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**CLINICAL SERIE** 

# Complicated acute appendicitis in pediatric patients with "empacho" and abdominal manipulation history: case series

Apendicitis aguda complicada en pacientes pediátricos con antecedentes de "empacho" y manipulación abdominal: serie de casos

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# What do we know about the subject matter of this study?

Acute appendicitis is a surgical emergency that must be treated immediately. Delay in diagnosis and treatment leads to various complications. Empacho is known as a folkloric syndrome with a highly variable clinical picture.

# What does this study contribute to what is already known?

The belief of empacho has an indirect influence on the clinical deterioration of acute appendicitis, through the diagnostic and therapeutic delay of it, which contributes to the generation of different complications already known.

#### **Abstract**

According to Latin American popular culture, *empacho* is a gastrointestinal disease caused by excessive intake of cold or under-cooked food. Abdominal manipulation or *sobada* is one of the popular treatments used for *empacho* and consists of an abdominal massage, where the skin is rubbed and stretched, applying pressure on the abdomen. **Objectives**: To characterize clinical evolution and complications secondary to diagnostic delay in pediatric patients with acute appendicitis, with history of *empacho* and abdominal manipulation. **Patients and Method:** Descriptive, observational,

# Appendicitis; Perforated Appendicitis;

**Keywords:** 

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prospective study of pediatric patients with complicated acute appendicitis and history of abdominal manipulation, identified in the pediatric surgery room between November 2019 and June 2020 at the Mario Catarino Rivas Hospital. Descriptive statistics were used for analysis. **Results**: 10 patients were studied, aged ranging from 6 to 16 years (median age 14 years). Half of them were from rural areas. The main symptoms that appeared before abdominal manipulation were vomiting, nausea, and loss of appetite, followed by sudden diarrhea or constipation, and increased pain intensity. Complications were gastrointestinal perforation (5/10), appendicular mass (3/10), intra-abdominal abscess (3/10), and gangrenous appendicitis (1/10). The median time between initial symptom and hospitalization was 60 hours [IQR, 33.0 – 72.0]. **Conclusion**: This study describes the influence of the *empacho* and its respective treatment on clinical deterioration due to the delay in diagnosis and treatment of acute appendicitis, resulting in different complications.

## Introduction

Appendicitis is the inflammation of the vermiform (worm-shaped) appendix, a narrow, pouch-like, blind-ended extension around 5 to 9 cm long of the cecum opening<sup>1,2</sup>. In the first year of life, the appendix has a funnel morphology, with a low predisposition to obstruction, because of the lymphoid follicles that can obstruct the appendix reach a maximum size during adolescence; however, infantile appendicitis is the main etiology of acute abdomen in preschool and school children worldwide<sup>3</sup>.

The exact pathogenesis of acute appendicitis is multifactorial, which main cause is the obstruction of the proximal lumen, produced by mucosal inflammation, lymphoid hyperplasia, or a fecalith, triggering an accumulation of mucosal secretions of the distal tract, with an increase in intraluminal pressure, collapsing the venous system and its circulation, with suppurative transmural inflammation. If the obstruction persists, the congestion causes ischemia with gangrene and finally the appendix perforation<sup>4-6</sup>.

Classically, the clinical picture of appendicitis has been described as the gradual onset of dull periumbilical pain that migrates to the right lower quadrant. This picture usually occurs with additional symptoms such as nausea, vomiting, loss of appetite, fever, and less frequent diarrhea<sup>7</sup>. The nonspecific clinical presentation in children under 5 years of age, as well as difficulties in communicating with them, inadequate physical examination, irritability, and overlap of symptoms with other common childhood illnesses, contribute to the late diagnosis of acute appendicitis and high rate of misdiagnosis<sup>8</sup>.

The insidious clinical presentation of acute appendicitis in pediatric patients, the lack of knowledge of the pathology, and sociocultural beliefs are all factors that may influence parents and/or caregivers to consider *empacho* (indigestion) as a diagnosis in children with acute appendicitis. In Latin American medical

history, there are reports on *empacho* since the XVI century, making it a recognized disease, within popular beliefs<sup>9</sup>. This disease has demonstrated a historical continuity and a pattern of generational cultural transmission throughout Latin America. This popular knowledge has indigenous American and European Mediterranean origins, indissolubly fused<sup>10,11</sup>.

In a review study by Campos-Navarro R. (2016), *empacho* is defined as intestinal indigestion, usually with diarrhea, which affects mainly infants and young children, occurring mainly in children after eating undercooked or cold food<sup>11</sup>. The clinical picture of *empacho* resembles a classic picture of acute appendicitis, therefore many parents go to *sobadores* (rubbers) or relatives to treat the clinical picture of *empacho*, consisting of natural herbal remedies and abdominal manipulation<sup>12</sup>.

Abdominal manipulation, also known as "sobada", is a kind of abdominal massage, in which the patient's abdomen is rubbed, stretched, and pressed to try to cure *empacho* syndrome<sup>13</sup>. Therapists often use oils or butter to facilitate the movements<sup>14</sup>, and after such procedure, a spoonful of olive oil, purgatives, and/or tea is administered, where a variety between 17 and 39 species of herbs are used for its elaboration<sup>15,16</sup>.

However, when dealing with acute appendicitis, abdominal manipulation contributes to diagnostic delay, modifying the clinical picture and making it difficult for the surgeon to identify, resulting in the possible complications of acute appendicitis<sup>17</sup>.

The objective of this study is to characterize the clinical evolution and complications detected in a series of pediatric patients, diagnosed with complicated acute appendicitis, and with a history of abdominal manipulation for suspected *empacho*.

#### **Patients and Method**

Observational, descriptive, prospective, case series study. We analyzed a series of 10 pediatric patients

post-operated due to complicated acute appendicitis, who had history of abdominal manipulation, detected in the pediatric surgery room between November 2019 and June 2020, at the Dr. Mario Catarino Rivas National Hospital, a second level care hospital, which is a major referral center for the entire northwestern area of Honduras.

We included pediatric patients diagnosed with acute appendicitis, who received abdominal manipulation by non-medical personnel due to suspicion of *empacho*. Acute abdominal cases of etiology other than acute appendicitis and simple appendicitis were excluded, as in the case of 3 patients with history of abdominal manipulation and postoperative diagnosis of simple appendicitis. All pediatric patients who met the inclusion criteria and whose clinical history confirmed abdominal manipulation were included.

A form was created to collect the variables with content validity by experts. Through a direct interview with the patient, we consider studying the clinical picture registered in the admission record (symptomatology, time of evolution and intensity of pain, and hospital stay), sociodemographic variables: age (treated as quantitative data), sex (male or female), zoning (rural and urban), parental schooling (no schooling, primary, secondary, and higher education), laboratory tests (blood count, CRP), and the clinical picture before and after abdominal manipulation. To quantify the variable of abdominal pain intensity, scales were used according to the patient's age: the Wong-Baker faces pain scale for patients aged 3 to 10 years<sup>18</sup> and an analogous numerical pain scale for those aged 10 to 18 years.

The data were recorded in comparison tables in the Statistical Software IBM SPSS version 25.0 (license in force), which was used for the statistical analysis. Descriptive statistics were used to obtain the frequencies and percentages of the categorical variables, and the median and interquartile ranges for the quantitative ones.

The project was approved by the institutional ethics committee of the Universidad Nacional Autónoma de Honduras under number #003-04. After approval of the study protocol, patients were actively recruited in the pediatric operating room, informed consent signed by the parents or guardians was requested, as well as the consent of the pediatric patient before inclusion.

#### Results

A sample of 10 pediatric patients with complicated acute appendicitis who received abdominal manipulation was analyzed. The median age was 14 years [IQR 11.8-15.0 years] ranging from 6 to 16 years. The 13-14

years age group presented most of the cases (4/10), followed by the 15-16 years age group (3/10). Six patients included in the study were male. Half of the patients resided in rural areas. Regarding the academic level of the parent or guardian responsible for the patient, six of them had completed elementary school, one had no schooling, and three parents had completed higher education (Table 1).

Abdominal pain was the initial symptom in 9 of the patients included in the series; only one patient presented fever as the initial symptomatology. The median pain intensity was 5 points [IQR 5.0-6.5 points] on the analog pain scale. Two patients reported intensity of 8/10 on the analog pain scale. All patients presented with abdominal pain in their clinical picture, nine of them located in the right lower quadrant, and nine patients reported having subjectively high fever in the first hours of the pain picture (Table 2). The main presenting symptoms before the abdominal manipulation were: vomiting (10/10), nausea (9/10), loss of appetite (8/10), Dunphy's sign known as abdominal pain with coughing (8/10), and abdominal hypersensitivity (7/10). The median time between the onset of the initial symptom and abdominal manipulation was 24 hours [IQR 19-30 hours].

After abdominal manipulation, patients reported a gradual clinical deterioration and increased pain intensity (7/10), with a generalization of abdominal pain in the four quadrants (6/10). Pain intensity increased on the analog scale, with a median of 8 points [IQR 6.5-10]. Diarrhea (4/10) and constipation (4/10) were added, while the presence of nausea (9/10), vomiting (10/10), and loss of appetite (8/10) remained constant (Table 3).

At the time of hospitalization, nine patients presented McBurney's sign (pain on abdominal palpation caused by pressure between the external 1/3 with the internal 2/3 of an imaginary line drawn between the right anterior superior iliac spine and the umbilicus) and positive rebound tenderness (pain on abdominal decompression).

In the blood count, 3/10 patients showed leukocytosis between 10,000-15,000/mm³, 2/10 between 15,000-20,000/mm³, and 3/10 over 20,000/mm³, and 2 cases presented leukocyte values below 10,000/mm³. Complementary tests included C-reactive protein in the normal range in 9 patients and urinalysis without alterations. Imaging studies were not performed in the patients analyzed in this study, due to the surgical emergency they presented.

All patients underwent surgery within hours of diagnosis. The most frequent complications were: intestinal perforation (5/10), appendiceal adhesions (3/10), intra-abdominal abscess (3/10), and other complications such as gangrenous appendicitis (1/10)

(Table 2). Case n°7 presented three consecutive complications: intestinal perforation, appendiceal adhesion, and an intra-abdominal abscess.

The mean hospital stay of all patients was 7-14 days, due to the different complications they presented. The median time between the initial symptom and hospitalization was 60 hours [IQR 33.0-72.0], equivalent to more than two and a half days to go to a health care center.

#### Discussion

Abdominal pain is one of the main symptoms of pediatric patients seen in the emergency department, and appendicitis represents the most common abdominal surgical emergency in this population<sup>19</sup>, even more so when the patient is exposed to external factors of abdominal manipulation, which could delay diagnosis and influence clinical deterioration.

Podany AB. et al. (2017) report that the perforation rate in the pediatric population decreases by increasing age, establishing rates of almost 100% in patients aged one year; 50-69% at the age of 5 years and generally in adolescents is 10-20%<sup>20</sup>. In this study, the most affected age group was adolescents older than 14 years, which contrasts with what is reported in the literature.

One of the factors possibly involved in the increased incidence of complicated appendicitis, rarely considered, are sociocultural beliefs. *Empacho* is a culturally popular disease, not recognized by the medical profession, but by all the popular slang in Latin America and by Hispanic groups in the USA. The symptoms frequently described are gastrointestinal such as abdominal distension, constipation, indigestion, diarr-

hea, vomiting, lethargy, and loss of appetite<sup>21</sup>. Other non-gastrointestinal symptoms described are general malaise, exhaustion, fever, sweating, dry tongue, and whitish or yellowish mucous<sup>10</sup>. In this case series, most patients presented a similar clinical picture with abdominal pain (100%), vomiting (100%), nausea (90%), fever (90%), and loss of appetite (80%).

Acute appendicitis is usually confused, generally by parents and/or caregivers, with an episode of *empacho* due to the similarity of the clinical manifestations popularly described. For this reason, many parents and/or caregivers of patients, decide to go to a family member or therapist commonly known as "*sobador*", who performs natural home treatments and manual procedures, among which is abdominal manipulation.

In this case series, all parents and/or caregivers went to a therapist or "sobador", which is a family member or a private individual, because of a suspected empacho. In a qualitative study conducted by Sandberg JC, et al. (2018), 24 Mexican immigrants were interviewed, and it was established that they perceived physicians as professionals who are dedicated to chronic and acute biomedical diseases, preferred for wounds and many internal pathologies, they also refer that they do not perceive medical personnel as sufficiently effective to treat culturally-rooted diseases, such as empacho<sup>22</sup>.

This study demonstrates the clinical deterioration of the patients, who evolved with generalization and increased intensity of abdominal pain. In some cases, the development of diarrhea after the "soba" led to suspect gastroenteritis, the most common misdiagnosis in patients with appendicitis, where it may be present in 33-41%<sup>23</sup>.

The conventional pathophysiologic model of acute appendicitis is based on a relationship between time

No. Patient	Sex	Age	Origin	Parent's academic level
1	Feminine	16 years	Rural	Elementary
2	Male	14 years	Urban	College
3	Feminine	6 years	Urban	Illiterate
4	Male	11 years	Rural	Elementary
5	Male	14 years	Urban	Elementary
6	Male	14 years	Urban	Elementary
7	Feminine	14 years	Urban	Elementary
8	Male	15 years	Rural	College
9	Male	12 years	Rural	Elementary
10	Feminine	15 years	Rural	College

No. Patient	Initial symptom	Time between initial symptom and hospitalization	Pain intensity <sup>a</sup> before manipulation	Pain intensity <sup>a</sup> after manipulation	Abdominal pain generalization	Leukocytes on admission Cell/mm <sup>3</sup>	Intraoperative findings	Inpatient stay (days
1	Pain	96 h	5	10	+	10-15,000	Intestinal perforation	7-14 days
2	Pain	36 h	5	8	+	< 10,000	Intestinal perforation	3-7 days
3	Pain	72 h	5	10	-	10-15,000	Intra-abdominal abscess	3-7 days
4	Pain	24 h	8	8	-	15-20,000	Gangrenous appendicitis	3-7 days
5	Pain	48 h	5	5	+	10-15,000	Intestinal perforation	3-7 days
6	Pain	72 h	5	10	+	> 20,000	Appendiceal adhesion	3-7 days
7	Pain	72 h	5	7	+	> 20,000	Intestinal perforation, appendiceal adhesion, intra- abdominal abscess	> 14 day
8	Fever	24 h	8	10	+	15-20,000	Appendiceal adhesion	3-7 days
9	Pain	48 h	5	5	-	< 10,000	Intra-abdominal abscess	7-14 day
10	Pain	72 h	6	8	-	> 20,000	Intestinal perforation	7-14 day

Table 3. Previous and subsequent clinical manifestations of the 10 patients with complicated acute appendicitis with abdominal manipulation history

Clinical manifestations	Before abdominal manipulation <sup>a</sup>	After abdominal manipulation <sup>a</sup>	
Pain intensity, median	5 [IQR, 5.0-6.5]	8 [IQR, 6.5-10]	
Mild	0	1	
Moderate	8	2	
Severe	2	7	
Generalized pain			
Yes	1	6	
No	9	4	
Symptomatology			
Nauseas	9	9	
Vomiting	10	10	
Loss of appetite	8	8	
Diarrhea	0	4	
Constipation	0	4	
Fever	9	9	
Dunphy's sign	8	8	
Abdominal hypersensitivity	7	9	

and disease progression. Delays can occur anywhere along the pathway from symptom onset to presentation, evaluation, and treatment<sup>24</sup>.

A study by Marmo AS et al. (2014) and by Amin P et al. (2012), mentions that the prevalence of perforation is 7% when symptoms are present in less than 24 hours, and increases exponentially to 98% when symptoms are present more than 48 hours<sup>25,26</sup>. The findings in this study, regarding the time of evolution of the symptoms onset and hospitalization, were greater than 48 hours in eight of the cases, agreeing with the aforementioned. Two of the cases had a shorter evolution time, so external factors that could drastically modify the course of the disease could be considered.

In this study, the median hospital stay of the ten patients ranged from 7 to 14 days, agreeing with the study by Chen CL. et al., (2017) conducted to identify risk factors for prolonged hospitalization in pediatric patients with appendicitis, who did not initially undergo surgical treatment, in which the median hospital stay was 14 days<sup>27</sup>.

Delay in diagnosis remains one of the main risk factors for complicated acute appendicitis. In a multicenter study, Pedziwiatr M. et al. (2019) demonstrated that a prolongation between the onset of symptoms and hospital care is a risk factor for complicated acute appendicitis. Likewise, in a cohort study of 115 patients by Khan J. et al. (2018), the different causes of delay in the diagnosis of acute appendicitis were identified, finding that the major contributing factor was the mismanagement and diagnosis by a witchdoctor in 32.17% and by doctors in 26%<sup>28</sup>.

Similarly, we should consider that the different maneuvers performed by the therapist could be one of the

factors contributing to the perforation of the appendix, due to the probable increase in intra-abdominal pressure. Similar to that proposed in the study by Zvizdic Z. et al. (2020) in a case of perforated appendicitis secondary to blunt abdominal trauma, which establishes that a sudden increase in intra-abdominal pressure can lead to acute appendicitis<sup>29</sup>. In this case series, which presents a picture of acute obstructive appendicitis already established, the increased intra-abdominal pressure could influence vascular collapse and increased intracecal pressure, resulting in perforation of the appendix.

The high incidence of complicated appendicitis, when there is history of abdominal manipulation, popularly performed to relieve and/or cure *empacho*, should be studied in greater depth and rigor, with a larger population of patients, to establish the possible causal relationship between these two variables.

#### Conclusion

Sociocultural beliefs, such as *empacho*, have an important role in the evolution and prognosis of the patient with acute appendicitis. The *empacho*, and its associated abdominal manipulation as empirical treatment, influence the clinical deterioration of the patient, increasing the intensity and generalization of abdominal pain, thus delaying the diagnosis and adequate management of appendicitis which would cause different complications.

It is recommended that community medical personnel raise awareness among the general population about the acute abdomen and, therefore, prevent the popular belief in *empacho*, avoid its practice, and encourage timely consultation.

# **Ethical Responsibilities**

Human Beings and animals protection: Disclosure the authors state that the procedures were followed according to the Declaration of Helsinki and the World Medical Association regarding human experimentation developed for the medical community.

**Data confidentiality:** The authors state that they have followed the protocols of their Center and Local regulations on the publication of patient data.

Rights to privacy and informed consent: The authors have obtained the informed consent of the patients and/or subjects referred to in the article. This document is in the possession of the correspondence author

## **Conflicts of Interest**

Authors declare no conflict of interest regarding the present study.

#### **Financial Disclosure**

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