanied by fever and elevated CK levels, clinicians must consider DHS. in DHS, dopaminergic overstimulation causes continuous dyskinesia, increasing CK levels through sustained muscle contraction. This intense activity, coupled with hypothalamic thermoregulatory failure, results in hyperpyrexia. A significant clinical challenge arises because Parkinsonism-Hyperpyrexia Syndrome (PHS)—triggered by the sudden withdrawal of levodopa—presents with nearly identical clinical features. Therefore, precise medical history is essential. the clinician must carefully decide whether to gradually taper the dosage (in the case of DHS) or reintroduce the medication (in the case of PHS).