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CASE REPORT | COLON

# Isolated Intestinal Angioedema Secondary to a Food Allergen

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# **ABSTRACT**

Intestinal angioedema is the dilatation or thickening, or both, of a segment of bowel. It is a diagnostic phenomenon that manifests itself clinically as acute abdominal pain, diarrhea, and emesis. Generally, this condition occurs in tandem with angioedema of the face and tongue and/or in association with the use of an angiotensin-converting enzyme inhibitor (ACE-I). We present a rare case of a 63-year-old woman who developed isolated intestinal angioedema due to the ingestion of a food allergen.

# INTRODUCTION

Classically, angioedema has a large genetic component (C1-inhibitor deficiency) and can be associated with the use of an angiotensin-converting enzyme inhibitor (ACE-I).<sup>1,2</sup> Additionally, the edema is usually located in the tongue and face or is at least concomitantly in the bowel as well.<sup>3</sup> The bowel mucosa is rarely the only site of angiodema.<sup>1</sup> Despite this, reports describing isolated intestinal angioedema do scantily appear in the literature. However, in most of these cases, the intestinal angioedema is secondary to the use of an ACE-I.

# CASE REPORT

A 63-year-old woman with no known medical history presented to the emergency room complaining of severe abdominal pain of acute onset. She mentioned that it occurred after drinking her daily green smoothie, after which she immediately developed severe abdominal cramping, nausea, vomiting, and watery diarrhea. She denied chest or back pain, blood in the stool, bloody emesis, facial or lip swelling, itching, or a rash. Upon further questioning, she mentioned the only addition to her normal morning smoothie was parsley. She remembered having a similar reaction years ago after ingesting kiwi fruit.

In the emergency room, she was febrile, tachycardic, and tachypneic. On physical examination, her abdomen was soft, nondistended, and tender to palpation, with normoactive bowel sounds throughout. Her laboratory results were notable for a white blood cell count of 18,000/mL and a lactate of 5.9 mmol/L. All other test results including blood cultures, stool analysis, erythrocyte sedimentation rate, cytoplasmic antineutrophil cytoplasmic antibodies (C-ANCA), perinuclear antineutrophil cytoplasmic antibodies (P-ANCA), C1-inhibitor, and complement levels were within normal limits. Computed tomography scan showed thickened bowel and submucosal edema, consistent with the diagnosis of isolated intestinal angioedema (Figure 1). She was given a 1-day course of antibiotics and antiemetics, after which her diet was advanced and soon after she was discharged home in the stable condition.

# DISCUSSION

Intestinal angioedema is marked tomographically by the dilatation and/or thickening of a segment of bowel.<sup>3</sup> This occurs in response to an offending agent, which in our patient was the new vegetable.<sup>1</sup> The most likely mechanism is immunoglobulin E-mediated mast cell degranulation releasing histamine leading to intestinal swelling.<sup>4</sup> Any hollow viscera is prone to this type of temporary swelling.

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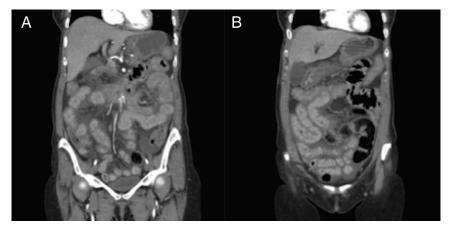


Figure 1. Abdominal and pelvic computed tomography showing (A) diffuse thickening of the small bowel and colonic walls and (B) the presence of mucosal enhancement and submucosal edema.

The severity of presentation of intestinal angioedema can vary, from mild colicky pain to severe vomiting and diarrhea.<sup>1</sup> Generally this condition occurs in tandem with angioedema of the face and tongue.<sup>1</sup> Moreover, nearly all cases are either genetic in nature due to a C1-inhibitor deficiency, which was not indicated in our case based on her normal C1-inhibitor levels, or associated with the use of an ACE-I, which acts as the offending agent.<sup>1,2,4,5</sup> Intestinal angioedema rarely occurs secondary to the ingestion of a food allergen.

Avoidance of the trigger is the best form of prophylactic treatment. Edema can be reduced by the administration of antihistamines, glucocorticoids, epinephrine, or a combination of these medications. Interestingly, fresh frozen plasma has also shown efficacy in aborting acute attacks.<sup>6</sup>

In conclusion, physicians must remain mindful of this entity in patients presenting with acute abdominal pain, vomiting, or diarrhea of unknown etiology. Attention to imaging combined with a scrupulous history can help reach a diagnosis. Although most cases typically resolve within 1–3 days, early recognition may prevent unnecessary imaging as well as possible exploratory surgery.<sup>4</sup>

#### **DISCLOSURES**

Author contributions: A. Hassan and S. Weissman wrote and edited the manuscript. S. Weissman, A. Hassan, and MA Sciarra drafted and critically revised the manuscript. MA Sciarra and

J. Sotiriadis provided images and edited the manuscript. S. Weissman and A. Hassan are the article guarantors.

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# REFERENCES

- Bork K, Staubach P, Eckardt AJ, Hardt J. Symptoms, course, and complications of abdominal attacks in hereditary angioedema due to C1 inhibitor deficiency. Am J Gastroenterol. 2006;101:619–27.
- Ciaccia D, Brazer SR, Baker ME. Acquired C1 esterase inhibitor deficiency causing intestinal angioedema: CT appearance. AJR Am J Roetgenol. 1993; 161:1215–6.
- De Backer AI, De Schepper AM, Vandevenne JE, Schoeters P, Michielsen P, Stevens WJ. CT of angioedema of the small bowel. AJR Am J Roetgenol. 2001;176:649–52.
- Palmquist S, Mathews B. Isolated intestinal type angioedema due to ACEinhibitor therapy. Clin Case Rep. 2017;5(5):707-10.
- Chase MP, Fiarman GS, Scholz FJ, MacDermott RP. Angioedema of the small bowel due to an angiotensin-converting enzyme inhibitor. J Clin Gastroenterol. 2000;31(3):254–7.
- Nzeako UC. Diagnosis and management of angioedema with abdominal involvement: A gastroenterology perspective. World J Gastroenterol. 2010; 16(39):4913–21.

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