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Hypertrophic Pyloric Stenosis in Infants Fed via Transpyloric Tube : Three Case Reports

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Abstract

Hypertrophic pyloric stenosis (HPS) is a well-known cause of gastric outlet obstruction in infants. However, its etiology is still controversial. Our experience has let us to consider that transpyloric (TP) tube feeding may be a potential cause of HPS. We report 3 cases of HPS in the context of TP tube feedings. All 3 patients had severe fundamental surgical disease and required enteral nutritional support via TP tube for an extended period. Within several months of TP tube placement, their gastric residuals gradually increased and a small amount of non-bilious vomiting occasionally occurred. Ultrasonography revealed gastric outlet obstruction due to hypertrophied pyloric muscles. All 3 infants required pyloromyotomy. The total duration of TP tube insertion ranged from 40 to 146 days.

Of the 107 patients who received enteral nutrition support via TP tube at the National Center for Child Health and Development in the past 8 years, 3 (2.8%) developed HPS, an incidence approximately 15 times higher than the overall prevalence of infantile HPS in Japan, which ranges from 1 to 2 per 1000 live births (0.1-0.2%). To the best of our knowledge, only 16 cases of HPS that were considered to be related to TP tube feedings have been previously reported. HPS is a very rare complication, but one that must be taken into account when symptoms of delayed gastric emptying are seen during TP tube feedings.

Keywords: hypertrophic pyloric stenosis, transpyloric tube, enteral nutrition, complication

Introduction

Transpyloric (TP) tube feeding, as an alternative to nasogastric tube feeding, is selected in certain clinical circumstances. TP tube feeding is expected to improve a patient's respiratory condition by minimizing the risk of aspiration. Infants with severe respiratory distress, recurrent aspiration, gastroparesis, and postoperative gastro-esophageal reflux are good examples of patients for whom TP feeding is typically used. However, some potential problems with TP tube feeding must be noted [1,2].

Three infants were referred to us with hypertrophic pyloric stenosis (HPS) after prolonged TP tube feeding. To the best of our knowledge, only 16 cases of HPS in infants receiving TP tube feeding have been previously reported [3-7].

Case Reports

Case 1

Case 1 is a male infant who was diagnosed prenatally with right diaphragmatic hernia. He was born through a normal vaginal delivery at 38 weeks and 1 day of gestation and had a birth weight of 2832 g. He required long-term mechanical ventilation due to extreme hypoplasia of the lungs. Enteral nutrition was provided through a TP tube. Occasionally, an increase in residual gastric contents became evident, and he vomited small amounts. Abnormal gastric emptying and an elongated pyloric canal were demonstrated on upper gastrointestinal (GI) cineradiography. Ultrasonography (US) confirmed the finding of a hypertrophied pylorus compatible with the diagnosis of HPS (Fig. 1). Pyloromyotomy was performed, and the gastric outlet obstruction improved after the surgery. The total period of TP tube insertion before the definitive diagnosis of HPS was 146 days.

Case 2

Case 2 is a female infant who was prenatally diagnosed with an extremely large lymphangioma of the cervical area. She was born through Caesarean section at the gestational age of 37 weeks and 1 day, and had a birth weight of 3268 g. Mechanical ventilation was required due to airway compression by the tumor. TP tube enteral feeding was provided. She was noted to occasionally vomit small amounts, with a gradual increase in the frequency of vomiting. Upper GI cineradiography revealed delayed gastric emptying. US confirmed a hypertrophied pylorus (Fig. 2). After a pyloromyotomy, gastric emptying improved significantly. The total period of TP tube insertion before the definitive diagnosis of HPS was 44 days.

Figure 1.

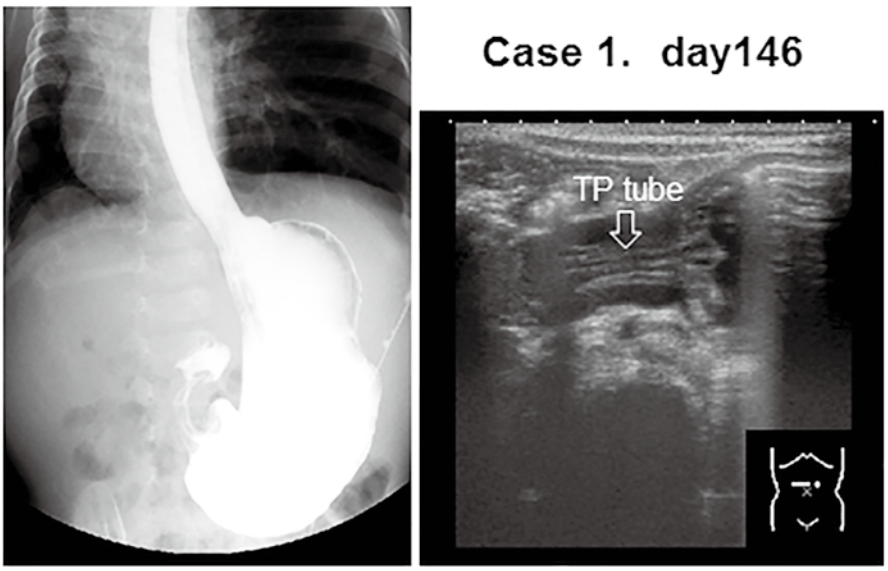

Figure 1. Upper gastrointestinal cineradiography showed abnormal gastric emptying and an elongated pyloric canal. Ultrasound showed a hypertrophied pylorus. The pyloric muscle thickness was 4.9 mm, and the canal length was 31 mm.

Figure 2.

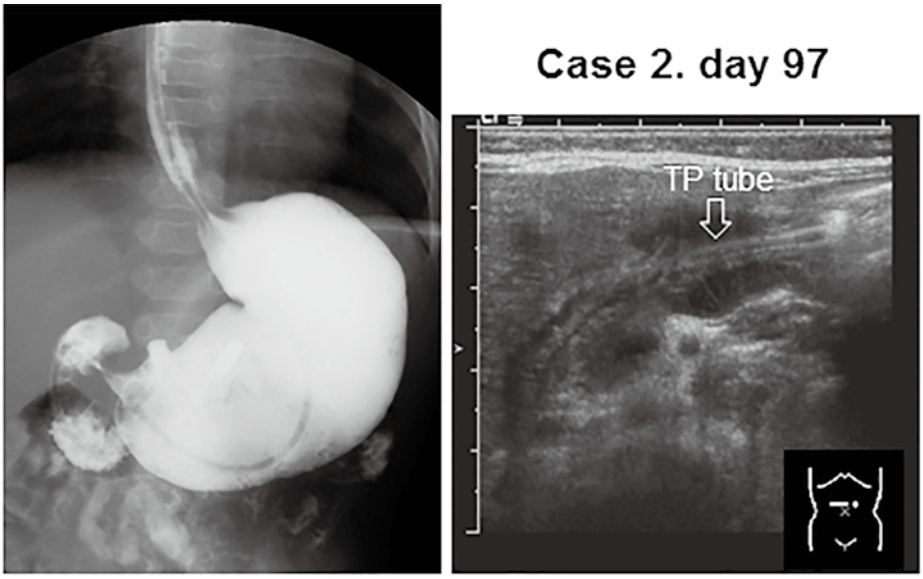

Figure 2. Upper gastrointestinal cineradiography showed gastroesophageal reflux and abnormal gastric emptying. Ultrasound showed a hypertrophied pylorus. The pyloric muscle thickness was 5.0 mm, and the canal length was 24 mm.

Figure 3.

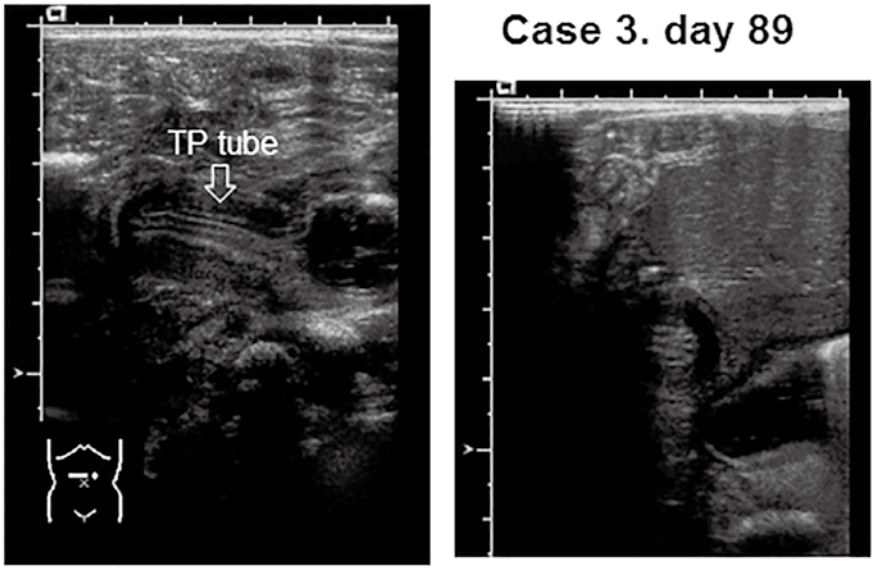

Figure 3. Ultrasound showed a hypertrophied pylorus. The pyloric muscle thickness was 6.6 mm, the canal length was 20.1 mm, and the canal diameter was 13.6 mm.

Case 3

Case 3 is a female infant who was prenatally diagnosed with omphalocele. She was born through Caesarean section at the gestational age of 36 weeks and 5 days and had a birth weight of 1622 g. Her respiratory status was unstable and mandatory mechanical support was prolonged. Erythromycin (EM) was administered for 3 weeks to prevent aspiration pneumonia and, subsequently, TP tube enteral feeding was started. A gradual increase in gastric residuals became evident; US confirmed HPS (Fig. 3). Pyloromyotomy improved the gastric clearance. The total period of TP tube insertion before the definitive diagnosis of HPS was 40 days.

US criteria for the definitive diagnosis of HPS

We used the following criteria to diagnose HPS on US: (1) wall thickness more than 3-4 mm on longitudinal and cross-sectional views, (2) pyloric diameter more than 15 mm on cross-sectional view, and (3) pyloric channel length more than 16-18 mm on longitudinal view [8-10].

Discussion

Infants who cannot maintain adequate oral intake due to pulmonary, cardiac, and/or neurologic disorders often require enteral tube feedings. Initially, such infants are fed through a nasogastric tube. Nevertheless, some of these infants suffer from gastroesophageal reflux, persistent vomiting, and recurrent aspiration due to gastric infusion of milk or formula. TP tube feedings are often selected as an alternative method of enteral nutritional support in such clinical situations. Some complications should be kept in mind when TP tube feeding is required. Displacement and clogging of the tube, mild transient epistaxis, nasal mucosal irritation, nasal skin erosion due to the fixed tape, feeding-related diarrhea, dumping-like symptoms, abdominal cramping, and hyperglycemia are common problems [1,2].

HPS was first mentioned as a complication of TP tube feedings in premature infants by Evans et al. in 1982 [3]. Since then, only 16 such cases have been reported (Table 1) [3-7]. To the best of our knowledge, literature is lacking regarding HPS directly caused by nasogastric tube feedings. The most common symptom of infantile HPS is projectile non-bilious vomiting [11], whereas the major symptoms of HPS associated with TP tube feeding are said to be an increasing volume of gastric residuals, an increase in the frequency and amount of vomiting, and difficulty in establishing oral or nasogastric tube feedings [3,4]. Compared with the projectile vomiting seen in infantile HPS, the symptoms of HPS in an infant receiving TP tube feeding are less obvious clinically. This is because

Table 1. Cases in the literature of hypertrophic pyloric stenosis (HPS) as a complication of transpyloric (TP) tube feeding

Cases		Mean gestation	Mean birthweight	Mean duration of TP tube insertion	Mean age at diagnosis
Evance NJ [3] (Sep, 1982)	2 males	27 weeks (26-28)	1.07 kg (0.87-1.27)	51 days (23-78)	90 days (78-91)
Raine PA [4]* (Oct, 1982)	4 males 2 females	30 weeks (29-33)	1.67 kg (0.93-2.81)	50 days (15-91)	67 days (45-93)
Muayed R [5]* (Aug, 1984)	6 males 4 females	30 weeks (28-34)	1.40 kg (0.93-2.81)	46 days (15-91)	unspecifed
Latchaw LA [6] (Aug, 1989)	3 cases	33 weeks (28-37)	2.58 kg (1.50-3.74)	47 days (21-90)	154 days (120-189)
Cosman BC [7] (Dec, 1992)	1 male	30 weeks	1.70 kg	16 days	35 days
Our cases	1 males 2 females	37 weeks (36-38)	2.57 kg (1.62-3.26)	77 days (40-146)	111 days (89-146)

(range)

Cases of HPS associated with TP tube feeding in infants. Sixteen cases have been reported; there was no difference by gender. The duration of TP tube insertion ranged from 2 weeks to several months. (* studies included the same cases). Although there is no clear description on the frequency, Raine et al estimated the risk of developing pyloric stenosis in infants with respiratory distress syndrome who had been fed via TP tube as 20 times greater than the normal population [4].

the major symptoms of HPS under TP tube feeding become gradually more evident with each passing day. In our 3 cases, we recognized the gastric outlet difficulties at 146, 44 and 40 days after the insertion of a TP tube. The etiology of HPS with TP tube feeding is not well-debated in the former reports. Environmental factors such as prematurity might be said to lead to a failure of the pyloric muscle to relax and to accelerated synthesis of growth factors [5,6]. Our 3 patients, however, were born at full-term. It was assumed that the TP tube in some way acted as an irritant and a stimulator for muscle hypertrophy.

EM, a motilin receptor agonist, is known to cause HPS in neonates. Four cohort studies concluded that infants exposed to a high dose of EM (about 40 mg/kg/day) in the first few weeks of life are at risk for developing HPS [12-15]. The patient described in Case 3 was treated with a therapeutic dose of EM (15 mg/kg/day) for 3 weeks from day 40. There was no clear causal relation between EM and HPS in Case 3. A standard amount of EM was administrated to the infant in this case.

We encountered 3 cases of HPS out of 107 infants who received temporary nutritional support with TP tube in the last 8 years. The fundamental disorders from which these 3 infants suffered were serious compared with the other 104 patients. The duration of TP tube feedings in the other 104 patients was 2 to 913 days (mean 114, median 41). There was no statistically significant difference in the placement period of TP tubes between the 2 groups ($p = 0.713$). Unfortunately, no further histological or hormonal investigation was performed, and the pathogenesis of HPS with TP tube feeding was not clear in our cases. Approximately 2.8% (3 out of 107) of TP tube-fed infants developed HPS. This incidence was about 15-fold greater than the prevalence of infantile HPS in Japan, which ranges from 1 to 2 per 1000 live births (0.1-0.2%).

The etiology of HPS itself remains a mystery [6,11]. Thus the effective policy to reduce the risk of HPS during TP tube feedings is not found at present. We should keep in mind that TP tube feeding may cause hypertrophy of the pyloric muscle and delay gastric emptying. It seems reasonable to perform an US assessment when TP tube-fed infants show an increasing volume of gastric residuals or an increasing frequency and amount of vomiting.

Conclusion

We encountered 3 cases of HPS in infants undergoing TP tube feeding. The etiology remains unknown, but the incidence was 15-fold (2.8%) compared to the overall prevalence of infantile HPS (0.1-0.2%) in Japan. Although it is a very rare complication, it must be taken into account when the symptoms of delayed gastric emptying are seen in an infant being fed via TP tube.

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経幽門経管栄養中に肥厚性幽門狭窄を発症した乳児３例

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肥厚性幽門狭窄は、新生児期から乳児時期の胃排出障害をきたす要因として周知されている。しかしながら、その病態生理は未だ不明な点が多く、明らかとはなっていない。今回我々は、市販のエンテラールフィーディングチューブを用いた経幽門経管栄養中に、幽門筋の肥厚による胃排出障害が惹起された３例を経験した。

呼吸器障害、循環器障害、あるいは神経系の障害のために経口摂取が困難な新生児においては、通常は経鼻胃管から母乳ないしミルクを注入し経腸栄養サポートを行っている。胃内注入により、呼吸窮迫を生じる、誤嚥を繰り返す、体位ドレナージによっても胃残が多い、胃食道逆流がみられる、などの場合には経鼻経幽門経管栄養を選択している。また、長期に人工呼吸サポートを要する状況で経腸栄養管理を行う場合にも誤嚥防止の観点から経幽門経管栄養が選択されることがある。胃を介さない経管栄養管理中の留意点としては、ダンピング様症状や高浸透圧性下痢症状などが一般的であるが、１９８２年に Evans らによって初めて経幽門経管栄養管理中に惹起された幽門筋肥厚症例が報告された。

自験３例は出生前診断された重度な右横隔膜ヘルニア、頚部巨大リンパ管奇形、臍帯ヘルニア症例であった。新生児期の手術後も、長期の人工呼吸換気を要し、この間の経腸栄養サポートとして経幽門経管栄養が選択された。チューブの留置から数か月間の間に、胃残は少しずつ増加し、少量の非胆汁性嘔吐が時折みられるようになった。腹部超音波検査により、幽門筋の肥厚が明らかとなり胃排出障害の要因と判断された。３症例とも粘膜外幽門筋層切開術を施行され、すみやかに症状の改善が得られた。チューブ留置期間は４０～１４６日間であった。

過去８年間で、総計１０７例に対して新生児期から経幽門経管栄養サポートを行っている。うち３例（2.8％）にのみ、肥厚性幽門狭窄の発症がみとめられた。両者のチューブ留置期間に統計学的有意差はみられなかった。本邦の肥厚性幽門狭窄発症頻度は、１０００出生に対し１～２例（0.1 ～ 0.2％）といわれており、経幽門経管栄養サポート下の同症発症率はおよそ１５倍に相当すると評価された。検索し得た限り、同様な症例の先行報告は１６例のみであった。通常みられる肥厚性幽門狭窄症の症状は噴水状嘔吐であり発見は容易であるが、経幽門経管栄養中の肥厚性幽門狭窄症状は胃残の漸増や少量嘔吐の持続などが主体であり、留意していなければ気づきにくい症状であった。経幽門経管栄養が幽門筋肥厚に及ぼす明らかな病態生理は未詳であるが、このような稀な事象があることは、今後、臨床上留意すべき点であると考えられた。