Case report

A case report on recurrent appendicitis: An often forgotten and atypical cause of recurrent abdominal pain

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\section{Introduction}

Appendicitis is the most common life threatening abdominal surgical emergency globally \cite{1} that requires prompt intervention \cite{2}. Physicians from a wide range of medical specialties including internal medicine and pediatrics, as well as surgeons encounter patients with this condition in their daily practice \cite{3}. Although acute appendicitis is the most common form of appendical illness, the existence of recurrent and chronic types of appendicitis is well recognized \cite{4}. The occurrence of chronic (continuous right lower quadrant (RLQ) abdominal pain for more than 3 weeks) and recurrent appendicitis (serial episodes of similar RLQ abdominal pain) is now widely documented in many medical literature \cite{5}. It is indicated that recurrent appendicitis and chronic appendicitis cases occur in 10\% and 1\% of patients, respectively \cite{6}.

Acute appendicitis is caused by the obstruction of the appendicular lumen, leading to rapid bacterial multiplication, inflammation, ischemia, distension of the appendix and ultimately perforation \cite{7} and the risk of perforation increases by 5\% in every subsequent 12 hours period after 36 hours have passed since the onset of symptoms \cite{8}. This suggests the need for expedient diagnosis and treatment. Mortalities associated with appendicitis are associated with perforation \cite{9} and subsequent progression in to systemic infections. Small number of patients present with recurrent or chronic episodes of right lower quadrant abdominal pain with no associated inflammatory changes \cite{10}. These patients are often difficult to diagnose and treat \cite{11}. The exact cause of pain in such patients is yet unknown \cite{12}. Herein, we described a case report of a male patient with recurring right lower quadrant abdominal pain that he got complete relief of the symptoms of pain after he underwent appendectomy. The case was atypical form of appendicitis in that the patient had very mild tenderness during physical examination with no signs of peritonitis and Rovsing’s sign. A unique aspect about the case is that it had an extended history with 18 years of right flank pains with an extended duration should suspect for recurrent appendicitis and carefully analyze patient history as well as line up their diagnosis.

\section{Discussion}

Recurrent appendicitis can be missed or delayed secondary to atypical presentation or prior treatment with antibiotics, which may lead to resolution of the infection. Missed diagnosis can lead to serious complications such as perforation, abscess formation and peritonitis. Medical practitioners who encounter patients with recurrent right flank pains with an extended duration should suspect for recurrent appendicitis and as well as early medical interventions.

\section{Conclusion}

Therefore, recurrent appendicitis should be considered as differential diagnosis in patients with recurring right lower quadrant abdominal pain and mild/or no tenderness.
2. Presentation of case

A 29-year-old male patient was admitted in Axum Saint Mary Hospital with the complaint of 18 years history of recurrent right lower quadrant (RLQ) abdominal pain associated with constipation, anorexia, mild fever, chills and malaise. The patient was working as lecturer at Aksum University. Ethnically, he belongs to the people of Tigray, Ethiopia. The pains have been resolving spontaneously and sometimes as a result of indiscriminate usage of antibiotics such as tetracycline, doxycycline and amoxicillin when the episodes begin. The patient has never been diagnosed acutely for two reasons: First, the episodes he faced were short in duration (usually less than 6 hours). Secondly, almost all of the episodes appear during the night time, where moving to health care centers might be difficult for him. Moreover, he always used to ignore the case because it developed progressively and had long history. The patient also complained that he has faced episodes once in every 2.5–3 months on average and he stays asymptomatic between the episodes. He also added that he always get warning symptoms of mild and progressive nature of pain, up to 1–2 hours prior to the episodes. The patient also noted that he had once scanned by computed tomography (CT-scan) after 8 hours of acute abdominal pain, where the intensity of the pain has been reduced to some extent. But he was informed that his appendix was normal. In January 2016 the pain began progressively as usual but its intensity increased as time advanced. Once the pain became unbearable, he came to emergency section of Saint Mary hospital with his colleague in search of better medical service. Saint Mary is a public hospital located in central Tigray, Ethiopia.

Up on physical examination, mild tenderness was observed during palpation of the right flank. Evaluation of vital signs indicate 122/65 mmHg of blood pressure, 60 beats/min of pulse rate, 20 breaths/min of respiratory rate and 36°C body temperature. Hematological evaluation, fecal examination and urinalysis results did not suggest any clue about his recurrent abdominal pain. Moreover, ultrasound and X-ray images of the digestive and urinary systems did not collect valuable information. Careful re-evaluation of the patient's history and the exact location of pain (right flank) pointed our diagnosis to the possibility of recurrent appendicitis. After the summary of examination results, the possibility of the case to be recurrent appendicitis was explained to him. Since appendectomy is the only treatment option, we advised him to undertake advanced diagnostic techniques such as CT scan and visit referral hospitals with such facilities as early as possible when the symptoms of the disease begin. However, the patient claimed that he was exhausted of the case and he was not interested to do so. Rather, he insisted to get his appendix removed.

Evaluation of patient history indicated that he was free from any chronic diseases and psychosis, which require regular administration of drugs and he had no history of adverse drug reaction. Besides, he had no history of previous operation. He was also free from alcoholic and smoking addictions, gastrointestinal infection and appendical trauma. Analysis of patient history also revealed that appendicitis has never been identified in his family members. Generally, the patient was in good state of health and his BMI was calculated to be 24 Kg/m².

The patient was prepared both physically and psychologically for surgery. He also signed an informed consent form acknowledging that he is aware of risks and complications he may encounter. He was informed that he would be receiving general anesthesia and the surgeon explained to him about the operation. Surgical procedures are not performed unless the consent form is signed by the patient. On the suspicious diagnosis of a recurrent appendicitis, we anesthetized the patient under general anesthesia and performed an open appendectomy. The surgical procedure was performed by team of eight professionals including major surgeon, trainees, anesthetists and nurses.

The total time taken for the operation was 30 minutes and there was loss of blood during the operation. Gross examination of external surface of the excised the appendix indicated that it was inflamed and dilated, some parts were ulcerated, scarified and with significant strands of periappendical fat (Fig. 1). Besides, the wall of the appendix was thick; its lumen was narrow and filled with purulent material.

Besides to the standard post-operative care, the patient was ordered to take two doses of 500 mg of Cephalexin daily for 10 days per os. At the first post-operative day, the patient began oral alimentation. We followed him in our hospital for two days. On the third postoperative day, he was discharged with full recovery after his wound was checked up. We suggested him to visit us if he encountered strange situations such as wound wetting or severe pains. On the seventh post-operative day, the stiches were removed and his wound was disinfected. Besides, the wound was dry and the patient was generally in a good state of health. It has been almost two years since his appendix was removed and the patient has got complete relief of the symptoms of abdominal pain after appendectomy.

3. Discussion

Appendicitis can occur in all ages of people but higher incidence is reported to occur within the range of ages between 10 and 19 years [14,15]. Estimates indicate that the lifetime risk of development of appendicitis is about 8.6% for males and 6.7% for females of all ages [14]. Although the concept of recurrent appendicitis has remained controversial, its existence has gained medical importance in recent years [6]. The diagnostic criteria for recurrent appendicitis relies on history of similar and recurrent attacks of right lower quadrant abdominal pain, a histopathological confirmation of chronic inflammation of the excised appendix and relief of symptoms after appendectomy [16,17]. The current case complies with the case definition of recurrent appendicitis. Moreover, gross examination of external surface of the excised the appendix indicated that it was inflamed and dilated, its wall was thick and its lumen was filled with purulent material and

Fig. 1. Picture of removed appendix.
narrowed. Some parts of the appendix were also ulcerated, scarified and had significant strands of periappendiceal fat (Fig. 1). These findings were concordant with the work of Chang and Chan [18]. The patient has also got complete relief of symptoms after he underwent appendectomy. However, we could not perform histopathological analysis of the excised appendix as we do not have the facilities in our hospital. CT scan may also be valuable method of diagnosis particularly if performed at early stage of appendicitis with an overall accuracy ranging from 93% to 98% [6]. But CT scan is found only in referral hospitals and people living in remote areas of developing countries usually do not have an access.

Unlike acute appendicitis, recurrent appendicitis is not considered a surgical emergency [19]. Diagnosis can be missed or delayed secondary to atypical presentation or prior treatment with antibiotics, which may lead to resolution of the infection [20]. A missed diagnosis can have serious complications such as perforation, abscess formation and peritonitis [19]. Upon the suspicion of recurrent appendicitis, the treatment of choice must be surgical exploration and appendectomy [2]. Incidental appendectomies are also commonly performed at the time of other abdominal or pelvic surgery to prevent the development of appendicitis [21] since the appendix has no known function in humans. Many surgeons prefer the surgical removal of the inflamed appendix, even if the diagnosis is only suspicious. However, one third of the patients have atypical symptoms confusing the diagnosis [17]. Atypical presentations of appendicitis are often difficult to diagnose and are usually overlooked [11]. Most importantly, abdominal attacks with prolonged history are neglected or misdiagnosed and treated as other ailments, especially in developing countries with low public health infrastructure settings. In such situations, the removal of the appendix is delayed or even not performed [22]. In contrast, earlier diagnosis and operation could prevent the spread of the inflammation and avoid the development of the related complications [17]. Moreover, morbidity associated with appendectomies is generally considered to be negligible [23]. The current case report presents 18 years of undiagnosed recurrent appendicitis. This had the longest duration of history of recurrent appendicitis among the documented such medical case reports. Previous reports indicated that the duration varies from three weeks to seven years [6,24]. It is always important to consider recurrent appendicitis as a differential diagnosis in patients with recurring pain in their right iliac fossa [18] so that patients could either be closely followed up or medical interventions options could be outlined.

4. Conclusion

Appendicitis is the most common life threatening abdominal surgical emergency that we frequently encounter in our hospital. Recurrent appendicitis does not manifest the classical clinical symptoms and signs of acute appendicitis. Abdominal attacks with long history of recurring nature are neglected or misdiagnosed in most cases. Surgeons/medical practitioners who encounter patients with recurring right flank pains with an extended duration should suspect for recurrent appendicitis and carefully analyze patient history as well as line up their diagnosis. Therefore, recurrent appendicitis should be considered as differential diagnosis in patients with recurring right lower quadrant abdominal pain and mild/or no tenderness.

Ethical approval

Proof of ethical clearance and informed consent was obtained from the Institutional Review Board of College of Health Science, Aksum University under the reference number IRB035/2017.

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Author contribution

Mesfin Demeke has developed the concept of the study, identified the case and conceived to convert it into scientific article. Gebreyohans Gebru wrote the paper. Mesfin Demeke and Gebreyohans Gebru reviewed the paper and approved for publication.

Conflicts of interest

The authors declared that they have no conflicts of interest.

Guarantor

Gebreyohans Gebru is the guarantor for this study.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Registration of research studies

Since this is a case report, it will not use human participants as subjects for study. However, we have gone through the formal procedure of acquisition of proof of ethical clearance and informed consent.

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