CASE REPORT

Case study of a migraine patient with multiple extracephalic symptoms including melena due to telangiectasia of the sigmoid colon : a possible new migraine-associated symptom

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Abstract : Migraine is a chronic neurological disorder characterized by headaches and extracephalic symptoms. We report a 73-year-old male patient with a history of migraines as well as several other chronic conditions including abdominal pain accompanied by nausea and vomiting, pain and ecchymosis of the limbs, dysmetropsia, syncope, and melena due to telangiectasia of the sigmoid colon. After a thorough evaluation of the migraine condition, we hypothesized that the patient's melena due to telangiectasia of the sigmoid colon might in fact be a migraine-related phenomenon. In this report, we discuss a possible mechanism for melena due to telangiectasia in migraine patients, as well as "tips" for identifying subtle and/or unreported clinical features of migraine conditions. J. Med. Invest. 70 : 298-300, February, 2023

Keywords **:** *migraine, extracephalic symptoms, telangiectasia*

INTRODUCTION

Migraine is a chronic neurological disorder characterized by headaches (1) as well as extracephalic symptoms, some of which, including cyclic vomiting syndrome (2) and abdominal migraines (3), are known as migraine-equivalents. In this report, we present a case of migraine co-present with various symptoms including abdominal pain accompanied by nausea and vomiting, pain and ecchymosis of the limbs, dysmetropsia, syncope, and melena due to telangiectasia of the sigmoid colon. After a thorough evaluation, we reached a hypothesis that could explain all of these symptoms through migraine-associated mechanisms. In order to minimize the risk of underdiagnosis, we emphasize the importance, in such cases, of evaluating many different aspects of migraine, all of which may not be obvious even to skilled physicians.

CASE REPORT

A 73-year-old male patient presented to our outpatient clinic with episodic headaches and various recurrent symptoms including abdominal pain with nausea and vomiting, pain in the limbs accompanied by ecchymosis, dysmetropsia, syncope, and melena due to telangiectasia of the sigmoid colon. The clinical course of the patient is shown in Figure 1. The patient's family history showed a consistent presence of migraines in the maternal family line. Beginning at age six, the patient began to experience severe periumbilical pain lasting approximately six hours, accompanied by nausea and vomiting. The patient also experienced episodic pain and ecchymosis of limbs without any obvious traumatic triggers. These symptoms always remitted spontaneously. The patient received supportive care because a medical investigation conducted between the episodes found no apparent abnormalities. Beginning at age 12, the patient began experiencing odd new episodes that lasted approximately one hour each. Regarding these episodes, the patient recounted : "I abruptly felt as if I became a 10-foot-tall giant." The patient also suffered several episodes of loss of consciousness triggered by mental stress, which were diagnosed as neutrally-mediated syncope. The patient was evaluated using electroencephalography several times, all of which showed no apparent abnormalities. Beginning at age 17, the patient started experiencing episodes of melena, which were treated with supportive care only. The patient then underwent a resection of the sigmoid colon due to a massive melena at age 23. Based on histopathological evaluation of this resected section, the patient was diagnosed with telangiectasia as a root cause of melena. At age 23, the patient began experiencing severe, throbbing unilateral headaches typically lasting around six hours without aura, although the headache was transiently accompanied by antecedent visual auras lasting approximately one hour from the age of 39 to 60. These headaches were also accompanied by photophobia. These episodes initially occurred weekly, although their frequencies gradually decreased to monthly after age 60 without any preventive medications. Findings of the patient's physical and neurological examinations between attacks were unremarkable. No abnormality was indicated on brain MRI. According to the International Classification of Headache Disorders 3 (ICHD-3), these types of headaches were compatible either with 1.1 : Migraine without Aura or 1.2 : Migraine with Aura. We hypothesized that all the above-mentioned symptoms could be explained by migraine-associated mechanisms, as detailed below.

DISCUSSION

This report describes a variety of migraine-associated extracephalic symptoms, most of which could easily be overlooked in many clinical settings. We believe that this report will facilitate

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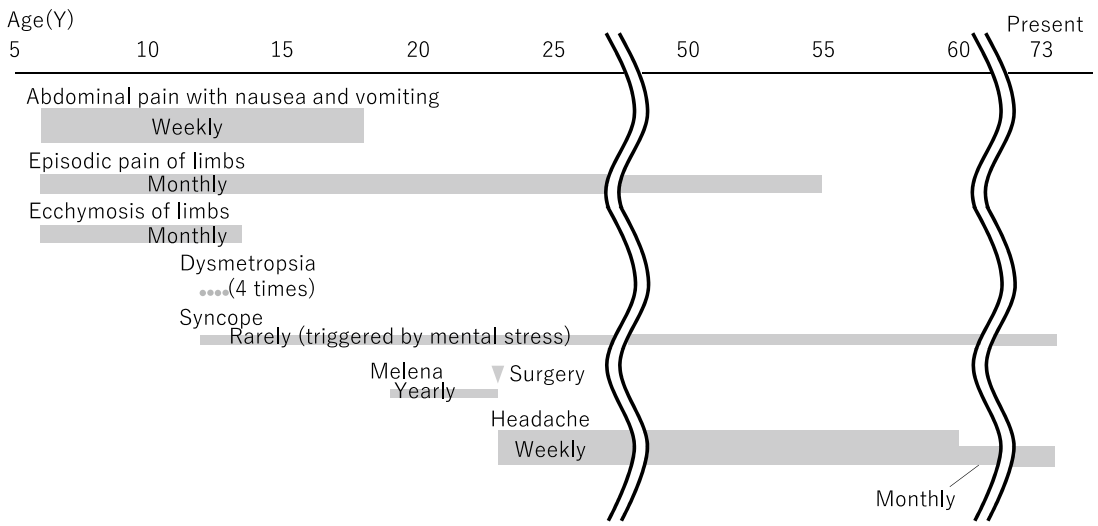

Figure 1.

the identification of a variety of migraine-associated features. This may contribute to better management of patients with migraine in the future.

We believe that this patient's symptoms can all be considered migraine-associated. His abdominal pain can be considered a form of abdominal migraine (3). An abdominal migraine attack can appear without an accompanying migraine headache (4, 5). Similarly, abdominal pain due to the mechanism of primary stabbing headache (a form of primary headache, which is reported to somewhat relate with migraine) can appear without stabbing cephalic pain (6). His body pain and accompanying subcutaneous bleeding could be symptoms of migrainous corpalgia (7, 8) and migraine-associated ecchymosis (4, 9), respectively. Dysmetropsia or "Alice in Wonder Syndrome" has also been reported to be associated with migraines (10). We believe that recognition of these peculiar forms of migraine is important to prevent diagnostic delay.

We believe that the patient's syncope can have some mechanism in common with migraine, as reported by Daas *et al*., who concluded that vasovagal syncope, a form of neurally mediated syncope, is associated with migraine (11). They reviewed three generations of a family with comorbid vasovagal syncope and migraine. It included 21 subjects. Eleven of the 14 subjects with a diagnosis of migraine (78%) also had vasovagal syncope, and 11 of the 12 subjects with vasovagal syncope (92%) also had migraine (11).

Moreover, our patient's intestinal hemorrhage due to telangiectasia of the sigmoid colon could also be explained by a migraine-related mechanism, similar to that of another case previously reported by Christensen *et al*. (12). In their meta-analysis, they found that the prevalence of migraines in patients with rosacea, a form of telangiectasia, was higher than in those without rosacea, although the authors could not find a detailed mechanism underlying the two conditions (12). More similar case-studies are needed in order to confirm our hypothesis, as, to the best of our knowledge, no other cases of intestinal hemorrhaging due to telangiectasia in migraine patients have been reported. This may be a novel aspect to consider in the setting of migraine patient care. However, we do not insist that all migraine patients with abdominal pain should receive colonoscopy. We understand that history focused on migraine will be important, especially when the migraine patient's abdominal pain is 1) repetitive, 2) severe, 3) localized to the midline (or periumblical region), 4) remitting within 2–72 h without specific intervention, and 5) each episode shows similar profile (stereotypic) (1).

The mechanism of occurrence of abdominal migraine may be speculated from that of migraine. Migraine is a condition with neuronal sensitization represented by allodynia (pain due to a stimulus that does not usually provoke pain) and hyperalgesia (increased pain from a stimulus that usually provokes pain) (13, 14). With an aim to clarify the mechanism of migraine-associated abdominal pain under this context, it may be reasonable to say that non-painful stimulus for non-hypersensitive brain (i.e., physiological intestinal movements) can cause prominent abdominal pain in the sensitized condition.

In future cases of this nature, we recommend a more "active" approach to history-taking (15). In other words, even if a patient does not report symptoms that may be related to migraine, the physician should still attempt to ascertain the presence or absence of candidate symptoms. This can occur even in situations where symptoms appear to be "distant" from the headache (e.g., abdominal pain in migraine patients). If physicians begin to adopt a more wholistic view of migraine treatment, in which any symptom may potentially be a feature of the patient's migraine, and attempt to develop unitary hypotheses for patients' conditions, we believe they may find subtle and/or unreported migraine-associated extracephalic symptoms, which may in turn contribute to better patient management.

CONFLICT OF INTEREST

None.

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