

Infant Rumination Syndrome

Report of a Case and Review of the Literature

David R. Fleisher, MD

• The infant rumination syndrome is an uncommon disorder difficult to differentiate from commoner conditions causing vomiting and weight loss. Its validity has recently been questioned. Its importance is heightened by the availability of newer diagnostic and operative procedures that might be misapplied. An 8-month-old male infant had been regurgitating for two months with increasing inanition. Results of diagnostic tests suggested pylorospasm or gastric pseudo-obstruction syndrome. Medical management failed. A therapeutic trial of special nursing eight hours a day resulted in rapid rehydration, weight gain, and avoidance of surgery. This case exemplifies each of the ten characteristics of the infant rumination syndrome.

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Infant rumination is an uncommon symptom, difficult to distinguish from vomiting and poor weight gain that are a result of diseases such as hiatus hernia, chaliasia, or gastroesophageal reflux with peptic esophagitis. The infant rumination syndrome has been well described,¹⁻⁹ and the theoretical basis for its pathogenesis and treatment have been ably presented in the pediatric and psychiatric literature. Recently, however, the importance of psychologic factors

has been questioned and the suggestion made that infants who ruminate do so because of a primary defect in the physiologic barrier to gastroesophageal reflux.¹⁰⁻¹¹ The patient described here was studied with the most modern diagnostic techniques, all of which failed to reveal any organic defect in gastroesophageal function. The importance of this very real illness is heightened by the availability of newer diagnostic and operative procedures that might be misapplied to infants whose vomiting should be diagnosed and treated more effectively by nontechnologic means.¹²

REPORT OF A CASE History

This 8-month-old infant began vomiting during the course of an upper respiratory infection when he was almost 6 months old. The respiratory symptoms cleared within a week, but frequent regurgitation persisted. There was no improvement with the use of antispasmodic drugs or elimination of milk. At age 7 months, he was hospitalized twice for intravenous hydration and diagnosis. Skull roentgenograms and an echoencephalogram were normal. Results of two upper gastrointestinal (GI) barium studies were normal. Esophageal manometry showed normal lower esophageal sphincter pressure (24.5 mm Hg) and normal relaxation with swallowing. Esophageal peristalsis was normal. Fiberoptic panendoscopy was normal. A third upper GI barium study showed antral spasm. Follow-up roentgenograms at 11 and 23 hours disclosed stasis of barium in the stomach.

Therapy consisted of intramuscular propantheline bromide; small, frequent feedings of soy formulas thickened with rice cereal; and upright positioning. Pressure lesions appeared on the buttocks six days later, and upright positioning was discontinued. The patient was discharged on a regimen of oral propantheline bromide, 1.75 mg four times daily (0.9 mg/kg/24 hr), with a diagnosis of pylorospasm and gastric motility disorder.

The patient returned eight days later still regurgitating and losing weight. He was hospitalized for the third time in order to instruct the mother in administration of propantheline by injection. The plan at the time of discharge was to consider pyloroplasty or gastrojejunostomy for correction of gastric retention should there be no improvement with intramuscular propantheline.

Regurgitation persisted despite propantheline injections at home. The patient was hospitalized for the fourth and final time at 8 months of age.

Physical Examination

Physical examination showed an apathetic infant with doughy skin turgor. At times, his mouth filled with gastric fluid; some would spill out, some was reswallowed. The mother spent much of the day devotedly sitting or standing near the crib. She held the baby only during feedings during which there was little eye contact.

RESULTS Hospital Course

Noninvasive observations during the following few days showed repeated episodes of regurgitation,

From the Department of Pediatrics, University of California at Los Angeles.

Reprint requests to 9735 Wilshire Blvd, Suite 480, Beverly Hills, CA 90212 (Dr Fleisher).

mostly occurring while the baby was alone in a passive, self-occupied state and usually ceasing whenever social eye contact was established with the observer. The mother and maternal grandmother were enlisted in an effort to hold the patient most of the day, to watch for the onset of his characteristic contractions of abdomen and pharynx, and immediately to respond by engaging him socially. Stressful procedures were discontinued to maximize the patient's physical and emotional comfort.

His weight stabilized, but the parents became exhausted within two days. The family agreed to hire a special nurse, who held the baby in the supine and upright positions while he was awake during the day. Formula was offered frequently but never force-fed. The patient's regurgitation decreased. His weight increased 1 kg during the next six days, with an accompanying fall in urine specific gravity (Fig 1). He was discharged on the 20th hospital day and continued on an unlimited diet and no medication. The mother agreed to have the special nurse help her with the baby during the ten days after discharge from the hospital.

Subsequent Course

Figure 2 shows the patient's weight before, during, and since his illness. Follow-up examination when he was 2½ years old found his weight to be near the 50th percentile; he had done no further vomiting and appeared well.

Description of the Family

The patient's mother suffered a severe emotional disturbance when she was 12 years old after the death of her father. Family disruption causing extreme rejection and loneliness was complicated by an out-of-wedlock pregnancy at age 16. This baby was given up for adoption. The mother married at age 23 and became pregnant with the patient when she was 30 years old.

The patient's father described himself as a "loner." His childhood was marred by emotional isolation. He was an executive who was made to feel indispensable by his superiors, and he

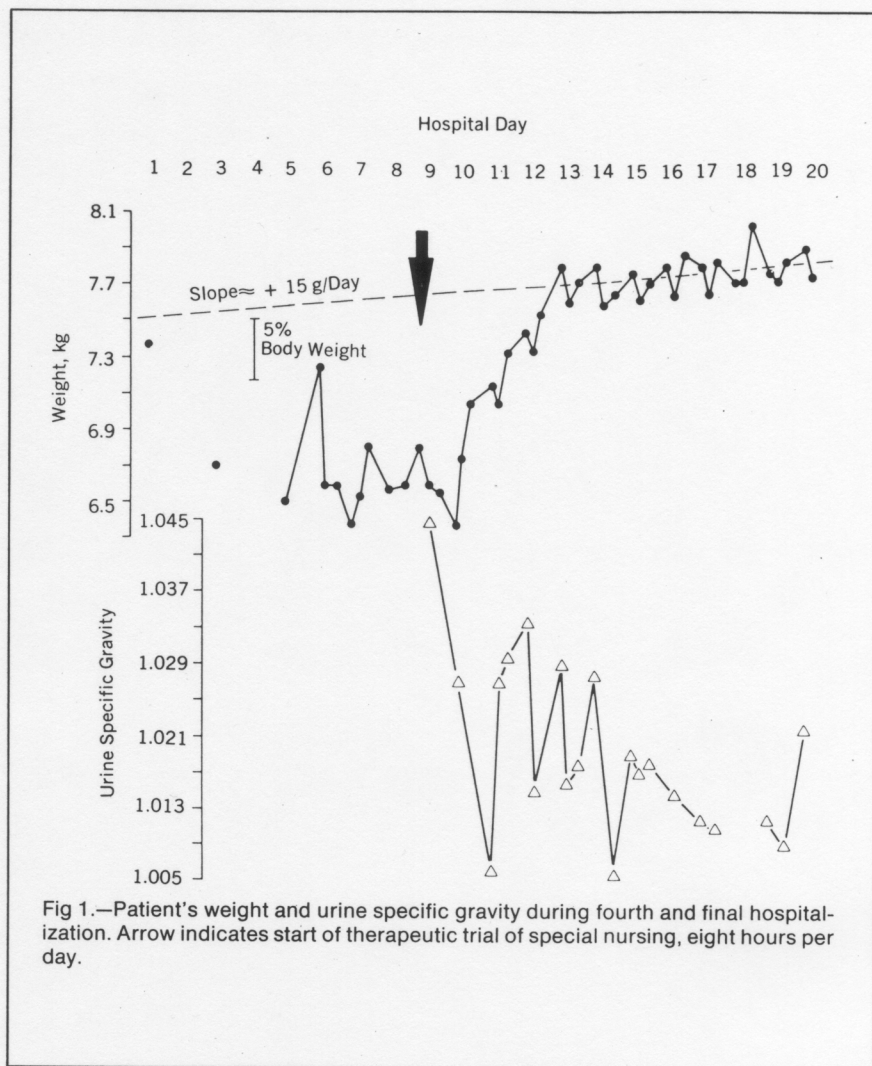


Fig 1.—Patient's weight and urine specific gravity during fourth and final hospitalization. Arrow indicates start of therapeutic trial of special nursing, eight hours per day.

often worked to exhaustion. The man suffered an episode of "nervous fatigue" at about the time his wife became pregnant with the patient. He praised his wife for being such an understanding person.

In a separate interview, she acknowledged that her husband was under great pressure from his work. Although she rarely complained, she described him as dictatorial, uncommunicative, and tired. She said she got along by virtue of her stoicism, personal religious commitment, and her continuous preoccupation with home and child.

COMMENT

In the present case as in most other reported cases, the correct diagnosis of infant rumination was made after a long, painful, and costly delay.

What causes these delays? First, a high index of suspicion is necessary for prompt recognition of the rumination process and to discern it from more common kinds of vomiting. Second, the diagnosis and management of infant rumination can only be achieved through use of interviewing¹³ and naturalistic observation,¹² and a collaborative parent-physician relationship.^{14,15} Third, ruminating infants sometimes display patterns of aberrant gastroesophageal function that may mislead the physician to more familiar but erroneous diagnoses. In the present case, one of three upper GI studies showed severe gastric retention,¹⁶ which prompted the diagnosis of gastric pseudo-obstruction syndrome or pylorospasm and the recommendation of gastrojejunostomy or pyloroplasty. Other reported

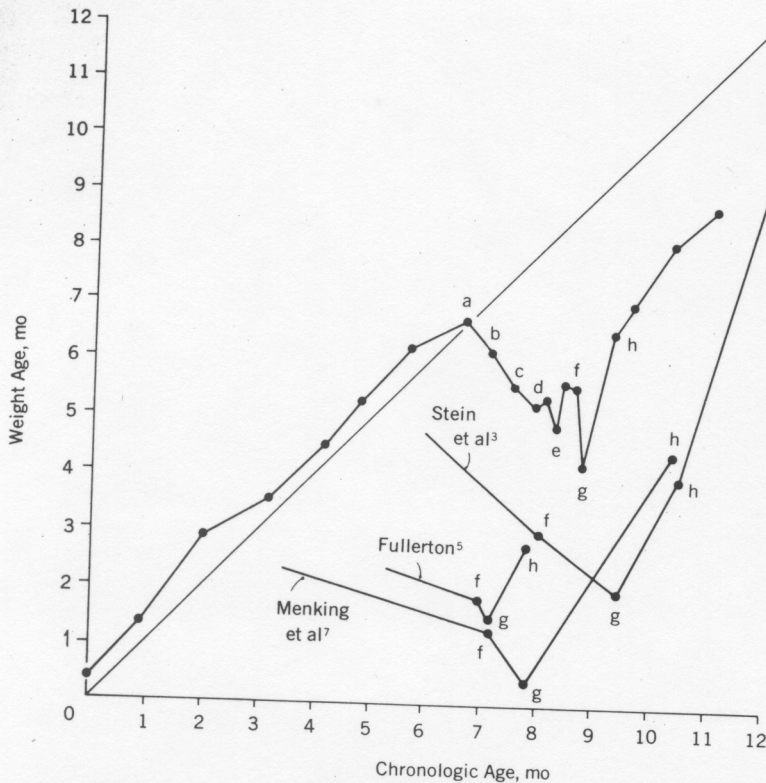


Fig 2.—Upper plot: patient's weight course: a, onset of vomiting; b, first hospitalization; c and d, admission and discharge, second hospitalization; e, third hospitalization; f, at time of fourth admission; g, at start of therapeutic trial of special nursing; h, at discharge from fourth hospitalization. Lower plots: f, on admission; g, at beginnings of therapeutic trials of special nursing; and h, at times of discharge of patients reported by Stein et al,³ Fullerton,⁵ and Menking et al.⁷

cases have shown evidence of free gastroesophageal reflux diagnosed as chalasia⁵ and hiatus hernia.¹⁰

The present case exemplifies the following ten characteristics of the infant rumination syndrome described in previously published reports: (1) The onset usually occurs between 3 and 8 months of age.²⁻⁷ (2) Loss of previously swallowed food causes increasing inanition.^{2-4,6-7,17} (3) The failure to thrive does not improve with thickened feedings, upright positioning, anticholinergic drugs, hand restraints, formula changes, or gastrostomy feedings.^{2-3,5} (4) The regurgitation is voluntary and done without distress.^{2-5,7} (5) Regurgitation usually does not occur during sleep or when the baby is actively interested in objects or people in his environment. It typically occurs while the baby is

awake, quiet, and self-absorbed. There may be other self-stimulation behavior such as head rolling, hand sucking, or sound-making.^{1-4,6-7,17} (6) The typical act of rumination begins with rhythmic contractions of the pharynx, tongue, and abdominal muscles; it culminates in the regurgitation of gastric contents into the mouth with more or less spillage to the outside. Whatever remains in the mouth is mouthed and reswallowed. The baby appears to enjoy this activity.^{1,4-5,7} (7) The appearance of the mothers' parenting may range from neglectful to almost slavishly attentive. But, in every case, there is an avoidance of enjoyable holding and difficulty in sensing what gives comfort and satisfaction to their babies.^{2-5,7} (8) The weight loss and rumination rapidly respond to nurturing

that is sensitive and interactive.^{1-2,4-7} (9) The aberrant nurturing relationship is one aspect of more pervasive difficulties the parents have with interpersonal relationships. There is usually a history of traumatic emotional deprivation during childhood and of marital dysfunction.²⁻⁸ (10) Once the rumination and failure to thrive has stopped, it usually does not recur, even in families whose overall mental health status remains poor.²⁻⁷

A theory for the pathogenesis of the infant rumination syndrome proposed by Richmond et al² is supported by earlier and more recent studies of the emotional and cognitive development of infants.¹⁸⁻²² Three phenomena emerge at about age 3 months: the baby begins to be aware of his separateness from the need-gratifying mother and experiences a feeling of helplessness when she is not responsive. Fortunately, two means of coping are also developing at that time. The infant becomes able to evoke a social response from the mother. He also becomes able to stimulate and entertain himself by producing interesting noises and activities. If the infant's efforts fail to elicit enough tension-relieving responses from the mother, he will resort to more self-stimulation. Brazelton et al²² studied face-to-face interaction between normal infants and mothers. They found a pattern of interactive behavior during which the baby's attention was closely coordinated with the mother's social behavior. When the mothers were asked to become completely unresponsive by sitting before their babies with a still face for three minutes, the babies responded first by becoming physically concerned and then by jerky movements, as they attempted to evoke a response from mother. When repeated attempts failed, the babies became withdrawn. This demonstration contributes toward the understanding of the development of rumination in infants of mothers with impaired responsiveness.

The diagnosis of rumination is confirmed by a favorable response to a therapeutic trial.^{23,24} Therapy for my patient consisted of employing one nurse eight hours a day to hold,

comfort, socially interact with, and feed the infant during his waking hours. The nurse was relaxed and maternal enough to enjoy this work. Although social stimulation was not continuous or forced, the nurse was sufficiently empathetic and observant to know when the baby slipped into the self-occupied withdrawal that fostered rumination, and she would promptly engage the baby in social interaction. The results of such management in the present case are similar to other reported cases for which weight data were given (Fig 2).

The best index of therapeutic response was the development of catch-up weight gain corroborated by a decrease in urine specific gravity that was due to improved hydration. Regurgitation did not stop abruptly; the patient regurgitated an average of 10.4 times per day and 5.8 times per day during the first and last five days of the 11-day therapeutic trial. Therefore, cessation of regurgitation should not be used as the most important indicator of the patient's course.

Another practical consideration of great importance is the mother's reaction to employment of a mother

substitute. She may think that the physician judges her to be inadequate. Such an understandable reaction should be anticipated and dealt with to prevent the collaborative relationship from deteriorating into an adversary one. Our respect for the parents was reinforced by trying to imagine what our own growing-up would have been like under similar conditions. Permission to employ the special nurse was requested of the parents for the purpose of getting help with difficult tasks of data collection and infant care. The nurse's role was that of helper to physician and mother, not didactic teacher of superior mothering technique.

Follow-up

The patient was seen after an interval of two years, at 37 months of age. Both height and weight were near the 50th percentiles, and he appeared well.

A sister was born when he was 29 months old. His behavior toward her was unusually affectionate. "He loves to get in bed with her, gives her toys, gives her her bottle, plays with her, hands me diapers when I change her,

has to walk right next to her when we go out," his mother said.

About one month previously, baby sister had been left for two weeks with the paternal grandmother in a distant city. During her absence, the mother said that her son "... just moped around, kind of lost." The parents noticed him chewing pieces of dinner he had eaten a few hours before. He swallowed all regurgitated material after chewing it, losing none from his mouth. This behavior was noticed repeatedly during his sister's absence. It ceased as soon as she returned home, except for one instance while she was napping and inaccessible to him.

This provides evidence of the occurrence of benign, adult-type rumination two years after recovery from the more pernicious infantile rumination with failure to thrive.²³ In this patient, loneliness seemed to trigger both episodes.

M. E. Greaney, Jr, MD, allowed the author to work with this patient; M. E. Ament, MD, and A. Euler, MD, performed the esophageal manometric and endoscopic studies; and Arthur Moss, MD, Eleanor Galenson, MD, R. T. Stone, MD, and A. Newburger, MD, gave criticism.

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