BMJ Case Reports

Unusual association of diseases/symptoms

Gliadin allergy manifested with chronic urticaria, headache and amenorrhea

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Summary

Gluten intolerance is an autoimmune enteropathy caused by heterogeneous mixture of wheat storage proteins. Malabsorption symptoms imply diarrhoea, abdominal pain/bloating and weight loss. This case describes a 22-year-old female subject, who had chronic headache, joint pain, urticaria and long period of amenorrhea. Skin prick tests revealed a sensitisation to α-gliadin, while neurological, gynaecological, endocrine and clinical-laboratory examinations did not justify the above-mentioned symptoms. Gluten-free diet resolved chronic symptoms and re-established the menstrual cycle, whereas a temporary gliadin daily diet re-exacerbated all clinical symptoms. Urticaria occurred 20 min and the chronic headache the next day after exposure to the gliadin-rich diet. In addition, the missing of the expected menstrual bleeding was observed. This case demonstrates that gliadin intake can induce malabsorption and 'idiopathic' neuronal or gynaecological symptoms.

BACKGROUND

Gluten intolerance (GI) or celiac disease is an autoimmune enteropathy caused by gluten, a heterogeneous mixture of wheat storage proteins which includes α-gliadins.[1 2] Patients usually manifest malabsortive symptoms such as diarrhoea, abdominal pain/bloating and weight loss.[3] Some reports described the correlation of gluten consumption with non-abdominal symptoms.[3 4] GI can be associated by several cutaneus, neuro-psychological or gynaecological pathologies, such as herpetiform dermatitis, peripheral neuropathy, psychoses, amenorrhea, infertility, etc.[3–6] The successful treatment of these cases consists on the elimination of gluten or suspected gliadin epitopes from the daily diet.[1] We are reporting on an unusual combination of urticaria, headache, joint pain and amenorrhea of a patient, caused due to gliadin intake.

CASE PRESENTATION

This is case presentation of a 22-year-old female subject, who had chronic headache, urticaria and joint pain, associated by amenorrhea (a menstrual cycle every 4–5 months). She had reported on previous recidivant urticarial episodes occurring about 15–30 min after non-steroid anti-inflammatory drug intake (ibuprofen, ketonal or nimesulid), as well as under psychological stress. Reference to family history, her mother was diagnosed with bronchial asthma to house dust mites, and her aunt with allergic rhinitis to grasses.

INVESTIGATIONS

Neurological, gynaecological and endocrinological tests and clinical/laboratory examinations did not explain the above-mentioned symptoms. Thyroid, ovarian and hypophyseal hormonal levels were normal. Gynaecological ultrasound and join x-ray exploration did not reveal any pathological aspect. The skin prick test (pneumoallergens and trophoallergens) revealed a sensitisation to house dust mites, grasses, cereals and α-gliadin, even though she never complained on respiratory allergic symptoms. The allergologist recommended a gluten-free daily diet, referring to previous report on secondary amenorrhea due to gliadin intake.[3] Gluten-free diet resolved chronic symptoms and re-established the menstrual cycle at the expected time. A later temporary diet with gliadin re-exacerbated the clinical symptoms, including the absence of the expected menstrual cycle. In addition, urticaria occurred 20 min after re-exposure and the headache 1 day after.

DIFFERENTIAL DIAGNOSIS

Several medical specialists evaluated the case. Based on clinical, imagery and laboratory tests (see above) they excluded neurologic disorders (diverse chronic headaches such as migraine, etc); endocrine disorders (endocrine amenorrhea, or hypophysis-related disorders); gynaecological disorders (ovarian and uterine-originating amenorrhea); and rheumatologic disorders (rheumatologic arthritis).

TREATMENT

The patient conducts a strict gluten-free daily diet.

OUTCOME AND FOLLOW-UP

Momentarily the subject is symptom-free.

DISCUSSION

Patients with GI usually complain about malabsorption symptoms, but the occurrence of certain non-abdominal

systemic symptoms are also reported.[1–6] Gastrointestinal symptoms, associated with additional non-gastrointestinal manifestations (such as peripheral neuropathy, amenorrhea or infertility) are reported on a few case-reports.[3–6] Our case is an unusual description of an atopic subject who complained on chronic urticaria, headache and joint pain, as well as gluten-related amenorrhea. Gluten allergy was confirmed by positive result of skin prick test to α-gliadin, and the elimination/reintroduction of gluten from her diet. This is a helpful diagnostic method of the food allergy.[7] The allergologist recommended a gluten-free diet after neurological, gynaecological, hormonal and ultrasound explorations were non-conclusive.[3] Gluten reintroduction confirmed the diagnosis of IgE-mediated allergy and the association of gluten intake with further delayed symptoms. After that, the allergologist suggested the elimination of gluten from the daily diet and this completely normalised the clinical situation.

Learning points

- This report indicates the multiple systems' implication due to gluten allergy as a form of gluten intolerance, accompanied with amenorrhea. In similar cases, the application of skin prick test, gliadin elimination and reintroduction could be considered as appropriate screening tests on affected female subjects.

Competing interests None.

Patient consent Obtained.

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