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Chronic Recurrent Appendicitis in Children: An Insidious and Neglected Cause of Surgical Abdomen

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Abstract: In addition to the familiar acute inflammation of the appendix, chronic inflammation has long been a controversial disease entity. However, some recent reports announced that chronic recurrent appendicitis is by no means an established disease of the appendix. Therefore, we reviewed our appendectomies in children to determine the underestimated cases of chronic recurrent appendicitis and discuss this disease entity in children.

The records of children who had undergone appendectomy upon initial clinical diagnosis of appendicitis were reviewed for a period of 42 months. Incidental appendectomies were excluded. The diagnoses of acute appendicitis, negative explorations and chronic recurrent appendicitis were made on clinical and histopathological grounds.

Of 72 children who underwent appendectomy, three cases showed chronic inflammation of the appendix, five cases showed normal appendix and the remaining were of acute inflammation histopathologically. Three girls of eight, 10 and 12 years old also had clinical presentation of chronic recurrent appendicitis with previous similar attacks of right lower abdominal pain and vomiting and showed relief of these symptoms in the postoperative follow-up.

For children with previous similar attacks of right lower abdominal pain and vomiting chronic recurrent appendicitis should be considered in differential diagnosis after a thorough clinical examination.

Key Words: Appendicitis, Chronic Recurrent; Abdomen, Acute; Children

Introduction

Although acute appendicitis is the most common disease of the appendix, chronic recurrent appendicitis is a previously condemned but existing disease of the appendix (1-9). Many reviews and case reports of chronic recurrent appendicitis in children and adults have also featured in non-English literature (10-12). Because of the general belief and attitude that seeks to avoid unnecessary operations shonly abdominal pain exist, especially when the signs and symptoms are atypical, many children with chronic recurrent appendicitis suffer a similar attack of pain, vomiting and other symptoms.

The present study aims to present our experience with chronic recurrent appendicitis in children and discuss this entity in the light of the literature.

Materials and Methods

The clinical and histopathological records of children who underwent appendectomy following suspicion of appendicitis at the Department of Pediatric Surgery between 15 June 1998 and 15 December 2001 were reviewed. Incidental appendectomies were not taken into account. Cases of acute and chronic inflammation together with negative explorations were recorded. Diagnosis of chronic recurrent appendicitis was made on the basis of a history of similar, recurrent attacks of right lower abdominal quadrant pain leading to appendectomy and histopathological diagnosis of chronic inflammation of the appendix and relief of signs and symptoms after appendectomy. Chronic inflammation was diagnosed when lymphocytes and eosinophils were present within the appendiceal wall with associated fibrosis in the histopathological examination with hematoxylin and eosin (1,2,13-15).

Results

A total of 72 children underwent appendectomy from 15 June 1998 to 15 December 2001. There were 43 boys and 29 girls. The male to female ratio was 1,5. The median and mean ± standard deviation of the ages of all patients were 7 and 7,2 ± 3,8 years respectively.

Histopathologically, five cases of normal appendixes and three cases of chronic appendiceal inflammation as described above were found, while the remaining 64 cases were of acute inflammation. Table 1 shows the descriptive analysis and the diagnoses of the patients and Table 2 summarizes the properties of three cases of chronic recurrent appendicitis. The histopathological examinations of 12-, 8- and 10- year-old girls confirmatory for chronic recurrent appendicitis are also shown in Figures 1-3 respectively.

Table 1. The descriptive analysis and histopathological results of patients who underwent appendectomy.

	BOYS	GIRLS	TOTAL
No. of Cases	43	29	72
Age range	2-12	4-15	2-15
Mean age ± SD	7.0±3.5	8.8±3.6	7.2±3.8
Median age	7	8	7
Chronic infl.	-	3	3
Normal	1	4	5
Acute infl.	42	22	64

(Ages were given in years; No, = number, SD = standard deviation; infl. = inflammation)

Discussion

The existence of chronic recurrent appendicitis has long been controversial and has recently been reconsidered and re-addressed (1-8). While acute appendicitis presents with a more typical sequence of signs and symptoms, the presentation of chronic recurrent appendicitis is more subtle. After early attempts to describe chronic and recurrent appendicitis in the last decades of the 19th century, the term of chronic recurrent appendicitis was then condemned for some time (1,2,16,17). However, recent reports from large series of patients have re-established the term and concept of *chronic recurrent appendicitis* (1-4,7).

The diagnostic criteria for chronic recurrent appendicitis are a history of similar, recurrent attacks of right lower quadrant pain leading to appendectomy, a histopathological diagnosis of chronic inflammation of the appendix and relief of symptoms after appendectomy (1,2,13-15). If acute inflammation is presented with similar criteria, it is termed recurrent appendicitis only. Chronic recurrent appendicitis comprises about 1,5 to 10% of all appendix inflammations (1-4). However, the precise disease mechanism of chronic recurrent

Table 2. The properties of three cases of chronic recurrent appendicitis.

	PATIENT 1	PATIENT 2	PATIENT 3
Age (years)	12	8	10
Sex	girl	girl	girl
Clinical presentation and duration of symptom	RLQ pain, bilious vomiting for 3 days	RLQ pain for 2 days	RLQ pain, bilious vomiting for one week
Previous similar attack(s)	twice within last year	once six months prior	once three months prior
Physical examination	RLQ tenderness	RLQ tenderness	RLQ tenderness
Fever	absent	absent	absent
White blood cell count/mL	7900	8800	11,200
Hemoglobin value (g/dL)	15	12.8	13.4
Urinalysis	normal	normal	normal
Fibrinogen (mg/dL)	227	-	258
C-reactive protein (mg/L)	< 3.6	< 3.6	< 3.6
Plain abdominal X-ray	nonspecific	nonspecific	local ileus in RLQ
Abdominopelvic USG and color Doppler USG for ovaries	right adnexial cyst of Morgagni of 3 cm diam	normal	normal
Laporotomy findings	no free fluid, ovaries normal, Morgagni cyst excised, appendix grayish and thickened	no free fluid, ovaries normal, appendix thickened with local adhesion	no free fluid, ovaries normal, appendix tense and thickened with increased vascularity in the wall
Histopathological examination of the appendix	Lypmphocyte infiltration and fibrosis	Lypmphocyte infiltration and fibrosis	Lypmphocyte infiltration and fibrosis
Postoperative complaints	complete relief	complete relief	complete relief

(RLQ = right lower quadrant, USG = ultrasonography, diam = diameter)

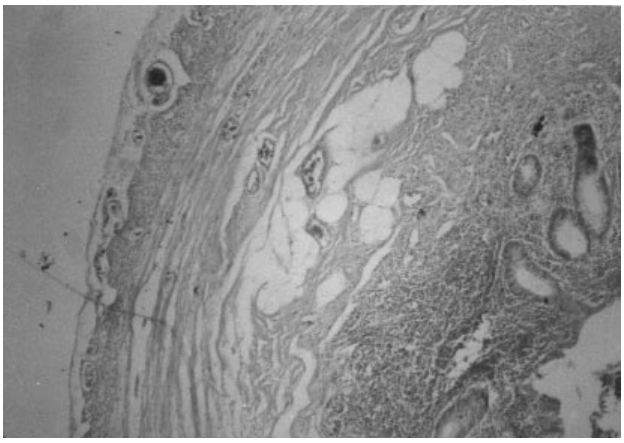

Figure 1. Prominent fibrosis and fatty infiltration in the wall of the appendix of Case 1 (hematoxylin-eosin, x 50).

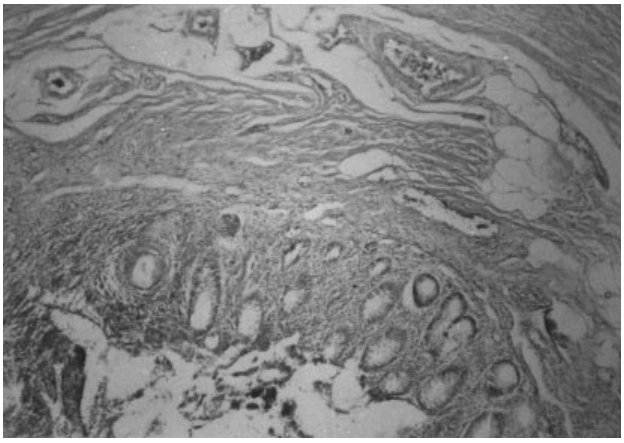

Figure 2. Photomicrograph of the appendix of Case 2 showing the dispersed submucosal lymph follicles, fibrosis in the muscular layer and fatty infiltration (hematoxylin-eosin, x 50).

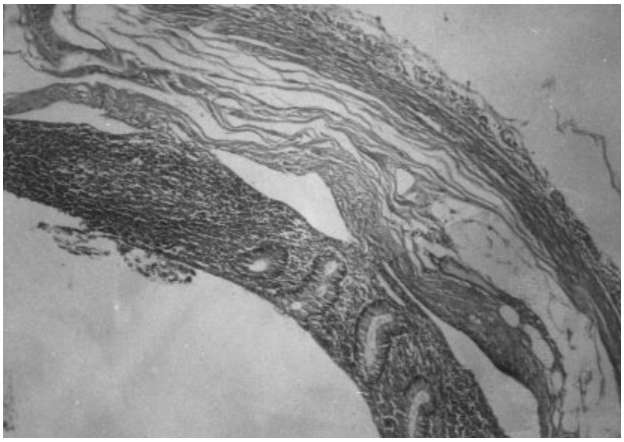

Figure 3. Eroded mucosal surface, dispersed submucosal lymph follicles, fibrosis in the muscular layer and fatty infiltration of the appendiceal wall of Case 3 (hematoxylin-eosin, x 50).

appendicitis is obscure. Besides the various inflammatory reactions, changes in neuroendocrine cells and formation of traumatic neuromas were held responsible for the disease (18,19).

In children, a recent clinicopathological study showed chronic active inflammation with increased numbers of immunocompetent cells, mostly T lymphocytes, subsequent scarring and an increase in the number of neural cells (1). Appendixes in chronic appendicitis cases harbor a chronic inflammatory reaction of an unknown etiology mediated by T lymphocytes and increased lymphoid tissue with concomitant hyperplasia of germinal centers indicating a simultaneous stimulation of B cell mediated immune response (1). Also, appendices from patients with prolonged clinical symptoms defined as chronic appendicitis were shown to have vascular cell adhesion molecule-1 (VCAM-1) expression (20). Panneuronal marker protein gene product 9.5 (PGP9.5) was found to be increased in chronic appendicitis as a neuronal factor in the pathophysiology of the disease and pain symptoms (21).

When reviewing our patients who had undergone appendectomy, the rates of chronic recurrent appendicitis and negative explorations were 4.1% and 6.9% respectively, which are comparable to those in the literature (1-4). However, we have not encountered recurrent acute appendicitis clinically, as we believe that similar attacks of symptoms and signs in acute appendicitis cases may have been neglected or omitted in the past history of the records. In the differential diagnosis of appendicitis, acute or chronic, barium enema examination and computerized tomography might have been helpful in our cases (2,7,9). On the other hand, both techniques have their own handicaps (7,8).

In this study, all patients with chronic recurrent appendicitis were female, although in the literature there is no such sex predilection. While the reason for this female sex predilection in our series may depend on the size of the patient population, it may also result from the developmental nature of adolescent and pre-adolescent girls, which could be investigated in prospective studies. Meanwhile, attention should be paid to pathologies related to adnexa when the underlying etiology of abdominal pain is explored because of the nature of the problems of this sex, especially at preadolescent and adolescent ages. However, this must not mislead or mask the differential diagnosis of chronic recurrent diagnosis.

We should also emphasize that the reluctance to operate on chronic recurrent appendicitis because of the views that disfavor this term may needlessly delay the surgical treatment and prevent relief from a surgically curable problem in children with chronic recurrent appendicitis. Therefore, upon suspicion of chronic recurrent appendicitis in a patient with right lower quadrant pain and tenderness with previous similar attacks, the treatment of choice must be surgical exploration and appendectomy.

In conclusion, chronic recurrent appendicitis should be considered in differential diagnosis in the evaluation of a child with abdominal pain. A history of prior similar episodes of pain, especially in the right lower quadrant with a tenderness on palpation where other diseases are excluded, should favor the possibility of chronic recurrent appendicitis and prompt surgical exploration. Furthermore, special care must be given in cases of the female sex in the differential diagnosis of acute abdominal pain.

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