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SCIENTIFIC REVIEW

Unfinished Business: A Systematic Review of Stump Appendicitis

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Abstract

Background Stump appendicitis is defined as interval inflammation of any residual appendicular tissue, after an appendicectomy. We present a systematic review of case series and case reports on stump appendicitis, emphasising on risk factors, diagnosis and surgical management.

Methods The English literature (1945–2018) was reviewed, using PubMed, Embase and GoogleScholar, combining the terms "appendix", "appendicitis", "stump", "residual", "recurrent" and "retained". In total, 127 studies were included, describing 164 patients (males 59%, mean age 36 ± 17 years).

Results Index surgery was open in 59% and laparoscopic in 38%. It was described as "difficult" or "complicated" in 31%. 20% of patients reported episodes of recurrent abdominal pain during the time interval between index and stump appendicitis (range 2 weeks to 60 years, median 2 years). Right lower quadrant pain was the most frequent complain (88%), leukocytosis was found in 56%, whereas 92% of patients underwent imaging testing, which was diagnostic or highly suspicious in 67.5%. Mean delay between beginning of symptoms and surgery was 2.4 ± 2.3 days. The operative approach was open in 61% and laparoscopic in 35% of cases. The operation was characterised as "difficult" or "complicated" in 45%. In the majority (88%), a completion stump appendicectomy was performed, with 11% requiring more extensive procedures. Mean length of resected stump was 3.1 ± 1.6 cm (range 0.5–10 cm).

Conclusions Stump appendicitis may occur following both open and laparoscopic approach, when the residual stump is > 0.5 cm. Its clinical significance lies in the delayed diagnosis, leading to higher incidence of complications and the need for more extensive surgery.

Introduction

Acute appendicitis carries a lifetime risk of 6–9%, and appendicectomy is still one of the most frequently performed emergency operations worldwide [1–4]. Stump appendicitis is a rare, long-term complication of appendicectomy, described first by Rose in 1945 [5, 6]. Yet, it still remains an underreported clinical entity, and its true prevalence cannot be determined [6, 7].

It is defined as the interval development of obstruction and inflammation of any remaining appendiceal tissue after an appendicectomy [6–9]. Stump appendicitis is usually

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not included in the differential diagnosis of abdominal pain in patients with prior history of appendicectomy. Its clinical significance lies in the delayed diagnosis, leading to higher incidence of perforation, the need for more extensive surgery and increased postoperative morbidity [8].

We systematically reviewed the literature on stump appendicitis and looked into its epidemiology, clinical presentation, diagnosis and treatment, in order to describe the clinical consequences and to determine potential risk factors and prevention strategies.

Materials and methods

We performed a systematic review of the English literature, following the Meta-Analysis Of Observational Studies in Epidemiology (MOOSE) guidelines, in order to identify all studies of patients with stump appendicitis [10]. Literature searches were conducted in PubMed/MEDLINE, Embase and GoogleScholar bibliographic databases, spanning years 1945 to 2018. The keywords "appendix", "appendicitis", "stump", "residual", "remaining", "retained" and "recurrent" were used in all possible combinations. Additionally, the reference lists of all eligible studies were assessed for additional articles.

All study designs were eligible for inclusion in the final analysis. Patient age was not an exclusion criterion, and both adult and paediatric cases were included in the review. Non-English articles and articles without full-text availability were excluded. Moreover, articles reporting non-infective stump complications, like haemorrhage, intestinal obstruction or malignancy, were excluded.

Titles and abstracts of all articles from the initial search were independently screened by two authors, to determine those articles for full-text review. Any discrepancies concerning the evaluation of the studies were arbitrated by all authors. A flow chart of study selection is shown in Fig. 1.

For each eligible study, data were extracted about demographic and clinical characteristics (age, gender, comorbidity, time interval, recurrent episodes of right lower quadrant (RLQ) pain, symptoms), the index appendicectomy (emergency or elective surgery, open or laparoscopic approach, intraoperative findings, appendix ligation method, postoperative complications), laboratory and imaging tests (white blood cell (WBC) count, ultrasound/CT findings) and the surgical operation (time to surgery, emergency or elective surgery, open or laparoscopic approach, intraoperative findings, type of resection, postoperative complications, appendix stump length).

Statistical analysis was performed on SPSS, version 20.0. All data were tabulated, and outcomes were cumulatively analysed. Continuous variables were expressed as mean ± standard deviation, while categorical variables were expressed as frequencies or percentages. A subgroup analysis of males versus females and children versus adults was furthermore conducted. Student's t-test was used for comparison of continuous variables, and Chi-square or Fisher's exact test for categorical variables. Statistical significance was set to $p < 0.05$.

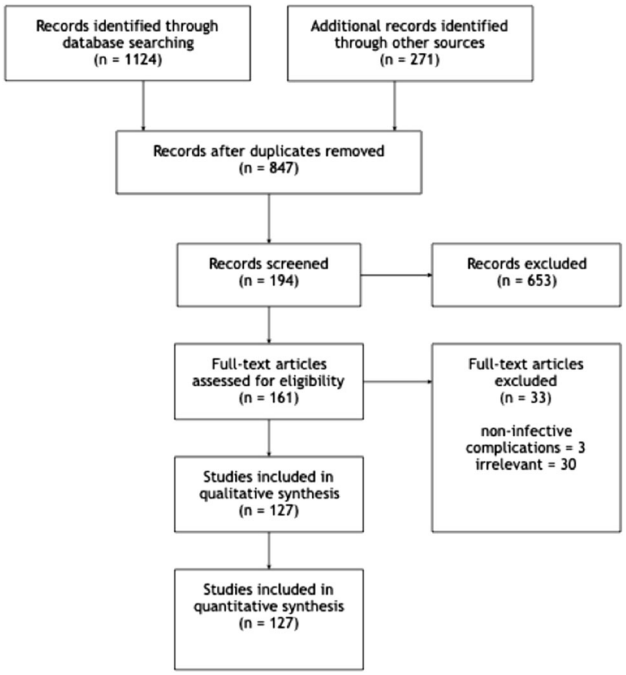

Fig. 1 Flow chart of the search strategy

Results

In total, 127 studies were included in the final analysis, describing 164 patients with stump appendicitis (Online Resource). Of these patients, 97 (59.1%) were male, 60 (36.6%) were female, and 7 (4.3%) did not report gender. The mean age at presentation was 35.8 ± 17.1 years (range 2–75 years) (Fig. 2). Past medical history was explicitly reported in 38 patients. Out of 10 patients with medical comorbidities, 2 were immunosuppressed (one case of lung cancer and one case of presumed Crohn's disease under immunosuppresants).

The index appendicectomy was performed on an emergency basis in 131 cases (79.9%), as elective case in 6 (3.7%), and it was not reported in 27 (16.4%). The operative approach was open in 97 patients (59.2%), laparoscopic in 63 (38.4%) and not reported in 4 (2.4%). Intraoperative findings during the index procedure were described in 84 cases. Of these patients, 26 (30.9%) represented a "difficult" or "complicated" case, either of

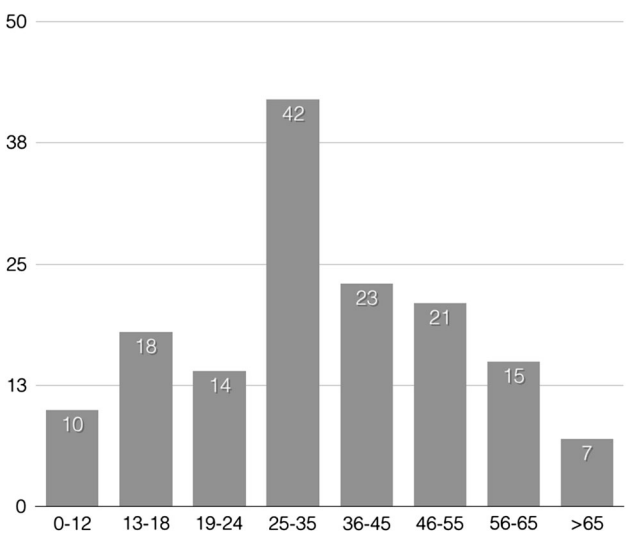

Fig. 2 Age distribution of stump appendicitis patients

gangrenous appendicitis, perforation, abscess or multiple adhesions. Appendix ligation method was reported only in 19 cases (suture ligation in 5, clips in 5, stapler device in 9 cases). Details of the postoperative course were included in 28 cases. Twenty-two patients had an uneventful course, whereas complications were reported in 6 (4 cases of surgical site infection, one intraperitoneal abscess, one admission to the ICU).

The time period between the index appendicectomy and stump appendicitis ranged from 2 weeks to 60 years (median 2 years) (Fig. 3). During this interval, 33 patients (20.1%) reported episodes of recurrent RLQ pain, following the appendicectomy.

As far as the symptomatology of stump appendicitis is concerned, the most frequent complaint was RLQ pain

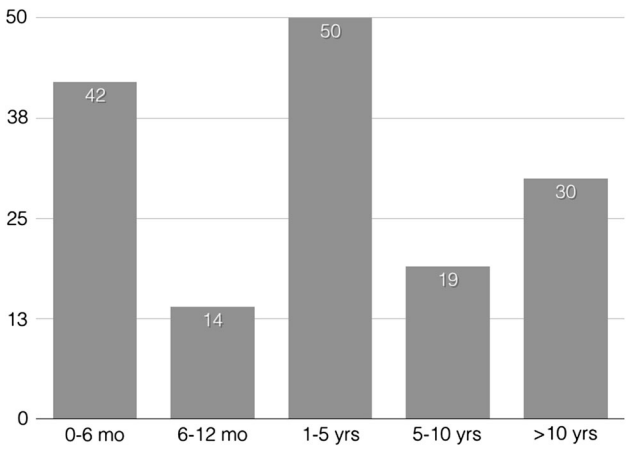

Fig. 3 Time interval between index appendicectomy and stump appendicitis

(144 patients, 87.9%), accompanied by fever in 61 (42.4%) and nausea/vomiting in 39 (27.1%) cases. Seven patients reported non-specific or diffuse abdominal pain (4.3%), 2 patients presented with draining cutaneous fistula (1.2%), and one patient presented with a retroperitoneal abscess (0.6%). Mean WBC count on admission was 14,500 ± 4500 (range 6900–27,000). Leukocytosis was observed in 92 cases (56.1%), normal WBC counts in 31 (18.9%) and was not reported in 41 (25%). In total, 151 (92.1%) patients underwent imaging studies (ultrasound, CT scan or both). CT scans were diagnostic or highly suspicious in 59/78 cases (75.6%), ultrasound in 18/28 (64%), whereas the combination of ultrasound plus CT in 22/32 (69%), $p = 0.47$.

Nine patients (5.5%) were successfully managed conservatively, with antibiotics and deferral of surgery, whereas 155 (94.5%) underwent surgical exploration. The mean delay between the beginning of symptoms and the definitive operative management was 2.4 ± 2.3 days. In 146 cases (94.2%), the second operation was performed on an emergency basis, whereas the remaining 9 cases (5.8%) were operated on electively. The approach at the second operation was open in 95 (61.4%), laparoscopic in 54 (34.8%) and not reported in 6 (3.8%) (Fig. 4). Ten patients required conversion from laparoscopy to open surgery (18.5%).

Intraoperative findings during the second operation were described in 147 patients. Of these cases, 66 (44.9%) were described as "difficult" or "complicated" (gangrenous appendicitis 3, phlegmon 3, cutaneous fistula 3, perforation 37, intra-abdominal abscess 20). In the majority of cases (136/155, 87.7%), a completion stump appendicectomy was performed. Two patients (1.3%) underwent drainage and cecostomy, whereas 17 patients (11%) required more extensive resections (ileocecectomy or right hemicolectomy). The postoperative course was reported in 134 cases, with 12 patients (7.7%) developing complications, mainly surgical site infections and prolonged paralytic ileus.

The mean length of the resected appendiceal stump was 3.1 ± 1.6 cm (range 0.5–10 cm). There was no statistical difference in stump length, when the index operation was either open or laparoscopic (3.02 ± 1.7 vs 3.3 ± 1.3 cm, $p = 0.17$).

Results of the subgroup analysis of males versus females and children versus adults are shown in Table 1. The rate of diagnostic or highly suspicious US/CT scans was comparable between the subgroups, although a delay in surgery was observed in both females (2.9 ± 3.2 vs 2.1 ± 1.6 days, $p = 0.050$) and children (3.2 ± 3.5 vs 2.2 ± 1.9 days, $p = 0.047$), compared to males and adults respectively. However, the rate of difficult or complicated surgery, as well as the length of the appendix stump, did not differ significantly.

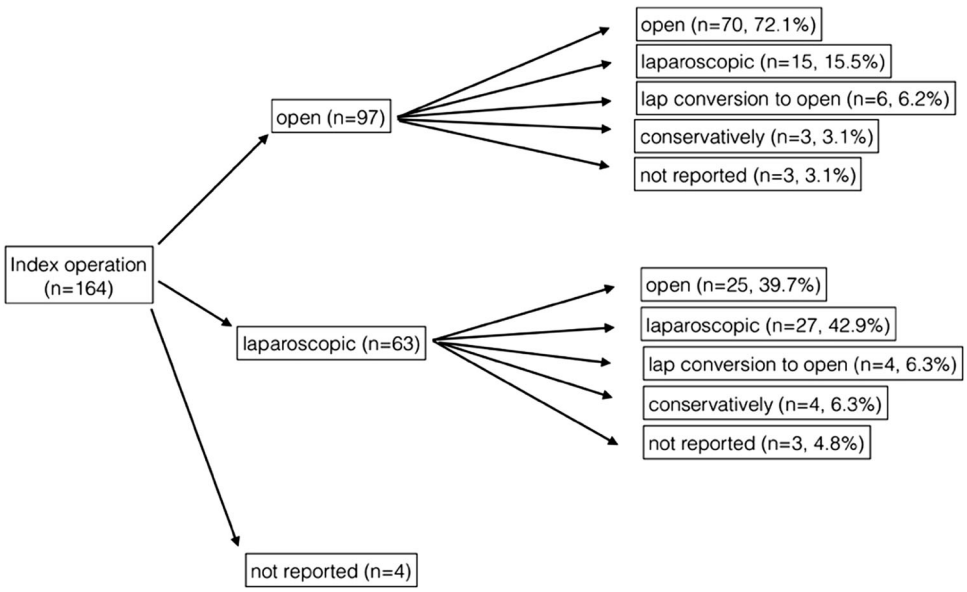

Fig. 4 Stump appendicitis management after initial open or laparoscopic surgery

Table 1 Subgroup analysis of males versus females and children versus adults

	Males	Females	*p* value
N	97	60	
Age (years)	32.3 ± 16.0	41.4 ± 17.2	< 0.001
CT/US diagnostic	63/88 (71.6%)	33/57 (57.9%)	0.088
Delay in surgery (days)	2.1 ± 1.6	2.9 ± 3.2	0.050
Complicated surgery	36/86 (41.9%)	26/55 (47.3%)	0.527
Stump length (cm)	3.3 ± 1.6	2.9 ± 1.6	0.105
	Children	Adults	*p* value
N	28	122	
CT/US diagnostic	21/27 (77.8%)	70/113 (61.9%)	0.121
Delay in surgery (days)	3.2 ± 3.5	2.2 ± 1.9	0.047
Complicated surgery	10/25 (40%)	52/110 (47.3%)	0.510
Stump length (cm)	3.3 ± 1.3	3.1 ± 1.7	0.293

Discussion

The true incidence of stump appendicitis is unknown and difficult to establish. Two large institutional reviews reported rates of 0.06–0.15% [11, 12]. Our systematic review, spanning years 1945 to 2018, describes 164 cases in total.

Clinical Presentation

Stump appendicitis occurs across all age groups, from a few days to several decades after appendicectomy [6, 9]. The prevailing symptom is RLQ pain; however, the classic history of colicky periumbilical pain migrating to the RLQ is rather infrequent. Rarely, it may present as non-specific or atypical abdominal pain, chronic draining sinus or distant abscess. Interestingly, one in five patients reported similar episodes of RLQ pain in the past, usually not requiring medical assistance.

Diagnosis

Unsurprisingly, the clinical suspicion of stump appendicitis in a patient with a history of appendicectomy would be very low in daily practice [6]. Most surgeons and emergency physicians opted for radiological investigation, with ultrasound, CT scan or both, and would not rely solely on the clinical presentation to proceed to surgical intervention.

Sonographic findings are generally non-specific and include fluid in the right iliac fossa, oedema of the cecum or inflammatory changes around the appendiceal stump [8]. In some patients, the thickened, inflamed stump with an appendicolith within its lumen may be visualised [8]. CT findings are also non-specific and include pericecal or right paracolic fluid, thickening of the cecum with fat stranding, phlegmon, abscess or free air in case of perforation [13–15].

Rates of correct diagnosis or high suspicion were comparable among CT, US, and their combination, and also did not differ depending on patient gender or age. It appears that ultrasound still has a role, although CT scans provide more detail and would be preferable in ambiguous cases. As with acute appendicitis, 24–36% of patients had a negative or inconclusive imaging study [16]. Admission and serial clinical reevaluation is therefore a valid option.

Similarly, leukocytosis, a surrogate systemic inflammatory marker, is usually present on admission, although WBC counts may be within normal range in up to 20% of patients [16]. C-reactive protein levels were not reported in most studies.

Treatment

Surgical exploration was the treatment of choice in the majority of cases, with only few patients managed conservatively. The delay in seeking medical assistance and the challenge of establishing an accurate diagnosis may explain the mean time interval of 2.4 days, between the beginning of symptoms and surgery, and the high rate of "difficult" or "complicated" cases (44.9%). This delay in definitive surgical treatment was more significant in females and children, in whom the differential diagnosis of RLQ pain is wider.

Completion stump appendicectomy is generally safe and adequate, when the base of the appendix can be identified. In cases of gangrene, plastron, perforation or inflammatory involvement of the cecum, an ileocecectomy or even right hemicolectomy may be needed. In our review, more extensive procedures were reported in 11% of patients, in contrast to acute appendicitis cases, where this percentage is much lower (around 1% in the POSAW study) [16].

Nevertheless, overall complication rate (7.7%) was comparable to acute appendicectomy morbidity (3–28%) [16]. Moreover, it appears that the laparoscopic approach is technically feasible in experienced hands, even after an initial open procedure, with an acceptable conversion rate of 18.5% (Fig. 4). No cases of stump appendicitis following robotic appendicectomy have been described so far.

Risk factors

The pathogenetic mechanism of stump appendicitis remains largely unclear. Impaction of a fecalith in the stump lumen and poor blood supply to the stump have been implicated.

The major predisposing factor is the length of the residual appendix after ligation. A stump of ≥ 0.5 cm is considered large enough for a fecalith to become impacted and cause obstruction and inflammation. No cases of stump length < 0.5 cm have been described in the literature. In our review, the mean stump length was 3.1 ± 1.6 cm, with a range from 0.5 cm to 10 cm.

Invagination of the appendiceal stump would theoretically prevent complications such as peritoneal contamination, adhesion formation and stump appendicitis. There is no evidence, however, to support inversion versus simple ligation [7, 8, 17]. Ligation method during the index appendicectomy (suture, clips, stapler) was available only in 11.5% of cases and consequently no conclusions can be drawn.

Speculation that stump appendicitis might be more frequent with the laparoscopic approach, due to the narrow, two-dimensional field of vision and the loss of haptic feedback, has not been confirmed [6–9]. Between 1990 and 2000, 35% of stump appendicitis cases followed an initial laparoscopic appendicectomy. Between 2001 and 2018, 42.7% of cases occurred after a laparoscopic procedure. As the penetrance of laparoscopy increases over the years, this number is expected to increase proportionately. However, this is by no means evidence that laparoscopy per se is a risk factor for stump appendicitis. Moreover, there was no difference in stump length, depending on the operative approach at the index appendicectomy (open 3.02 ± 1.7 cm vs laparoscopic 3.3 ± 1.3 cm, $p = 0.17$).

Regardless of the method to remove the appendix and treat the stump, it is imperative to trace the taeniae coli to the base of the cecum, in order to recognise the appendiceal-cecal junction. Subramaniam et al. propose a laparoscopic appendicitis "critical view of safety", equivalent to that in cholecystectomy, where the appendix, the terminal ileum and the taenia libera form a triangle [8]. Failure to identify the appendiceal-cecal junction should be an indication for conversion to an open procedure [6–9].

Inadequate visualisation of the appendiceal base is more likely to occur in cases of multiple adhesions, retrocecal or subserosal appendix and particularly in severe inflammation [6]. In large observational studies, rates of complicated acute appendicitis (gangrene, phlegmon, abscess, perforation) range between 25 and 45% [4, 16, 18–20]. In accordance with the literature, a history of complicated index appendicitis was noted in 31% of patients. Although a difficult appendicitis may contribute to an incomplete

resection, the majority of cases did not fall into this category [8].

Given the rarity of stump appendicitis and the long-term follow-up required, a prospective observational study appears rather impractical. Systematic reviews of case reports and case series remain therefore the highest available level of evidence to date. Another limitation is the English language restriction in our literature search. Moreover, our systematic review spans more than 7 decades, implying that the study sample could be at risk of heterogeneity, in terms of diagnostic modalities (US and CT scans) and surgical procedures (laparoscopy, stapling devices, clip appliers). Finally, the pending issue of laparoscopy being a risk factor cannot be definitely addressed, since the true incidence of stump appendicitis with the open and laparoscopic approaches cannot be calculated.

Conclusion

Stump appendicitis is the interval obstruction and inflammation of any remaining appendiceal tissue following an appendicectomy. It is a rare and underreported late complication, observed following both open and laparoscopic approaches. A high index of clinical suspicion, along with appropriate imaging studies, may establish a timely diagnosis. Completion stump appendicectomy is usually feasible; however, stump gangrene, perforation or abscess may result in more extensive resections. The major risk factor is failure to identify the appendiceal base during appendicectomy and a residual appendix stump of > 0.5 cm.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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