Chapter 1

Introduction

Bowel obstruction is a critical condition in pediatric populations. Undiagnosed conditions lead to morbidity and mortality. The etiologies of bowel obstruction are various and differ among age groups.Causes of bowel obstruction in pediatric patients can be congenital (Meckel's diverticulum, duplication cyst, malrotation or hernia), infection and inflammation (appendicitis, intussusception or inflammatory bowel disease), iatrogenic (adhesions), vascular (Henoch-Schonleinpurpura), neoplastic (adenocarcinoma) and others (ingested foreign body, distal intestinal obstruction syndrome or colonic volvulus).[1] Most common causes of bowel obstruction in infants and children is "intussusception".

Intussusception is a gastrointestinal disease. The two Latin words, i.e., "intuss", meaning within and "suscipere", meaning to receive, are combined together form intussusception. Thus, intussusception is the invagination of the proximal bowel in the distal part of the bowel. The invaginated bowel is called intussusceptum and the received bowel is called intussuscipiens.

The pathogenesis of intussusception may be causedby an inhomogeneity of longitudinal forces along the bowel wall. Imbalanced wall forces occur due to the leading point. The leading point can be nonpathologic, and is called an idiopathic and pathologic leading point. An idiopathic leading point is the thickened bowel wall lymphoid tissue at the ileocolic area called, Peyer's patch, the cause of 95% of intussusceptions. The pathologic leading point is found in about 1.5 to 12% up in reported studies. The incidence of the pathologic leading point increases with age, from 5% in the first year to 60% in children 5 to 14 years old. The inversions of Meckel's diverticulum are the most common pathologic leading points. The others can be the appendix, foreign body, parasite, mass etc.[2]

The occurrence of intussusception causes bowel obstruction. Not only the bowel, but also the mesentery of the proximal bowel is invaginated in the intussuscipiens. The mesenteric vein is compressed resulting in intestinal edema. The venous congestion leads to vascular sloughing of the mucosa combined with mucoid discharge from the colon resulting in currant jelly stool. When the reduction treatment is not accomplished, eventually the mesenteric artery will be compromised resulting in intestinal ischemia, gangrene and necrosis.[3]

Worldwide incidence of intussusception is about 1 to 4 in 2000 infants and children. The incidence of intussusception varies widely throughout the world. Jiang, 2013 studied the

distribution of intussusception around the world and reported mean incidence of 74 per 100,000 infants yearly. In Asia, about 100 per 100,000 were found accounting for 36 per 100,000 infants yearly in Thailand.[4]

Intussusception can be found in all ages with differing pathogenesis. More than 40% occur in 3 to 9 months of age. Idiopathic intussusception occurs uncommonly before 3 months and after 3 years. A total of 75% of cases are found within 2 years and 90% of cases are found within 3 years of age. Many studies show about 20% have a previous viral infection including the respiratory tract, gastrointestinal tract or both. Reports about rotavirus and adenovirus associated with the pathogenesis of intussusception has been proposed.[5]

Diagnosis of intussusception can be made with the clinical symptoms in 50% of cases. Radiologic investigation can help to confirm diagnosis in suspected cases and establish diagnosis in infants and children with unknown cause of small bowel obstruction. Plain abdominal X-ray, ultrasonography and contrast enema play a role in diagnosis.

The treatment aim of intussusception is to reduce the proximal invaginated bowel out of the distal received bowel. The treatment modalities comprise of surgical and nonsurgical reduction. Nonsurgical reduction can be performed safely when no contraindications are present. Contraindications are peritonitis, evidence of free air in the abdominal radiography and clinical evidence of shock. Nonsurgical radiologic reduction can be performed with hydrostatic or pneumatic reduction under ultrasonographic or fluoroscopic guidance. The success rate of reduction varies from 12.5 to 100% around the world.[6] The surgical treatment is done in the case of failed nonsurgical reduction and presence of contraindications.

Treatment methods vary widely. In some institutions, surgical management remains the initial method. Reports from Tanzania and Kenya show that 73 to 100% of patients underwent surgery as initial treatment after resuscitation due to limited resources and lack of referral system.[7, 8] In Thailand, the most common initial treatment is nonsurgical reduction when contraindications are not observed. The success rate of nonsurgical reduction in Thailand is about 50 to 68%.[9, 10] However, in some areas lacking specialists including pediatric surgeons, pediatricians and pediatric radiologists, the initial treatment is still surgery.

The treatment options of nonsurgical reduction of intussusception are not established in all areas of Thailand and the technique of reduction and radiological guidance varies. The success rate of reduction is not as high as some studies report. Studies are needed to fill in these knowledge gaps.

Scope of the thesis

This thesis was conducted in response to the three research questions below.

1. What is the reduction technique used most associated with better success rate of nonsurgical reduction of intussusception?
2. What are the factors that predict the reduction failure of intussusception?
3. Is constructing a simple prediction rule for reduction failure possible?

To answer those questions, we conducted 3 studies including pediatric intussusception patients from Chiang Mai University Hospital and Siriraj Hospital. These 3 retrospective cohort studies used the same data set to solve the 3 research questions under the titles listed below.

1. **Enema reduction of intussusception: the success rate of hydrostatic and pneumatic reduction**
2. **Prognostic indicators for failed nonsurgical reduction of intussusception**
3. **Clinical prediction rules for failed nonoperative reduction of intussusception**

The first part of this dissertation concerns the clinical manifestation and diagnosis of intussusception presented in Chapter 2. The second part concerns the modalities of nonsurgical reduction associated with study I presented in Chapter 3. The prognostic factors and clinical prediction rules of failed nonsurgical reduction of intussusception, associated with studies II and III, are presented in Chapter 4. The conclusion of nonsurgical reduction of intussusception, clinical application of the prediction rules and limitations are presented in Chapter 5.

Philosophical context of clinical epidemiology research including theoretical, data collection, and data analysis design for each study is presented in Appendix A. The three published studies are presented in Appendix B.

REFERENCES

1. Hryhorczuk AL, Lee EY. Imaging evaluation of bowel obstruction in children: updates in imaging techniques and review of imaging findings. Semin Roentgenol. 2012;47(2):159-70.
2. Columbani PM, Scholz S. Intussusception. In: Coran AG, Adzick NS, Krummel TM, Laberge J-M, Shamberger RC, Caldamone AA, editors. Pediatric Surgery. 2. 7th ed. the United States of America: Saunders; 2012. p. 1093-110.
3. Maki AC, Fallat ME. Intussusception. In: George W. Holcomb III, J. Patrick Murphy, Ostlie DJ, editors. Ashcraft's Pediatric Surgery. 1. 6 ed. China: Elsevier Saunders; 2014. p. 531-8.
4. Jiang J, Jiang B, Parashar U, Nguyen T, Bines J, Patel MM. Childhood intussusception: a literature review. PLoS One. 2013;8(7):e68482.
5. Ukarapol N, Khamrin P, Khorana J, Singhavejsakul J, Damrongmanee A, Maneekarn N. Adenovirus infection: A potential risk for developing intussusception in pediatric patients. Journal of medical virology. 2016.
6. Daneman A, Navarro O. Intussusception. Part 2: An update on the evolution of management. Pediatric radiology. 2004;34(2):97-108; quiz 87.
7. Chalya PL, Kayange NM, Chandika AB. Childhood intussusceptions at a tertiary care hospital in northwestern Tanzania: a diagnostic and therapeutic challenge in resource-limited setting. Ital J Pediatr. 2014;40(1):28.
8. Omore R, Osawa F, Musia J, Rha B, Ismail A, Kiulia NM, et al. Intussusception Cases Among Children Admitted to Referral Hospitals in Kenya, 2002-2013: Implications for Monitoring Postlicensure Safety of Rotavirus Vaccines in Africa. Journal of the Pediatric Infectious Diseases Society. 2015.
9. Khumjui C, Doung-ngern P, Sermgew T, Smitsuwan P, Jiraphongsa C. Incidence of intussusception among children 0-5 years of age in Thailand, 2001-2006. Vaccine. 2009;27 Suppl 5:F116-9.
10. Kruatrachue A, Wongtapradit L, Nithipanya N, Ratanaprakarn W. Result of air enema reduction in 737 cases of intussusception. Journal of the Medical Association of Thailand = Chotmaihet thangphaet. 2011;94 Suppl 3:S22-6.