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Short Report

Severe anal pain caused by food allergy?: A case report

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SUMMARY

Introduction: Food allergy such as cow milk protein allergy (CMPA) is most common in early childhood, but may persist or develop in later life. Gastrointestinal symptoms are a common manifestation of food allergy but anal pain is highly unusual.

Case presentation: We report the case of a ten-year-old boy who presented with severe anal pain that completely resolved on exclusion of cow milk from his diet.

Conclusion: Severe anal pain is rare in children; this case suggests that once other pathology has been excluded a trial of dietary exclusion of common food allergens should be implemented.

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1. Introduction

Manifestations of food allergy include gastrointestinal symptoms. Cow milk protein allergy (CMPA) as with other food allergy is most common in early childhood, but may persist or develop in later life. We report the case of a ten-year-old boy who presented with severe anal pain that completely resolved on exclusion of cow milk from his diet, and recurred with its reintroduction. Subsequently he came to tolerate milk and milk products, but reacted in the same way to chocolate.

2. Case presentation

A ten-year-old boy was referred for a surgical opinion by his general practitioner with a six month history of intermittent pain localised to the area of his anus and perineum. The pain was severe and troublesome, occurring repeatedly during the day and often persisting for up to an hour or more. It could be severe enough to make him cry, and unlike proctalgia fugax was not transient and sharp in nature. At such times he was unable to sit down, but gained some relief by lying prone. He reported passing a soft stool without difficulty every other day, but frequently noticed small amounts of

fresh blood and mucus on the toilet paper. Treatments that had failed to provide symptomatic relief included laxatives (lactulose and senokot), topical anaesthetic ointment (lidocaine) and glyceryl trinitrate. He was fit and well except for mild asthma which was under good control with regular inhaled treatment. A history of colic and regurgitation as an infant suggested possible CMPA in early life. He enjoyed playing football, had a good appetite and was growing normally. Both parents and two siblings were in good health, and there was no family history of illness. During the outpatient consultation his pain prevented him from sitting down. General clinical examination was unremarkable; the anus was normally sited and very mild inflammation of the perianal skin was noted.

Investigations showed normal haemoglobin of 130 g/L, white cell count $7.60 \times 10^9/L$, eosinophil count $0.19 \times 10^9/L$, platelet count $306 \times 10^9/L$, C-reactive protein 9 mg/L, total bilirubin 14 $\mu\text{mol/L}$, and alanine transaminase 27 IU/L. In view of symptom severity an examination under anaesthetic was arranged. This revealed a normally sited anus, with slightly friable perianal skin; no masses were palpable on digital examination, and proctoscopy showed only very mild inflammation of the rectal mucosa with an increased amount of mucus. Stool for parasites and perianal swab for group A haemolytic streptococci was negative. Rectal swabs sent for culture grew only mixed enteric organisms; nucleic acid amplification tests for *Neisseria gonorrhoea* and *Chlamydia trachomatis* were negative. Histological examination of rectal mucosal biopsy showed focal mild acute cryptitis with no evidence of glandular distortion and well preserved goblet cells. The changes were mild and insufficient to make a diagnosis of inflammatory bowel disease. In view of persistent symptoms, he underwent oesophago-gastro-

Abbreviations: CMPA, Cow's Milk Protein Allergy.

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duodenoscopy and colonoscopy. Endoscopic appearances were unremarkable, confirming only minimal erythema around the anus and in the rectum. Biopsy from the caecum showed some mild focal non-specific inflammation with an increase in eosinophils, but all other biopsies including those from the rectum were now normal.

The possibility that pruritis ani might sometimes be an oligo-symptomatic form of food allergic proctocolitis^{1–3} prompted us to consider whether this could also be true of anal pain. A trial of strict cow milk protein exclusion under the supervision of a paediatric dietitian, lead within a fortnight to a considerable reduction in both anal pain and the frequency of blood on the toilet paper; symptoms then completely resolved over the next three months. An unplanned cow milk challenge while staying away from home for the weekend resulted in a return of anal pain two days later. This once again resolved with exclusion of milk from his diet, only to recur three months later when milk chocolate was eaten. Two years later, he was able to reintroduce milk into his diet, but ingestion of chocolate continued to provoke pain and bleeding.

3. Discussion

Anal pain is highly unusual in children and may be idiopathic or a manifestation of coccydynia, infections of the rectum or perianal area, thrombosed external haemorrhoids, fissure in ano, tumour⁴ or nerve root pathology.⁵ In the light of a report of a child with pruritis ani precipitated by yogurt,⁶ we felt that a trial of milk exclusion was warranted in our patient with anal pain. The European Academy of Allergology and Clinical Immunology advises that in non-IgE-mediated food allergy presenting with gastrointestinal symptoms IgE based tests will be negative so that the diagnosis must rely principally on the history and the relapsing character of the symptomatology.⁸ In our patient, resolution of symptoms with cow milk exclusion and recurrence following challenges therefore strongly suggest the diagnosis of anal pain as a manifestation of CMPA. While the delayed reaction to cow milk protein and the time lapse between milk elimination and resolution of symptoms points more to a non-IgE-mediated reaction, our patient declined to have further blood tests or skin testing so that we were unable to exclude the possibility of an IgE-mediated response. Additional supportive features for a diagnosis of CMPA include the history of colic and regurgitation in infancy, atopic background (asthma), slight redness of the perianal skin on examination, and the histological findings in the colon of non-specific patchy inflammation, with eosinophils prominent in one biopsy. Although a double-blind placebo-controlled food challenge is the 'gold standard' investigation in selected cases of food allergy⁷ our patient did not agree to undergo this procedure. After a period of two years he came to tolerate milk and dairy products, but continued to react to milk chocolate suggesting sensitivity to cocoa or another component of this confectionary. Chocolate induced gastrointestinal symptoms associated with mucus in stools is well described in adults.⁸

Children with a history of CMPA in infancy are more likely than controls to suffer from abdominal pain at the age of 10 years, the mechanism involved remains uncertain.⁹ Symptoms in food allergy are considered to result from an aberrant immune response leading to inflammation and tissue damage. Food allergy is classified as IgE-mediated or non-IgE-mediated. The former is the best understood mechanism and more easy to diagnose since the onset of symptoms is often within minutes to hours of exposure. Poor understanding of the immunopathological mechanisms underlying non-IgE-

mediated allergy has hindered the development of simple and reliable diagnostic tests. These reactions tend to be delayed, with onset of symptoms occurring from 1 h to several days after ingestion of milk. IgE and non-IgE-mediated reactions are not mutually exclusive and reactions to food can involve a mixture of immunological mechanisms. Type-1 helper T cell (Th1) mediated reactions, the formation of immune complexes leading to activation of complement, or T cell/mast cell/neuron interaction inducing functional changes in smooth muscle activity and intestinal motility have all been implicated.¹⁰

4. Conclusion

Severe anal pain is rare in childhood; this case suggests that once other pathology has been ruled out, a trial of dietary exclusion of common food antigens may be worthwhile. Although a double-blind placebo-controlled food challenge is the most rigorous way of confirming diagnosis it is a complex process and therefore infrequently used in practice. Diagnosis can be made on clinical grounds in the light of response to dietary elimination and challenge.

Consent

"Written informed consent was obtained from the patient and his guardian for publication of this case report. A copy of the written consent is available for review by the Editor-in-Chief of this journal."

Conflict of Interest

The authors declare that they have no conflict of interests.

Authors' contributions

VZ and JWLP drafted the manuscript and reviewed the literature. JWLP and IDS were involved in the patient care and follow-up. All authors have read and approved the final manuscript.

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