

Concurrent Streptococcal Pharyngitis and Appendicitis in a Boy with Neurodevelopmental Disorder

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Abstract Acute appendicitis is the most common surgical emergency in children. A Japanese 8-year-old boy with attention deficit hyperactivity (ADHD) and comorbid autism spectrum disorder (ASD) exhibited fever, anorexia, and frequent vomiting. Gastrointestinal symptoms were initially regarded as associated with streptococcal pharyngitis and adverse effects of ADHD medication. Empirical antibiotic therapy did not improve his clinical condition. Absence of abdominal distension and tenderness led to a misled diagnosis and delayed diagnosis and treatment of acute appendicitis. Surgical intervention was made eventually for perforated appendicitis. He recovered without sequelae. Streptococcal pharyngitis with concurrent appendicitis is rare. Children with ASD sometimes have difficulty explaining their signs because of communication problems and insensitivity to pain. Special consideration is necessary when examining children with neurodevelopmental disorders, especially in emergency surgical conditions.

Keywords: appendectomy, attention deficit hyperactivity (ADHD), autism spectrum disorder (ASD), methylphenidate hydrochloride, strep test

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1. Introduction

The most common surgical emergency in children is acute appendicitis [1]. Appendicitis typically begins with abdominal pain, low-grade fever, anorexia, and abdominal tenderness [2]. Precise diagnosis of appendicitis is often difficult because many variants of appendicitis symptoms and signs occur [2]. Pain might not be localized, particularly in infants and children [2].

Streptococcal pharyngitis frequently shows various gastrointestinal manifestations including nausea, vomiting, and abdominal pain, which sometimes mimic acute appendicitis [1,3]. Concurrent occurrence of streptococcal pharyngitis and appendicitis is rare [1]. The association between pharyngitis and appendicitis is particularly deceptive because those overlapping symptoms must complicate the making of a definitive diagnosis of acute appendicitis [1]. Seeing patients with gastrointestinal symptoms associated with streptococcal pharyngitis still poses clinical challenges.

Attention deficit hyperactivity (ADHD) and autism spectrum disorder (ASD) are neurodevelopmental disorders. ASD and ADHD often co-occur and might share some clinical features [4]. Detection of serious illness is well known to be missed or delayed often in children with ASD because of their communication

difficulties and lower sensitivity to somatic pain [5]. Herein, we report a boy with ADHD and comorbid ASD presenting with rare association between streptococcal pharyngitis and acute appendicitis.

2. Case Report

An 8-year-old Japanese boy was ill, reporting a five-day history of vomiting, anorexia, and intermittent fever. Many cases of streptococcal infection were reported at his school. A rapid strep test (BinaxNOW® Strep A Test; Alere, Abbott) gave positive results 3 days before admission. A physician diagnosed streptococcal pharyngitis and prescribed oral azithromycin hydrate (10 mg/kg/day for 3 days). Non-bilious vomiting containing no mucous persisted. He was then referred to our hospital.

He regularly attended a mental health clinic. He showed not only inattention and impulsivity but also showed impaired communication and socialization. He had no intellectual disability. He was diagnosed as having ADHD and comorbid ASD based on DSM-5 criteria [5]. He took methylphenidate hydrochloride (18 mg/day).

On admission, he was lethargic and emotionless with a temperature of 38.0°C and oxygen saturation of 98% on air. The throat was infected slightly, without strawberry tongue or any skin rash. No throat swab culture was obtained. His chest was clear with equal air entry. He had

no complaint of tenderness. Abdominal examination was unremarkable. Urinalysis showed 1+ proteinuria and 2+ ketonuria without pyuria. The white blood cell counts were 17,100/mm³ with neutrophils of 83%, C-reactive protein 21.6 mg/dl, blood urea nitrogen 13.8 mg/dl, creatinine 0.48 mg/dl, sodium 123.4 mEq/L, potassium 4.4 mEq/L, and chloride 87.8 mEq/L. Chest X-ray was normal. Blood culture was negative. He was resuscitated with fluid bolus and reassessed. Intravenous piperacillin of 3 g/day was administered, yielding no improvement.

On hospital day 3, he was transferred to another hospital on request from his mother. He was completely anorexic with temperature of 37.0°C. Subsequent abdominal examination revealed no tenderness, but the abdomen became slightly distended and tense. Abdominal ultrasound confirmed acute perforated appendicitis and retroperitoneal abscess (Figure 1). He received surgical intervention. Delivery and lavage of the bowel was conducted together with appendectomy and lavage. He recovered without sequelae.

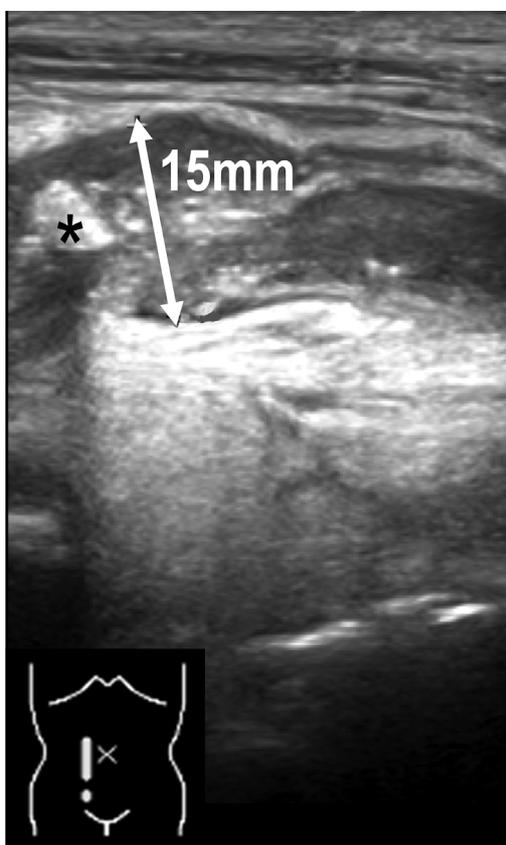


Figure 1. Abdominal ultrasonography shows a swollen appendix with fecal stone (asterisk): well compatible with a diagnosis of acute appendicitis

3. Discussion

This case highlights two important clinical issues. Concomitant streptococcal pharyngitis and appendicitis are rare. Children with neurodevelopmental disorders require special consideration when diagnosing acute appendicitis.

First, concomitant streptococcal pharyngitis and appendicitis are uncommon. Frequent vomiting and anorexia were initially thought to be associated with streptococcal

pharyngitis and adverse effects of methylphenidate hydrochloride [1,3,6]. Nielsen et al. reviewed cases of 1,572 children with abdominal pain at an emergency department, although none of 35 children with streptococcal pharyngitis had appendicitis [1]. Fulminant streptococcal infection usually causes peritonitis and intra-abdominal infection: in three cases, *Streptococcus pyogenes* grew from a blood or ascites culture [7,8,9]. Ghadage et al. reported a case of pneumococcal appendicitis [10]. In another comorbid case, the excised appendix culture showed *Escherichia coli* and *Pseudomonas aeruginosa* [11]. The clinical course of our patient was not rash. No culture of intra-abdominal abscess was obtained. The fulminant streptococcal infection was not applicable in this case. He might have been merely a streptococcus carrier in whom acute appendicitis occurred coincidentally.

Second, special consideration must be devoted to children with neurodevelopmental disorders in diagnosis of acute appendicitis. The primary physician happened to use azithromycin at first under circumstances that 30–40% of streptococcus is macrolide-resistant [12]. The first line treatment should be penicillin [12]. Inadequate first treatment by azithromycin might mask the presence of appendicitis. He actually never complained of abdominal distension or tenderness, although we repeatedly asked him about it. Children with ASD tend to struggle to express their signs because of communication difficulties and hyposensitivity to their somatic pain, which might delay detection of serious illness [5,13]. More polite inquiry and more careful abdominal examination are expected to be more effective. Lacking such care, no definitive diagnosis of appendicitis might ever have been made in this case.

4. Conclusion

Acute appendicitis is the most common target of surgical intervention in children. Childhood appendicitis sometimes shows protean manifestations, which might lead to misdiagnosis or delayed diagnosis. In fact, concurrent streptococcal pharyngitis and appendicitis are rare, but are expected to be likely. Children with ASD, because of their communication problems and their lessened sensitivity to somatic pain, are not good at explaining their symptoms by themselves. Special consideration is necessary for children with neurodevelopmental disorders, especially in emergency surgical conditions.

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