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Case Report

A Case of Trientine Overdose

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ABSTRACT

Wilson disease is a rare genetic hepatic and neurological disorder of copper accumulation. Trientine is usually used as a second line in the management of patients with this condition. We present a case of a large overdose of Trientine (60 g) resulting in self-limiting dizziness and vomiting with no further clinical sequelae or significant biochemical abnormalities. This case shows that Trientine has a good safety profile and hence could be used as a first line treatment in patients with Wilson's disease who suffer from psychiatric complications and who might be at risk of self-harm.

Key words: Overdose, trientine, Wilson's disease

INTRODUCTION

Wilson disease is a rare genetic hepatic and neurological condition, the management of which can be very challenging due to the limited treatments available. Trientine is a copper chelator that is used in the treatment of Wilson's disease.[1,2] In this case report we discuss the largest overdose of Trientine recorded.

CASE REPORT

A 41-year-old gentleman well known to our gastroenterology unit with Wilson's disease presented to the emergency department following an overdose of Trientine. He was known to have liver cirrhosis with evidence of portal hypertension and neurological involvement that mainly manifested as head tremor. He was on Trientine 300 mg three times a day as he previously developed a reaction to Penicillamine. He had no past history of mental illnesses and had no para-suicidal episodes prior to this presentation.

He presented to hospital after taking 200 tablets of 300 mg Trientine (total of 60 g), with suicidal intentions. However, some of the tablets were claimed to be 2 years out of date. He did not take any other medications and there was no concomitant alcohol intake. The incident was witnessed by his partner. At presentation the patient was complaining of dizziness but there were no focal neurological deficits. The rest of his physical examination was normal. On the following day, the patient started to experience vomiting and nausea. He had no further symptoms or physical complications and his dizziness and vomiting settled within less than 48 hours. His blood results showed a normal full blood count except for known mild chronic thrombocytopenia which was secondary to his liver disease. His renal function showed mild acute kidney injury,

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Creatinine 117 mmol/L (normal 62–106) which responded rapidly to intravenous fluids. The National Poisons Information Service (NPIS) indicated that Trientine had been found to be safe at a dose of 30 g; however, it was suggested that iron, zinc, renal functions and other electrolytes needed to be closely monitored. His calcium, magnesium and iron levels were normal. Nonetheless, he developed mild hypophosphatemia (0.69 mmol/L, normal 0.81–1.45) and low serum Zinc (7 mmol/L, normal 11–24 mmol/L). The latter improved spontaneously. The serum copper was low 6–7 mmol/L as expected at two separate readings (normal 11–20) and the urinary copper output was elevated 14.78 mmol/dL (normal less than 0.95). The patient remained medically fit and was discharged on the fourth day after being deemed at low risk of further suicidal attempt by the psychiatric team.

DISCUSSION

In medical practice, Trientine dihydrochloride is used almost exclusively in the management of Wilson's disease. It works primarily as a cupriuretic by promoting urinary copper excretion from the kidneys.[1] Trientine also reduces the intestinal absorption of copper. Trientine is used as a second line in patients with Wilson's disease and commonly in those who are intolerant to the first line medication, D-penicillamine.[2] Some anecdotal reports suggested it could be used as a first line treatment in view of its non-inferiority to penicillamine and the low side effect profile.[3] The adverse side effects of Trientine are rare but include reversible sideroblastic anaemia, lupus like reaction and worsening of neurological manifestations.[3]

To our knowledge this is the largest single overdose of Trientine reported to date. It is unclear how the patient gained access to that large number of tablets but this could be explained by possible poor compliance over the years and tablet hoarding. Our case shows that Trientine in large doses is reasonably safe and may result in only minor and temporary symptoms such as dizziness and vomiting notwithstanding the doubt that remains if some of the tablets were out of date. Biochemical disturbance was not demonstrated except for mild renal impairment and a slight reduction in the phosphate and zinc levels. It is known that Trientine is capable of binding and reducing the serum levels of both Fe and Zinc.[1] The serum iron concentration was not affected in this case and despite the transient decrease in Zinc concentration, there were no serious clinical consequences.

CONCLUSION

As Trientine appears to be safe in massive doses, assuming full biochemical activity in this case, and in view of the increased incidence of mental health problems in patients with Wilson's disease,[4] it may be reasonable to recommend its use as a first line treatment in Wilson's disease patients who have significant psychiatric complications with ongoing risk of self-harm. An early switch from D-penicillamine to Trientine in this subgroup may also be suggested.

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Conflicts of interest

There are no conflicts of interest.

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