Case report

Pulmonary emboli causing sudden death in ulcerative colitis

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Sudden death as a complication of disease in children past infancy is rare. Sudden death in children with ulcerative colitis occasionally occurs following intestinal perforation, malignancy of the colon and surgical complications, but seldom preoperatively. Death secondary to bilateral pulmonary emboli associated with venous thrombosis is recognized as a complication of inflammatory bowel disease in adults, but has not been described in children.

The following case is that of a 15-year-old girl who died suddenly and unexpectedly from massive bilateral pulmonary emboli and is reported as an unusual complication and cause of death in ulcerative colitis in childhood.

Case report

A 15-year-old white girl first had bloody diarrhea in September 1967. Symptoms slowly increased in frequency and in December 1967 the diagnosis of ulcerative colitis was confirmed by barium enema and proctosigmoidoscopy. A low residue diet and an antimotility agent were prescribed. She continued to have one or two loose bowel movements daily. In December 1968 the diarrhea ceased and bowel movements were normal. In December 1969 diarrhea recurred and continued until November 1970, when the patient was first seen at the Bernalillo County Medical Center, Albuquerque, New Mexico.

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Before or after the onset of diarrhea, the patient experienced no extracolonic complications of the disease. There was no family history of inflammatory bowel disease. The history did not reveal any serious physical illness. Six months prior to the initial onset of diarrhea, the girl's mother, while under the influence of alcohol, was involved in an automobile accident resulting in the death of the other driver. The patient was emotionally upset for several months following the accident. The mother associated the onset of the patient's diarrhea with her emotional distress concerning the accident.

On November 17, 1970, the patient was admitted to the Bernalillo County Medical Center for institution of a medical program for relief of the diarrhea. She was attractive, well developed, but thin, and quite pale, weighed 42 kg, and was 167.6 cm tall. She was alert and cooperative. The physical findings included pallor of the skin, mucous membranes, and nail beds; a systolic murmur was heard along the left sternal border. There was slight tenderness of the abdomen in the lower left quadrant over the colon. The remainder of the physical examination and vital signs were normal. Proctoscopic examination revealed diffuse mucosal granularity and friability. Several small ulcers were seen. A rectal mucosal biopsy specimen obtained by suction was interpreted as "consistent with severe mucosal colitis." Barium examination of the colon revealed loss of normal haustration and mucosal pattern and narrowing of the entire colon.

The initial hematocrit value was 18.5% and the hemoglobin level was 5.3 g/100 ml. Five hundred milliliters of fresh, whole blood was transfused on the first hospital day and again on the fourth hospital day, after which the hematocrit value was 30%. She also received iron dextran injection (Imferon) intramuscularly while in the hospital. Therapy was started with salicylazosulfapyridine (Azulfidine), diphenoxylate hydrochloride with atropine sulfate (Lomotil), diazepam (Valium), vitamin B complex (Sur-bex) with ascorbic acid (vitamin C) and methylprednisolone retention enemas nightly. After 2 days of therapy, rectal bleeding ceased and during the next 6 days the frequency of loose bowel movements decreased. On the morning of discharge, November 25, 1970, her stool was formed. The same medical regimen that was instituted in the hospital was prescribed.

She did well for approximately 3 weeks, and then for 2 weeks prior to her return to the hospital on December 28, 1970, she had increased frequency of stooling without bleeding. On discharge from the hospital she had weighed 44 kg, and on return weighed 39 kg. She appeared weak, pale and depressed. On the day following admission her temperature was 39.3 C and she appeared to be in a toxic condition. Her abdomen was slightly distended with minimal tenderness in the left lower quadrant. Blood cultures were taken and no growth was reported 72 hours later. She was given tetracycline 500 mg intravenously every 6 hours, and her original medical program was restarted. This medication was continued until the day of her death. During the next few days her weight dropped to 35 kg. She then began to improve gradually. Her weight increased to 42 kg on the day before her death. She was up and around the ward daily and participated in the activities of the child care program. At no time was she in bed for more than a 12-hour period.

During the second hospitalization she was seen by a child psychiatrist. The patient vigorously and intelligently cooperated with the psychiatrist in daily sessions.

Five days before death she had an episode of crying and hyperventilation lasting about 1 hour. No chest pain was noted at that time, although epigastric pain, relieved by antacids, occurred 2 hours later. A roentgenogram of the upper gastrointestinal tract with barium and a roentgenogram of the chest on that day were normal. The following day the

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abdominal pain disappeared. Twenty-four hours prior to death she had a second episode of hyperventilation and anxiety which was again interpreted as an anxiety reaction. At no time during this 5-day period did she complain of pain in her legs, and there were no signs of circulatory disturbances.

On the day of death she was up and around the ward intermittently. Late in the afternoon she began to hyperventilate, and in a few minutes this was followed by vomiting, headache, unconsciousness, and a generalized clonic seizure. She was resuscitated successfully, became alert, expressed an expectation of impending death, and 5 minutes later again became unconscious. Resuscitation attempts were unsuccessful.

The postmortem examination revealed severe ulcerative colitis of the ascending, transverse, and descending colon and extensive recent bilateral thrombi completely occluding the pulmonary arteries. Intraabdominal and pelvic vessels did not contain thrombi, and dissection and examination of the vessels of the lower extremities were not performed. The cause of death was felt to be extensive bilateral pulmonary emboli.

Discussion

In retrospect, the hyperventilation and epigastric pain experienced by this child before her death may have been symptoms associated with small pulmonary emboli. The fear of impending death expressed by the patient may also have been associated with massive pulmonary emboli. The source of these emboli was not evident as no clinical signs or symptoms of thrombophlebitis existed. She had been making remarkable physical progress. Her mental state was also improving.

In adults with ulcerative colitis, thrombophlebitis and pulmonary embolism have been well documented.[1] In one retrospective study, 40 of 624 patients with the disease had venous thrombosis of the legs and 10 had pulmonary embolism.[1] In reviews of the disease in childhood, venous thrombosis and pulmonary emboli were not mentioned.[2-4] In a published abstract concerning extracolonic complications of the disease in childhood, reference is made to one case with thrombophlebitis.[5] Certainly death from bilateral pulmonary emboli in children with ulcerative colitis must be rare.

The etiology of venous thrombosis and pulmonary emboli in ulcerative colitis is obscure. Prolonged bed rest with venous stasis, corticosteroid therapy, hypercoagulability of the blood and trauma to vein walls have been thought to contribute to the pathologic process.[6-8] Stress has also been thought to affect the components of the blood coagulation mechanism.[9-13] Corticosteroid therapy and changes in blood coagulation associated with her physical and mental state most likely contributed to the formation of pulmonary emboli in this patient and caused her unexpected, sudden death.

Summary

A case of sudden death secondary to bilateral pulmonary emboli in a 15-year-old girl with severe ulcerative colitis is reported to alert physicians caring for children of another extracolonic complication of ulcerative colitis.

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