Paraneoplastic chorea associated with gallbladder cancer

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DESCRIPTION
An 81-year-old woman presented with a 1-week history of involuntary movement of her left extremities. She also reported weight loss in the last few months. Her left upper and lower extremities were in a constant state of violent ‘dance-like’ movements, regardless of her intent to move them. Involuntary movement of some facial muscles were also observed (video 1), affecting her daily activities. Other ancillary neurological signs, including neuropathy, encephalopathy, dementia, psychiatric disorder and visual defects, were absent. She had no family history of Huntington’s chorea. Brain contrast-enhanced MRI revealed no obvious abnormal findings (figure 1A,B). Laboratory tests, including blood glucose and autoantibody assays (antinuclear antibody, antiphospholipid antibody, antineutrophil cytoplasmic antibody and rheumatoid factor), were unremarkable, except the marked elevation of CA19-9 level. Contrast-enhanced abdominal CT showed a gallbladder tumour with multiple liver metastases (figure 2). Chest CT showed no lung metastases. Examination of histopathological specimens based on needle biopsy confirmed hepatoid adenocarcinoma. Serum paraneoplastic antibodies (anti-CV2/collapsin response mediated protein (CRMP)5, Hu, Yo, Ri and amphiphysin) were negative. Antibodies against neuronal cell surface antigens or synaptic proteins, for example, N-methyl-D-aspartate receptors (NMDAR) were not measured. Cerebrospinal fluid analysis was not performed because her movement was restricted due to hemichorea; hence, it was difficult to maintain the stationary state for lumbar puncture. Nevertheless, the symptoms satisfied the diagnostic criteria for paraneoplastic neurological syndrome (PNS). Therefore, we diagnosed hemichorea as possible PNS. When administration of haloperidol for hemichorea commenced, the symptoms improved but did not completely disappear. However, after 2 months of gemcitabine–cisplatin chemotherapy for gallbladder cancer, the tumour shrank and symptoms disappeared completely. Therefore, haloperidol dose was tapered off, and her symptoms did not recur thereafter. Since her symptoms resolved after cancer treatment, we eventually diagnosed hemichorea as definite PNS.

Paraneoplastic chorea is an extremely rare PNS. It generally occurs subacutely and severely, progresses rapidly, and is drug-resistant. The symptoms are often asymmetrical or unilateral. The most common underlying cancer type is small-cell lung cancer. However, lymphomas and bowel or kidney cancer have been reported to also cause paraneoplastic chorea. To the best of our knowledge,
Images in...

Learning points

- It is important to probe for malignancy in patients with adult-onset chorea, particularly in elderly patients presenting with weight loss and asymmetrical symptoms.
- The fundamental principle in the treatment for paraneoplastic chorea is to treat the underlying cancer.
- For diagnosis, other known causes of chorea should be ruled out, especially cerebrovascular disease, infection (HIV), systemic lupus erythematosus, metabolic disease and drugs.

paraneoplastic chorea secondary to gallbladder cancer, especially related to hepatoid adenocarcinoma, has not been reported. In the diagnosis of PNS, other causes of chorea should be ruled out. Other potential causes include cerebrovascular disease, hyperglycemia, infection (HIV), systemic lupus erythematosus, metabolic disease and drug use (lamotrigine, methadone and lithium). Paraneoplastic antibodies are often positive in patients with paraneoplastic chorea. Anti-CV2/CRMP5 antibody has the highest frequency of positivity, followed by anti-Hu antibody. However, as in this case, there are patients without positive results for any antibody. Nevertheless, the absence of paraneoplastic antibodies does not rule out the possibility of the paraneoplastic origin of the disease. Immunological treatment (steroids and intravenous immunoglobulin), haloperidol and clonazepam have been reported to be effective in paraneoplastic chorea; however, the fundamental principle in the treatment for PNS, including paraneoplastic chorea, is to treat the underlying cancer (tumour resection and chemotherapy). Our report demonstrates the importance of probing for malignancies in patients with adult-onset chorea, particularly in elderly patients presenting with weight loss and asymmetrical symptoms.

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Case reports provide a valuable learning resource for the scientific community and can indicate areas of interest for future research. They should not be used in isolation to guide treatment choices or public health policy.

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