

# Intestinal mucus accumulation in a child with acute myeloblastic leukemia

*Akut myeloblastik lösemili bir çocukta intestinal mukus birikimi*

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## Abstract

Intestinal mucus accumulation is a very rare situation observed in some solid tumors, intestinal inflammation, mucosal hyperplasia, elevated intestinal pressure, and various other diseases. However, it has never been described in acute myeloblastic leukemia. The pathogenesis of intestinal mucus accumulation is still not clear. Here, we report a 14-year-old girl with acute myeloblastic leukemia and febrile neutropenia in addition to typhlitis. She was also immobilized due to joint contractures of the lower extremities and had intestinal mucus accumulation, which was, at first, misdiagnosed as intestinal parasitosis. We speculate that typhlitis, immobilization and decreased intestinal motility due to usage of antiemetic drugs might have been the potential etiologic factors in this case. However, its impact on prognosis of the primary disease is unknown. (*Turk J Hematol 2009; 26: 207-9*)

**Key words:** Acute myeloblastic leukemia, intestinal mucus accumulation, parasite

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## Özet

İntestinal mukus birikimi, solid tümörler, intestinal inflamasyon, mukozal hiperplazi, intestinal basınç artışı gibi çeşitli hastalıklarda nadiren görülen bir durumdur. Ancak, bu durum akut miyeloblastik lösemili hastalarda bildirilmemiştir. İntestinal mukus birikiminin patogenezi ise hala tam olarak bilinmemektedir. Burada, 14 yaşında akut miyeloblastik lösemili, febril nötropenili ve tiffilitli bir kız hastayı sunduk. Hastamız, ayrıca alt ekstremitte eklem kontraktürü nedeniyle immobilize idi. İntestinal mukus birikimi nedeniyle başlangıçta intestinal parazitoz tanısı almıştı. Biz burada, tiffit, immobilizasyon ve kullanılan antiemetikler nedeniyle barsak hareketlerinin azalmasının potansiyel etyolojik faktörler olabileceğini düşündük. Ancak, bu durumun primer hastalığın prognozu üzerine etkisi bilinmemektedir. (*Turk J Hematol 2009; 26: 207-9*)

**Anahtar kelimeler:** Akut miyeloblastik lösemi, intestinal mukus birikimi, parazit

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## Introduction

Intestinal mucus accumulation is a rare clinical presentation manifested as mucocele in benign and malignant intestinal tumors and obstructions [1]. The entity has not been reported in patients with acute leukemia. Inflammation, mucosal

hyperplasia, elevated intestinal pressure similar to that seen in intestinal lesions, and mucus accumulation due to intestinal tumors play a role in the pathogenesis of intestinal mucus accumulation [2]. The morphology of worm-like mucus accumulations may be confused with parasites such as *Ascaris lumbricoides*, which can result in the unnecessary use

of antiparasitic drugs. In such cases, a pathology examination is required for definitive diagnosis. Here, we report an adolescent with acute myeloblastic leukemia and intestinal mucus accumulation.

### Case Report

A 14-year-old girl had been diagnosed with acute myeloblastic leukemia (FAB M2) and had been started on an acute myeloid leukemia Berlin-Frankfurt-Münster 2004 (AML BFM-2004) chemotherapy protocol at another center. After the induction therapy, febrile neutropenia and invasive pulmonary aspergillosis developed. She was given wide-spectrum antibiotics (meropenem, teicoplanin) and antifungal (voriconazole) treatment, and was also given total parenteral nutrition. During this time, she had lower right abdominal pain and constipation. Afterwards, a 35-cm long parasite was observed in her stool, and she was therefore treated with levamisole hydrochloride. She was referred to our hospital following the development of stupor during the consolidation phase of the chemotherapy protocol.

The physical examination revealed joint contractures due to immobilization. The findings of cranial magnetic resonance imaging were typical of a posterior reversible leukoencephalopathy syndrome. Her stupor resolved in one week, after which the remainder of her treatment protocol was started. After the consolidation phase, the patient developed febrile neutropenia and recurrent invasive pulmonary aspergillosis. On the 8<sup>th</sup> day of febrile neutropenia, she experienced vomiting, bloody stool, and again, lower right abdominal pain. Results of another physical examination were negative for abdominal masses, perianal lesions, and rectal masses. Thickening of the cecum wall (4.8 mm) was observed on abdominal ultrasonography. A diagnosis of neutropenic enterocolitis (typhlitis) was made.

She was treated with wide-spectrum antibiotics and antifungal combination therapy that included imipenem, amikacin, metronidazole, liposomal amphotericin-B, and caspofungin. She was also given granulocyte colony-stimulating

factor and granulocyte suspension infusions. Enteral nutrition was stopped and total parenteral nutrition was started. On the 10<sup>th</sup> day of neutropenia, a round, worm-like, yellowish substance approximately 35 cm long was observed in her watery stool (Figure 1). During this time, serum electrolyte levels were normal. Microbiologic examination of the stool showed no parasites or other pathogens. On pathologic examination, the sample was defined as acellular and basophilic mucus (Figure 2). We concluded that the material was due to mucus accumulation. Intestinal mucus accumulation did not recur. Treatment of the patient ended successfully. The patient has been followed without chemotherapy for 7 months and has had no problems.

Informed consent was obtained from the patient and the family.

### Discussion

Intestinal mucus accumulation is rare; it presents mostly as mucocele and is usually located in the appendix [1]. Clinical manifestations of appendiceal mucocele include lower right abdominal pain, nausea, vomiting, weight loss, changes in bowel habits, gastrointestinal bleeding, and a palpable abdominal mass [1,3,4]. Two major pathological mechanisms have been postulated for the formation of intestinal mucocele. One is elevated intestinal pressure as a sequela of luminal obstruction caused by inflammation, mucosal hyperplasia, or intestinal lesions such as fecaliths, endometriosis, diverticula, and polyps. The other is mucus accumulation due to intestinal tumors [2]. In diagnosing mucocele, imaging techniques including ultrasonography, computed tomography, and intestinal endoscopy may be beneficial [5,6]; however, certain diagnosis is always made by pathology.

The pathologic diagnosis in our patient was intestinal mucus accumulation but not mucocele, because no cells were seen in the specimen. To the best of our knowledge, there are no reports in the literature regarding intestinal mucus accumulation in patients with acute leukemia. In our patient, we concluded that the mucus accumulation had been caused



Figure 1. Macroscopic view of mucus accumulation

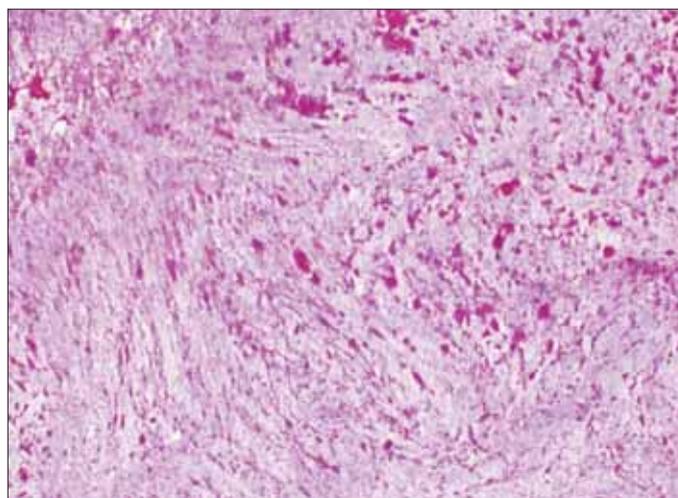


Figure 2. Basophilic mucus without cellular component (hematoxylin-eosin  $\times 10$  original magnification)

by neutropenic enterocolitis. The massive mucus production could have been caused secondarily by inflammation due to an invasive microorganism in the cecum.

Our patient had lower right abdominal pain, nausea, vomiting, weight loss, constipation, and gastrointestinal bleeding, as also seen in mucocele. The evaluation of the clinical findings plus the results of the abdominal ultrasonography showing intestinal wall thickening led to the diagnosis of neutropenic enterocolitis. However, we were unable to distinguish whether the mucus accumulation had any effects on the clinical manifestations or determine its impact on the prognosis of the primary disease. The intestinal mucus accumulation could have been the consequence of neutropenic enterocolitis or changes in the intestinal flora arising from the various wide-spectrum antibiotics administration or of the reduction in intestinal motility due to antiemetic use and immobilization. While we were unable to reach a diagnosis based on the clinical, microbiological, and radiologic methods, the definitive diagnosis was made with pathological examination.

In such cases, the clinician should be able to distinguish this situation from parasitosis to avoid unnecessary use of antiparasitic treatment. Pathologic examination is an efficient tool for the differential diagnosis.

No author of this paper has a conflict of interest, including specific financial interests, relationships, and/or affiliations relevant to the subject matter or materials included in this manuscript.

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