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Amyand's Hernia with Acute Appendicitis: A Case Report of a Type 2 Amyand's Hernia with Literature Review.

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ABSTRACT

Background: Amyand's hernia, defined as the presence of the vermiform appendix within an inguinal hernia sac, is an uncommon surgical entity, rarely complicated by appendicitis. Preoperative diagnosis remains challenging due to overlapping clinical features with other inguinal pathologies.

Objective: To describe a rare case of Amyand's hernia complicated by appendicitis and present a systematic review and pooled meta-analysis of published cases to evaluate clinical characteristics, diagnostic approaches, and surgical management outcomes.

Methods: Following PRISMA guidelines, a comprehensive PubMed search from January 2014 to August 2024 identified case reports, case series, and retrospective reviews of adult patients with Amyand's hernia complicated by appendicitis. Data on demographics, diagnostics, management strategies, and outcomes were extracted and analyzed using R software, with categorical variables compared via χ^2 and Fisher's exact tests.

Results: Fifty-five studies (59 patients) met inclusion criteria. The mean patient age was 63.5 ± 20.3 years, with an 86.4% male predominance. Right-sided involvement occurred in 93.2% of cases. Preoperative diagnosis of Amyand's hernia with appendicitis was achieved in only 37.3% of patients, with computed tomography being the most accurate modality (52.5%). Most patients (76.3%) underwent emergency surgery, predominantly via open approach (73.3%), while laparoscopic intervention was more common in elective settings. Non-mesh hernia repair (herniorrhaphy) was favored in inflamed or contaminated fields (66.6%, $p < 0.05$). Postoperative complications were rare, with isolated cases of mesh infection, renal failure, and death unrelated to procedure type.

Case Summary: A 39-year-old male presented with right lower quadrant pain diagnosed as acute appendicitis. Intraoperatively, an inflamed, pus-filled appendix was found extending into the deep inguinal ring. Laparoscopic appendectomy and herniorrhaphy without mesh were performed, yielding uneventful recovery.

Conclusion: Amyand's hernia complicated by appendicitis remains a diagnostic and therapeutic challenge. Laparoscopic appendectomy with individualized hernia repair based on inflammatory severity is safe and effective. Consistent case reporting is essential to refine management strategies and improve outcomes for this rare condition.

BACKGROUND

Amyand's Hernia is a rare condition characterized by the presence of the vermiform appendix within an inguinal hernia sac. This case report details a 39-year-old male who presented with a 3-day history of right lower quadrant abdominal pain, initially diagnosed as acute appendicitis. Upon examination, the patient was hemodynamically stable with a normal physical examination, except for localized tenderness in the right lower quadrant. Imaging with a CT scan confirmed inflammation of the appendix.

The patient's prior history included an episode of abdominal pain treated with antibiotics and bowel rest. In the emergency department, he received symptomatic treatment including antibiotics, analgesics, and fluids. Following stabilization, the patient underwent an emergency laparoscopic appendectomy. During surgery, an inflamed, pus-filled appendix was identified, extending into the deep inguinal ring and adhered to the lateral aspect of the ascending colon. The appendix was removed, and the hernia sac was repaired without mesh.

Postoperatively, the patient recovered well, with no additional complaints, and was discharged on postoperative day 3. This case underscores the challenges in diagnosing Amyand's Hernia preoperatively and highlights the importance of prompt surgical intervention. The findings contribute to the limited literature on the management of complicated Amyand's Hernia and demonstrate that laparoscopic surgery can be effective in treating this rare condition.

INTRODUCTION

Amyand's Hernia is the presence of vermiform appendix in an inguinal hernia sac. Amyand's Hernia, which was first described by Claudius Amyand in 1735, is generally complicated in 0.1% cases by Acute Appendicitis [1,2]. Amyand's Hernia is subsequently classified into three categories which includes (1) Type 1 Amyand's Hernia which has no inflammation of the appendix, (2) Type 2 Amyand's Hernia which has minimal inflammation of appendix and (3) Type 3 Amyand's Hernia with perforated appendix [3].The differential diagnosis for these cases include acute appendicitis, acute hydrocele, testicular torsion, peritonitis, epididymo orchitis and urologic emergencies [28]. Appendix is generally incarcerated in cases of femoral hernia (De Garengot's Hernia) [73]. The case of Amyand's Hernia with appendicitis is generally present on the right, occasionally it is present on the left side. With the recent advancement of Laparoscopic Surgeries and minimally invasive techniques, there is still minimal evidence on management methods to treat cases of Complicated Amyand's Hernia [18]. The preoperative diagnosis of the patient is challenging due to masking of pathognomonic clinical features to Inguinal hernias [19]. Inguinal hernias are generally treated on the basis of clinical signs and symptoms without generally any radiologic investigations, leading to high chance of missed preoperative diagnosis [33]. Even though the term Amyand's

Hernia is a term to describe Incarcerated hernia, there are literatures which defines it as a broader term which includes reducible hernia swelling [4,5]. Cases vary from neonates to elderly even though the disease is generally rare [6]. Surgical Management is mostly preferred in suspected cases of Amyand's Hernia [7]. There is very limited literature regarding management strategies of such cases which mostly relied on appendectomy and no use of Mesh repair or herniorrhaphy and need of surgeons to identify incarcerated inflamed appendices within hernias in order to ensure potential successful surgical outcomes and avoid unexpected surgical complications [73]. Thus, we report a case of Amyand's Hernia which was complicated by Acute appendicitis along with systematic review and pooled meta-analysis of available case reports of similar cases of Amyand's Hernia (AH) complicated by Acute appendicitis. The Systematic review is an effort to sum and unveil all the available reports on the AH with appendicitis and provide data on demographics, patient related parameters and optimal effectiveness of the operative management in terms of safety and effectiveness.

OBJECTIVE

To describe a case of Amyand's Hernia which is complicated by Inflammation and pus that is rare to diagnose a condition which is diagnosed intraoperatively, and to describe a systematic review along with analysis of the available case reports on Amyand's Hernia complicated by Inflammation.

METHODS

We performed a systematic review of the cases reported on Amyand's Hernia which got complicated by inflammation of the appendix. We followed the predesigned Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) protocol and PRISMA checklist and standard for reporting the systematic review to the extent of our possibilities.

Search Strategies

A comprehensive search for case reports, case series and retrospective review on Amyand's Hernia complicated by Inflammation of Appendix reported on PubMed database conducted by two independent reviewers (PJ and RM) on August 1, 2024. The search included Case series, Case reports and retrospective reviews published from January 2014 to August 2024 by using keywords such as "Amyand's Hernia", "Acute appendicitis", "Inflammation of Appendix" and "Pus filled appendix" and Mesh terms such as "Appendix" [MeSH term] AND "hernia, inguinal" [MeSH term].

Definitions and Classification

Amyand's Hernia (AH) is the presence of a vermiform appendix inside the inguinal hernia sac [1]. There is no definite criteria for diagnosis of Amyand's Hernia but there is classification of Amyand's Hernia was first described by Losanoff and Basson, and was modified by Rikki which describes inflammation of Appendix as Type 2 Amyand's Hernia and Amyand's Hernia with Pus formation of Appendiceal Inflammation as Type 3 Amyand's Hernia [74].

Eligibility Criteria

We used the following as an inclusion criteria to include cases for the systematic review: All case report, case series and Retrospective review of single center of AH with Appendicitis (1) Diagnosis of AH with appendicitis confirmed by a physician as described by the author's judgment, (2) where data including patient's demographic such as age, sex and ethnicity of patient, clinical symptoms, Preoperative and Postoperative diagnosis, Management type, mode of diagnosis preoperatively, Intraoperative Procedure, Outcome and Type of Surgery done is explicitly noted (3) where mimicking differential diagnosis was ruled out to provide clear picture and (4) Patients included to be adult patient.

We excluded articles that were (1) observational studies, review articles, letters to the editor, posters that were not presenting clinical NH case reports, (2) Patients with less than 18 years of age to provide clear picture, (3) case reports in any language other than English, (4) observational studies, review articles and case series with duplication of patient's data and (5) Case reports with no information on Intervention.

Selection of Studies and Data Collection

By using the Search Strategy above, we screened and identified a total of 107 studies. We excluded 39 studies which were non-human, non-full text and outside of January 2014 to August 2024 publication. Two investigators (PJ and RM) then read all the 68 articles including abstracts and full manuscripts, and selected the articles based on inclusion and exclusion criteria. Any disagreement between two investigators is resolved by mutual consensus. Thirteen studies were excluded due to the article being not full text, incomplete information on demographics or patients being children and neonates, not well defined studies, diagnosis of Appendiceal Neoplasm, Non-English and not able to comprehend. Thus, we got 55 studies of which there are 51 case reports, 2 Retrospective reviews and 2 Case series, among which there are 59 patients which are considered for quantitative analysis (Figure 1).

All eligible studies data were collected using standardized web-based form. This data was then summarized in descriptively in form of Country, No. of Patients in the study, Date published, Type of study, Age, Sex, Complaints being Primary or Recurrent, Management type, History of Hernia repair, Clinical Complaints, Side of Hernia complaint, Diagnostic modality, Preoperative Diagnosis, Type of Surgery, Use of Mesh, Hernia Repair done or not, Post-operative Diagnosis, Outcomes and Post-operative days till Discharge.

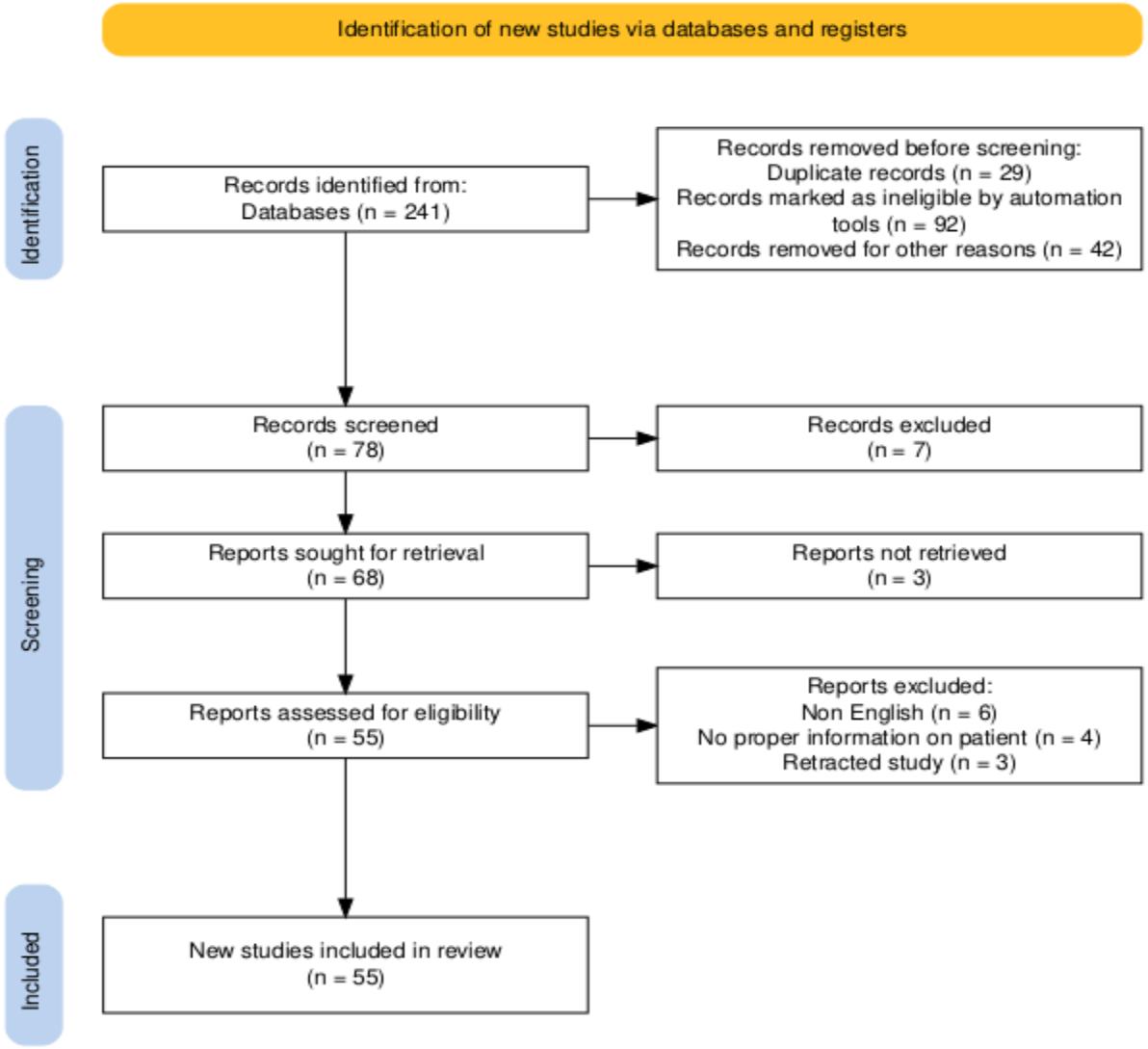

Figure 1: PRISMA flow diagram

Outcomes

The primary outcomes for our systematic review of the cases is to evaluate variations in clinical presentations of AH with Appendicitis, Diagnostic Modalities and treatment interventions as most cases of AH with Appendicitis are often misdiagnosed as Inguinal Hernia strangulation, testicular torsion, Enterocutaneous fistula and Acute Appendicitis. The secondary outcomes of this study were to compare utilization of mesh vs doing herniorrhaphy or No repair done to delay Mesh repair, compare the utilization of clinical acumen, CT scan and Ultrasound for diagnosis and compare approaches for the surgery i.e. Laparoscopic vs Open approach between two categories i.e. Elective and Emergency surgery management type and find the complications seen in both management types. Use of Mesh is done during Hernioplasty for which generally Polypropylene Mesh is taken which provides tension free Hernia repair. While in Herniorrhaphy, There is suturing of the layers of hernia sac after reducing the contents which is not tension free. In delayed Mesh repair, only reduction of hernia sac contents is done and when the inflammation is controlled, Mesh repair is done.

Statistical Analysis

We used Apple Numbers to collect data from those 55 studies which were converted to csv file and analysis of the data was done by a statistical software known as 'R' to evaluate the data on 59 patients(given as Table 1). The χ 2 and fisher's exact test was done for the categorical variables comparing utilization of Mesh repair and non-mesh repair in emergency and elective procedures and comparing utilization rates of different types of surgery for elective and emergency management type. A p value less than 0.05 is considered statistically significant in this study. No statistical power calculation was conducted prior to the study and the sample size was based on the available data. For individual variables such as Complaints, Gender, Use of diagnostic modality, Side of hernial complaints, history being primary or recurrent and Preoperative diagnosis of AH with appendicitis or not, the n(%) of the variables was calculated by dividing the number of patients with such variables by number of patients in data, multiplied by 100 which was then plotted on a clustered bar chart and Pie chart representing clinical complaints.

No	Study - Year published	No. Of Patients in Study	Type of Study	Country	Type of surgery	Sex (M/F)	Age	Primary / Recurrent Complaints	History of Hernia Repair	Side of previous hernia repaired	Chief complaints	Diagnostic modality	Preoperative diagnosis	Side	Post-operative Diagnosis	Management	Mesh Repair attime of operation	Herniorhapphy	Outcome	Post operative Days till discharge
1	Sengi et al. 2024 [18]	1	Case Report	Italy	Elective	M	85	Recurrent	Yes	Right sided	Painful irreducible bilateral inguinal mass, Vomiting, Constipation	CT scan	Amy and's Hernia with inflammation of appendix and Left side Inguinal hernia	Right sided and Left sided	Amy and's Hernia with Appendicitis	Bilateral Laparoscopic Hernia repair with Mesh (Bard 3D Max Polypropylene) and Appendectomy	Yes	Yes	Uneventful Post-operative course	4
2	Kawata et al. 2024 [19]	1	Case Report	Japan	Emergency	M	74	Recurrent	No	N/A	Fever, Firm Swelling in the right inguinal area, discomfort of scrotum	CT scan	Amy and's Hernia with inflammation of appendix	Right sided	Amy and's Hernia with Gangrenous Appendicitis	Emegency Appendectomy and Open Inguinal hernia repair without Mesh	No	Yes	Uneventful Post-operative course	9
3	Fjleh et al. 2024 [20]	1	Case Report	Syria	Emergency	M	77	Primary	No	N/A	Non-Painful irreducible right inguinal mass, Vomiting, Constipation	Ultrasound	Inguinal Hernia with isolated bowel loops	Right sided	Amy and's Hernia with Pus filled Perforated Appendicial Abscess	Open Inguinal Hernia repair and Appendectomy with J-P drain without Mesh	No	Yes	Uneventful Post-operative course	2
4	Choi et al. 2024 [21]	1	Case Report	Australia	Emergency	M	71	Recurrent	Yes	Left sided	Painful irreducible right inguinal mass	Ultrasound	Inguinal Hernia with isolated bowel loops	Right sided	Amy and's Hernia with Appendicitis	Emergency Laparoscopic Appendectomy and Laparoscopic Inguinal hernia repair with Mesh after 8	Yes	Yes	Uneventful Post-operative course	N/A
5	Chang et.al. 2024 [22]	1	Case Report	Taiwan	Emergency	F	25	Primary	No	N/A	Painful irreducible right inguinal mass	CT scan	Amy and's Hernia with inflammation of appendix	Right sided	Amy and's Hernia with Pus filled Perforated Appendicial Abscess	Laparoscopic Emergency Appendectomy and Inguinal hernia repair without Mesh with J-P drain	No	Yes	Uneventful Post-operative course	5
6	Rathod et.al. 2024 [23]	1	Case Report	India	Elective	M	71	Recurrent	Yes	Right sided and Left sided	Painful irreducible right inguinal mass	Ultrasound	Inguinal Hernia with isolated bowel loops	Right sided	Amy and's Hernia with Appendicitis	Emergency Appendectomy and Open Inguinal hernia repair without Mesh	No	Yes	Uneventful Post-operative course	2
7	Chagam et.al. 2024 [24]	1	Case Report	USA	Elective	M	76	Primary	No	N/A	Painful irreducible right inguinal mass, Vomiting, Constipation	CT scan	Inguinal Hernia with isolated bowel loops with enterocutaneous fistula	Right sided	Amy and's Hernia with Appendicitis	Laparoscopic Hernia repair with Mesh (Polypropylene) and Appendectomy with drain placement	Yes	Yes	Mesh infection	10
8	Zhimomi et.al. 2023 [25]	1	Case Report	India	Emergency	M	64	Primary	No	N/A	Painful irreducible right inguinal mass, discharge	Ultrasound	Inguinal Hernia with isolated bowel loops with enterocutaneous fistula	Right sided	Amy and's Hernia with Pus filled Perforated Appendicial Abscess	Emergency Appendectomy and Open Inguinal hernia repair without Mesh with J-P	No	Yes	Uneventful Post-operative course	5
9	Samsami et al. 2023 [26]	1	Case Report	Iran	Elective	F	74	Primary	No	N/A	Painful irreducible right inguinal mass	CT scan	Amy and's Hernia with inflammation of appendix	Right sided	Amy and's Hernia with Appendicitis	Open Inguinal Hernia repair and Appendectomy with J-P drain without Mesh	No	Yes	Uneventful Post-operative course	2
10	Rojas-Garza et al. 2023[27]	1	Case Report	Mexico	Emergency	M	57	Primary	No	N/A	Fever, Firm Swelling in the right inguinal area	CT scan	Amy and's Hernia with inflammation of appendix	Right sided	Amy and's Hernia with Appendicitis	Open Inguinal Hernia repair and Appendectomy with J-P drain without Mesh	No	Yes	Death due to unrelated Organ failure	N/A

11	Radboy et al. 2023 [28]		Case Report	Iran	Emergency	M	48	Primary	No	N/A	Painful irreducible right inguinal mass	Ultrasound	Amy and's Hernia with Inflammation of appendix	Right sided	Amy and's Hernia with Appendicitis	Laparoscopic Hernia repair without Mesh and Appendectomy	No	Yes	Uneventful Post-operative course	2
12	Litchinko et al. 2023 [29]		Case Report	Switzerland	Emergency	M	56	Primary	No	N/A	Fever, Firm Swelling in the right inguinal area, discomfort of scrotum	CT scan	Inguinal Hernia with isolated bowel loops	Right sided	Amy and's Hernia with Pus filled Perforated Appendicial Abscess	Open Inguinal Hernia repair and Appendectomy with J-P drain without Mesh	No	Yes	Uneventful Post-operative course	7
13	Jakovljevic et al. 2023[30]		Case Report	Serbia	Emergency	M	84	Primary	No	N/A	Painful irreducible right inguinal mass	Clinical	Inguinal Hernia with isolated bowel loops	Right sided	Amy and's Hernia with Gangrenous Appendicitis	Open Inguinal Hernia repair and Appendectomy with J-P drain without Mesh	No	Yes	Uneventful Post-operative course	4
14	Sun et.al. 2023 [31]		Case Report	Australia	Emergency	M	57	Primary	No	N/A	Fever, Firm Swelling in the right inguinal area, discomfort of scrotum	Clinical	Inguinal Hernia with isolated bowel loops	Right sided	Amy and's Hernia with Appendicitis	Open Inguinal Hernia repair and Appendectomy with J-P drain With Mesh	Yes	Yes	Acute oligoanuric renal failure due to ATN	7
15	Raj et al. 2023[32]		Case Report	UK	Emergency	M	76	Primary	No	N/A	Fever, Firm Swelling in the right inguinal area, discomfort of scrotum	CT scan	Acute Appendicitis	Right sided	Amy and's Hernia with Gangrenous Appendicitis	Laparoscopic Emergency Appendectomy and Inguinal hernia repair without Mesh with J-P drain	No	No	Uneventful Post-operative course	2
16	Olamaeian et al. 2022[33]		Case Report	Iran	Emergency	M	55	Recurrent	No	N/A	Painful irreducible right inguinal mass	Ultrasound	Inguinal Hernia with isolated bowel loops	Right sided	Amy and's Hernia with Appendicitis	Open Inguinal Hernia repair and Appendectomy with J-P drain without Mesh	No	Yes	Uneventful Post-operative course	N/A
17	Marzocchi et al. 2022[34]		Case Report	Italy	Emergency	M	50	Recurrent	Yes	Right sided	Painful irreducible right inguinal mass, discharge	CT scan	Inguinal Hernia with isolated bowel loops with enterocutaneous fistula	Right sided	Amy and's Hernia with Pus filled Perforated Appendicial Abscess	Open Inguinal Hernia repair and Appendectomy with J-P drain without Mesh	No	Yes	Uneventful Post-operative course	5
18	Gasinskaya et.al. 2022 [35]		Case Report	USA	Emergency	M	97	Primary	No	N/A	Fever, Firm Swelling in the bilateral inguinal area, discomfort of scrotum	CT scan	Amy and's Hernia with Inflammation of appendix	Right sided and Left sided	Amy and's Hernia with Appendicitis And Left sided indirect hernia	Laparoscopic Emergency Appendectomy and Inguinal hernia repair with Mesh with J-P drain	Yes	Yes	Uneventful Post-operative course but with concominant condition	N/A
19	Parekh et al. 2021 [36]		Case Report	USA	Emergency	M	81	Primary	No	N/A	Painful irreducible right inguinal mass	CT scan	Amy and's Hernia with Inflammation of appendix	Right sided	Amy and's Hernia with Appendicitis	Laparoscopic Emergency Appendectomy and Inguinal hernia repair without Mesh with J-P drain	No	Yes	Uneventful Post-operative course	5
20	Khalid et.al. 2021 [37]		Case Report	Pakistan	Emergency	M	28	Primary	No	N/A	Painful irreducible right inguinal mass	Ultrasound	Inguinal Hernia with isolated bowel loops	Right sided	Amy and's Hernia with Appendicitis	Open Inguinal Hernia repair and Appendectomy with J-P drain with Mesh	Yes	Yes	Uneventful Post-operative course	2
21	Gao et al. 2021 [38]		Retrospective Review of 6 cases	China	Emergency	M	77	N/A	N/A	N/A	Fever, Firm Swelling in the right inguinal area	CT scan	Acute Appendicitis and Inguinal hernia	Right sided	Amy and's Hernia with Pus filled Perforated Appendicial Abscess	Open Inguinal Hernia repair and Appendectomy with J-P drain without Mesh	No	Yes	Uneventful Post-operative course	N/A
				China	Emergency	M	64	N/A	N/A	N/A	Painful irreducible right inguinal mass	CT scan	Acute Appendicitis and Inguinal hernia	Right sided	Amy and's Hernia with Pus filled Perforated Appendicial Abscess	Open Inguinal Hernia repair and Appendectomy with J-P drain with Mesh	Yes	Yes	Uneventful Post-operative course	N/A
				China	Emergency	M	30	N/A	N/A	N/A	Painful irreducible right inguinal mass	Ultrasound	Amy and's hernia	Right sided	Amy and's Hernia with Appendicitis	Laparoscopic Emergency Appendectomy and Inguinal hernia repair with Mesh with J-P drain	Yes	Yes	Uneventful Post-operative course	N/A
				China	Emergency	M	77	N/A	N/A	N/A	Fever, Firm Swelling in the right inguinal area	CT scan	Acute Appendicitis and Inguinal hernia	Right sided	Amy and's Hernia with Pus filled Perforated Appendicial Abscess	Laparoscopic Emergency Appendectomy and Inguinal hernia repair without Mesh with J-P drain	No	No	Uneventful Post-operative course	N/A
22	Corvatta et al. 2021 [39]		Case Report	Argentina	Emergency	M	58	N/A	N/A	N/A	Painful irreducible right inguinal mass	CT scan	Amy and's Hernia with Inflammation of appendix	Right sided	Amy and's Hernia with Gangrenous Appendicitis	Laparoscopic Emergency Appendectomy and Inguinal hernia repair without Mesh with J-P drain	No	No	Uneventful Post-operative course	2
23	Caruso et al. 2021 [40]		Case Report	Italy	Emergency	M	80	Primary	No	N/A	Painful irreducible right inguinal mass, Vomiting, Constipation	Clinical	Inguinal Hernia with isolated bowel loops	Right sided	Amy and's Hernia with Pus filled Perforated Appendicial Abscess	Open Inguinal Hernia repair and Appendectomy with J-P drain without Mesh	No	Yes	Uneventful Post-operative course	2
24	Samsami et al. 2020 [41]		Case Report	Iran	Elective	M	21	Recurrent	No	N/A	Painful irreducible right inguinal mass, Vomiting, Constipation	CT scan	Inguinal Hernia with isolated bowel loops	Right sided	Amy and's Hernia with Appendicitis	Laparoscopic Emergency Appendectomy and Inguinal hernia repair without Mesh with J-P drain	No	Yes	Uneventful Post-operative course	2
25	O'Connor et.al. 2020 [42]		Case Report	UK	Elective	M	90	Primary	No	N/A	Fever, Firm Swelling in the right inguinal area, discomfort of scrotum	CT scan	Amy and's Hernia with Inflammation of appendix	Right sided	Amy and's Hernia with Appendicitis	NO surgical intervention. Patient on IV antibiotics, Supportive management for	No	No	Uneventful Post-operative course	12
26	Lee et al. 2020 [43]		Case Report	New Zealand	Emergency	M	87	Primary	No	N/A	Painful irreducible right inguinal mass	CT scan	Inguinal Hernia with isolated bowel loops	Right sided	Amy and's Hernia with Gangrenous Appendicitis and necrotic fasciitis	Open Inguinal Hernia repair and Appendectomy with J-P drain without Mesh	No	Yes	Uneventful Post-operative course but with concominant condition	2
27	Dadzan et.al. 2020 [44]		Case Report	Denmark	Emergency	F	79	Primary	No	N/A	Painful irreducible bilateral inguinal mass, Vomiting, Constipation	CT scan	Inguinal Hernia with isolated bowel loops	Right sided and Left sided	Amy and's Hernia with Gangrenous Appendicitis And Left sided indirect hernia	Emergency Appendectomy and Open Inguinal hernia repair without Mesh	No	No	Uneventful Post-operative course but with concominant condition	5
28	Syllaios et al. 2019 [45]		Case Report	UK	Emergency	M	58	Primary	No	N/A	Painful irreducible right inguinal mass, Vomiting, Constipation	Clinical	Inguinal Hernia with isolated bowel loops	Right sided	Amy and's Hernia with Appendicitis	Laparoscopic Hernia repair with Mesh and Appendectomy	Yes	Yes	Uneventful Post-operative course	2
29	Schaaf et al. 2019 [46]		Case Series	USA	Emergency	F	82	Primary	No	N/A	Painful irreducible right inguinal mass, Vomiting, Constipation	CT scan	Amy and's Hernia with Inflammation of appendix and femoral hernia	Right sided	Amy and's Hernia with Appendicitis and femoral hernia	Laparoscopic Hernia repair without Mesh and Appendectomy	No	Yes	Uneventful Post-operative course	2
30	Ornelas-Cortinas et al. 2019 [47]		Case Report	Mexico	Elective	F	78	Primary	No	N/A	Painful irreducible right inguinal mass	Ultrasound	Amy and's Hernia with Inflammation of appendix	Right sided	Amy and's Hernia with Gangrenous Appendicitis	Open Inguinal Hernia repair and Appendectomy with J-P drain with Mesh	Yes	Yes	Uneventful Post-operative course	2
31	Malik et al. 2019 [48]		Case Report	UK	Emergency	M	50	Recurrent	No	N/A	Painful irreducible right inguinal mass	CT scan	Inguinal Hernia with isolated bowel loops	Right sided	Amy and's Hernia with Pus filled Perforated Appendicial Abscess	Open Inguinal Hernia repair and Appendectomy with J-P drain without Mesh	No	Yes	Uneventful Post-operative course	2
32	Junttila et.al. 2019 [49]		Case Report	Finland	Emergency	M	32	Primary	No	N/A	Pain in abdomen at right lower quadrant at McBurney point	CT scan	Acute Appendicitis	Right sided	Amy and's Hernia with Pus filled Perforated Appendicial Abscess	Open Inguinal Hernia repair and Appendectomy with J-P drain without Mesh	No	No	Uneventful Post-operative course	1
33	Han et.al. 2019 [50]		Case Report	Taiwan	Elective	M	49	Primary	No	N/A	Painful irreducible right inguinal mass	CT scan	Amy and's Hernia with Inflammation of appendix	Right sided	Amy and's Hernia with Appendicitis	Laparoscopic Hernia repair with Mesh (Polypropylene) and Appendectomy	Yes	Yes	Uneventful Post-operative course	2

CASE REPORT

Case Presentation

A 39 year old male came to the emergency department(ED) with a 3 day history of Abdominal pain located at Right Lower Quadrant (RLQ). Upon arrival, the patient appeared to be non-toxic and was able to give a relevant history of the symptoms. The pain was gradual in onset, constant in character starting 3 days ago but got worse 1 day ago which prompted the patient to come to the emergency department. Patient described the pain as sharp and radiating to the back. It was not associated with vomiting, nausea, diarrhea, constipation, chills, urinary symptoms or fever. Patient also described there was a history of a similar episode of abdominal pain 3 months ago when he was traveling to Cuba. There the patient was diagnosed for acute appendicitis using ultrasound of the abdomen and was treated using oral antibiotics and bowel rest. Since then, the patient felt well and there were no such episodes till now. The patient's past medication history included 25 mg hydrochlorothiazide and losartan which he had also taken prior to coming to hospital for his medical history of hypertension. Family history of the patient was positive for Diabetes and hypertension present in him and his father, Bladder cancer present in his mother and Cardiac disease and Renal insufficiency present in his mother and father.

Physical Examination and Investigations

Upon arrival, Patient was non-toxic in appearance. He was afebrile with 101 beats/minute, respiratory rate 19/min, pulse ox. 98% and blood pressure of 114/61 mm Hg. Abdomen at the time of arrival was soft, non-distended, no masses, pain at the RLQ with palpation and presence of rebound tenderness with normal bowel sounds. Other systems were within normal limits. The urinalysis was shown to be unremarkable, while complete blood count (CBC) showed increased White blood cells i.e 19,300 /mm3 with high neutrophil count of 75%. Other parameters of CBC were within normal range along with the complete metabolic panel being unremarkable. As the patient was hemodynamically stable, he was taken for CT scan for the abdomen which revealed Inflammation of the Appendix as shown in Figures 2 and 3. The patient was diagnosed with Acute appendicitis.

Management

Patient was administered Piperacillin/Tazobactam 100 ml at 25 ml/hr IV 8 hourly, Isotonic Sodium Chloride(NaCl) 1L at 75 ml/hr IV, Morphine sulfate 2 mg/ml IV Inj. 4 hourly, Ondansetron 4 mg/2 ml Inj. IV 6 hourly and Zolpidem 5 mg tablet for suspected appendicitis, controlling dehydration, Severe pain, Nausea and Insomnia respectively. After stabilizing the patient, he was then admitted into our hospital where the NaCl was then stopped and initiation of Enalaprilat sodium in form 1.25 mg/ml Inj. IV 4 hourly. Patient was then scheduled for an Emergency Laparoscopic Appendectomy with the consent of the patient.

During the operation, we saw an inflamed pus filled appendix with the tip going inside the deep inguinal ring. It was inflamed but not perforated and adhered with the lateral aspect of the ascending colon as shown in Figure 4. There was presence of adhesions near the appendiceal mass which is consistent with inflammation of the peritoneum. A harmonic scalpel was used to dissect the adhesions present around the appendix. The tip of the appendix was pulled from the deep inguinal ring with the help of a laparoscopic grasper. A gastrointestinal anastomosis stapler was used to divide the base of the appendix. During this, there was some leakage

of pus which was then drained. No mesh repair was done and Herniorrhaphy of the patient was done during the operation. It measured 4.6 cm long × 1.5 cm diameter with a lumen up to 0.5 cm.

Outcome

Patient recovered well after the surgery, and was ambulated till morning, the next day after surgery. Patient was then discharged at Post Operative day 3 after no acute findings were seen on the patient. She came to follow-up for no additional complaints after the surgery.

Figure 2: Inflamed Appendix

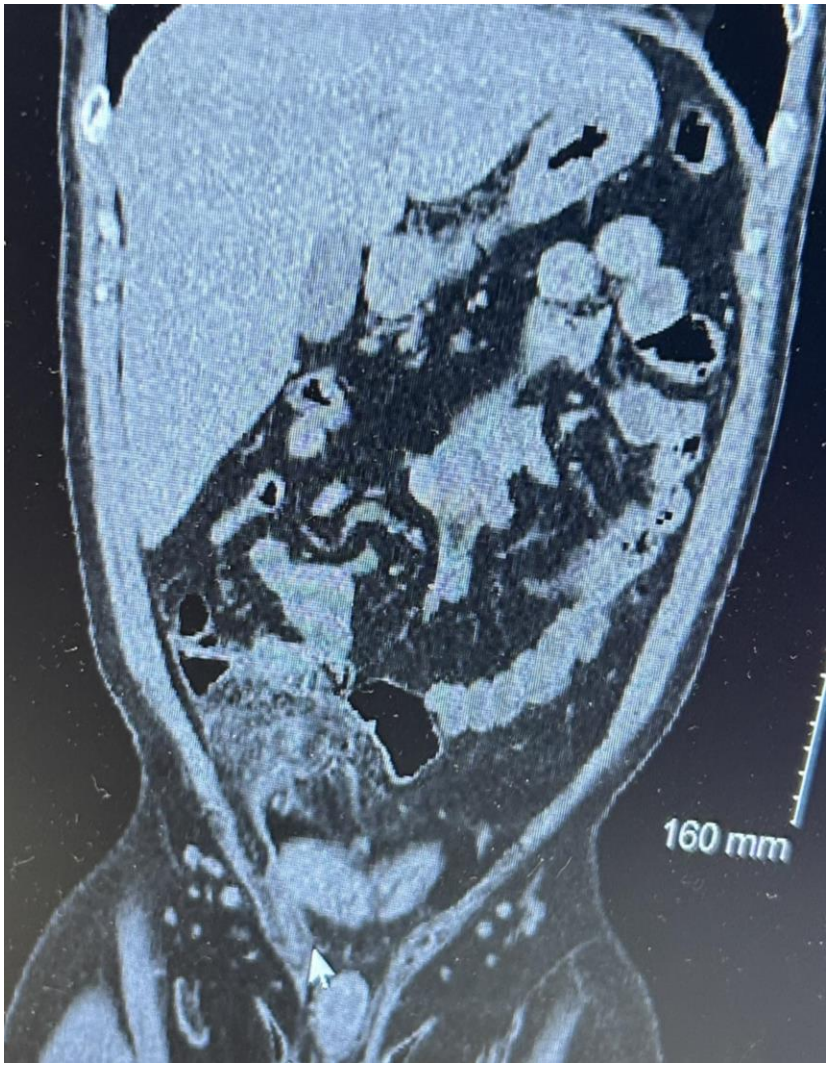

Figure 3: Appendix in Inguinal Canal

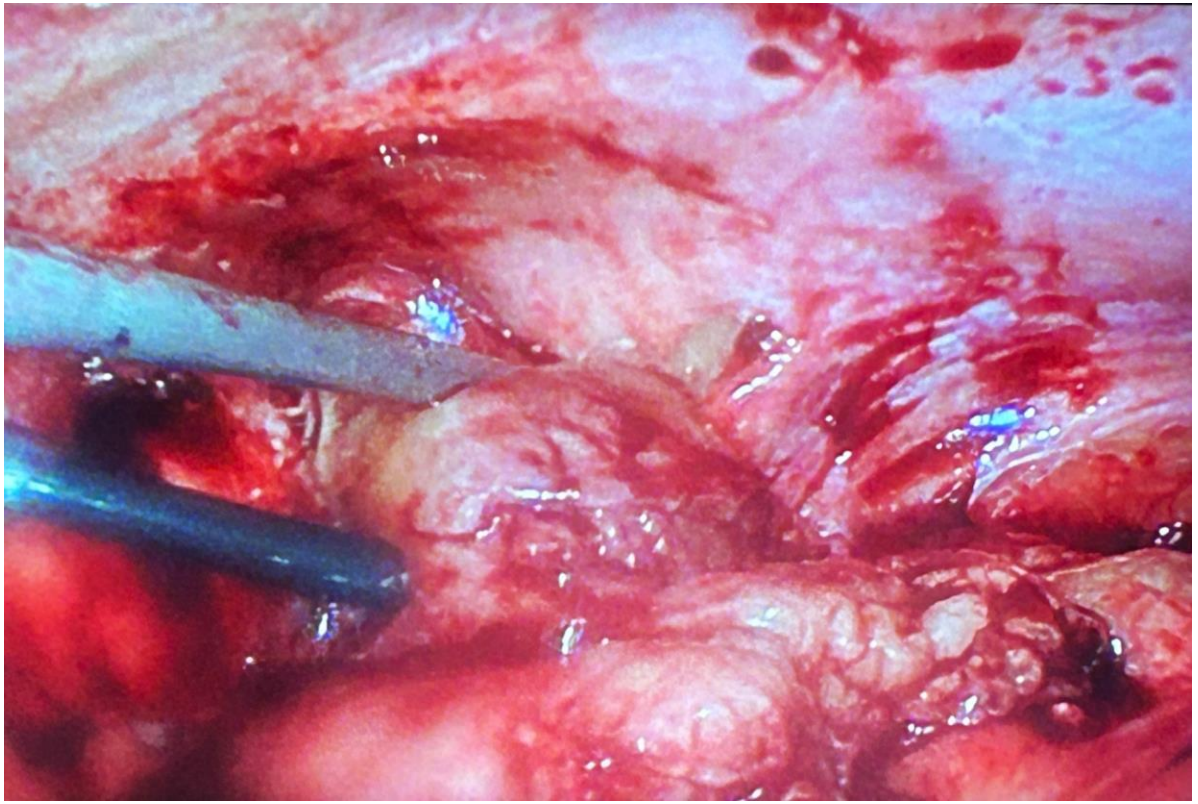

Figure 4: Inflamed Pus Filled Appendix in Lap. Appendectomy

RESULTS

We analyzed 59 patients in 55 published studies which included 51(92.7%) Case Reports, 2(3.67%) Case Series and 2(3.67%) Retrospective reviews which fulfilled the inclusion criteria for this review. Table 1 represents the distribution for all the studies. The cohort of the studies and its publication year is shown in Figure 5 and Table 2.

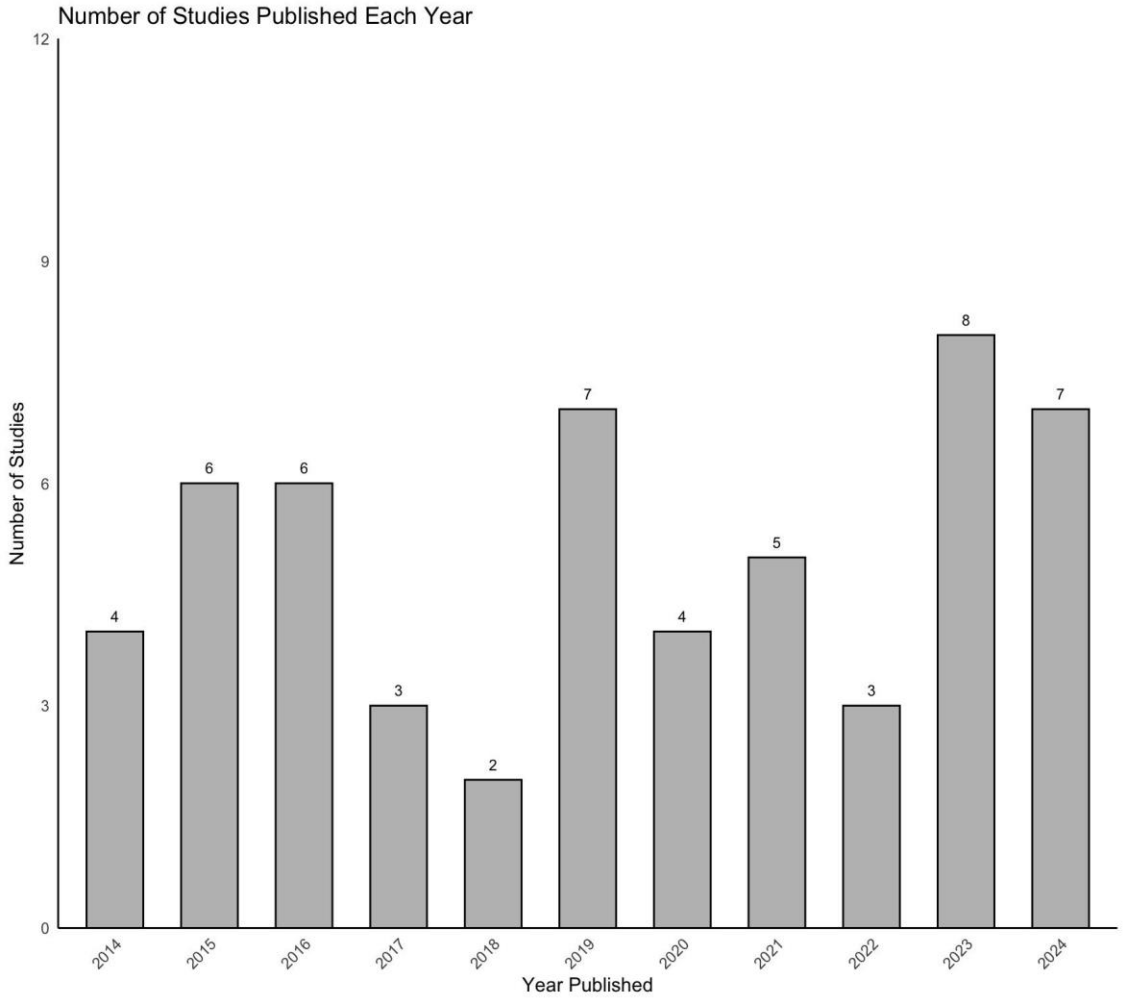

Figure 5: No. Of Studies Published Each Year

Table 2

Study type	Number of studies (n %)
Case report	51 (92.7%)
Retrospective reviews	2(3.67%)
Case Series	2(3.67%)

Epidemiological and Clinical Characteristics

Overall, the mean age of the patient was 63.54 ± 20.33 with 51 (86.4%) being males and 8 (13.6%) being females. There were 55 (93.2%) patients with Right sided inguinal mass with appendicitis, 1 (1.7%) patient presented with left sided inguinal mass with appendicitis and 3 (5.1%) patients presented with bilateral inguinal mass with appendicitis. There were 14 (23.7%) patients which had similar recurrent complaints, while 40 (67.8%) patients had primary complaints and there were 5 (8.5%) patients where there was no data that was available. Based on the data, we found that there were 45 (76.27%) cases of the patients who were treated in an emergency setting while 14 (23.73%) cases of the patients were treated in an elective setting after control of inflammation. From the 59 cases of AH with appendicitis, there were patients who showed constitutional symptoms such as fever, vomiting and constipation which includes 13 (22%) patients who presented with Constipation and 14 (23.7 %) patients who presented with fever and vomiting each. Thus, most patients with AH with appendicitis mostly show localized complaints of the disease. Out of the 55 cases with right inguinal mass, 39 (66.1%) cases showed painful inflammation over the right inguinal area, while 13 (22%) cases showed a firm reducible swelling in the right inguinal area and other 3 (5.1%) cases showed pain at the McBurney's point. There were 3 (5.1 %) patients presented with discharge at the right inguinal area mass and 7 (11.9%) patients presented with discomfort of the scrotum. The one case with left sided AH with appendicitis also presented with localized Pain at the left sided inguinal mass (1.7%) and out of 3 cases which has bilateral inguinal mass AH with appendicitis, 2 (3.4%) cases presented with localized pain in the bilateral inguinal area and 1 (1.7%) case with bilateral firm swelling with constitutional symptoms. The data for Chief complaints of the patient is summarized in table - 3. Based on the data, 40 (67.8%) patients presented with primary complaints and 14 (23.7%) patients had recurrent complaints of the complaints mentioned above. There were 5 studies of 5 (8.5%) patients which did not mention these complaints being primary or recurrent in the patients. The patient with preoperative diagnosis of AH with appendicitis were noted to be 22 (37.3%), patients with preoperative diagnosis of Inguinal Hernia with strangulation was noted to be 32 (52.5%) and there were 5 (8.5%) patients with preoperative diagnosis of Acute Appendicitis. Figure - 6 shows the above data in the form of a clustered bar chart for the patient characteristics.

Diagnostic and Management Outcomes

The patients are divided in two categories namely Elective group and Emergency group to compare the utilization of CT scan, Clinical diagnosis and Ultrasound in patients with elective surgeries and emergency surgeries, compare between utilization of Mesh and Non-mesh approach between Emergency and Elective surgery and compare the utilization of Laparoscopic approach with Open approach in emergency and elective surgery, all of which is described in Table 4 to 6.

Table 3

Chief Complaint	N (%)
Constipation	13 (22%)
Diffuse pain in abdomen	1 (1.7%)
Discharge from scrotum	3 (5.1%)
Discomfort of scrotum	7 (11.9%)
Fever	14 (23.7%)
Firm Swelling in the bilateral inguinal area	1 (1.7%)
Firm red Swelling in the right inguinal area	13 (22%)
Non-Painful irreducible right inguinal mass	1 (1.7%)
Pain in abdomen at McBurney point	3 (5.1%)
Painful irreducible bilateral inguinal mass	2 (3.4%)
Painful irreducible left inguinal mass	1 (1.7%)
Painful irreducible right inguinal mass	39 (66.1%)
Vomiting	14 (23.7%)

Table 4

Type	Clinical Diagnosis Utilization	CT Scan Utilization	Ultrasound Utilization	p- value
Elective	2(14.3%)	9(64.3%)	3(21.4%)	0.72064
Emergency	13(28.9%)	22(48.9%)	10(22.2%)	0.22809

Table 5

Type	Laparoscopic Approach Utilization	Open Approach Utilization	p-value
Elective	5(41.67%)	7(58.33%)	0.68427
Emergency	12(26.67%)	33(73.33%)	0.00002

Table 6

Type	Herniorrhaphy U	Mesh Repair Util	No Repair done	p - value
Elective	6(50.0%)	6(50.0%)	0(0.0%)	N/A
Emergency	30(66.7%)	10(22.2%)	5(11.1%)	0.00041

Based on the data in Figure 6, use of Computed Tomography (CT scan) was done on 31 (52.5%) patients which was more than Clinical diagnosis done on 15 (25.4%) and use of ultrasound done on 13 (22%).

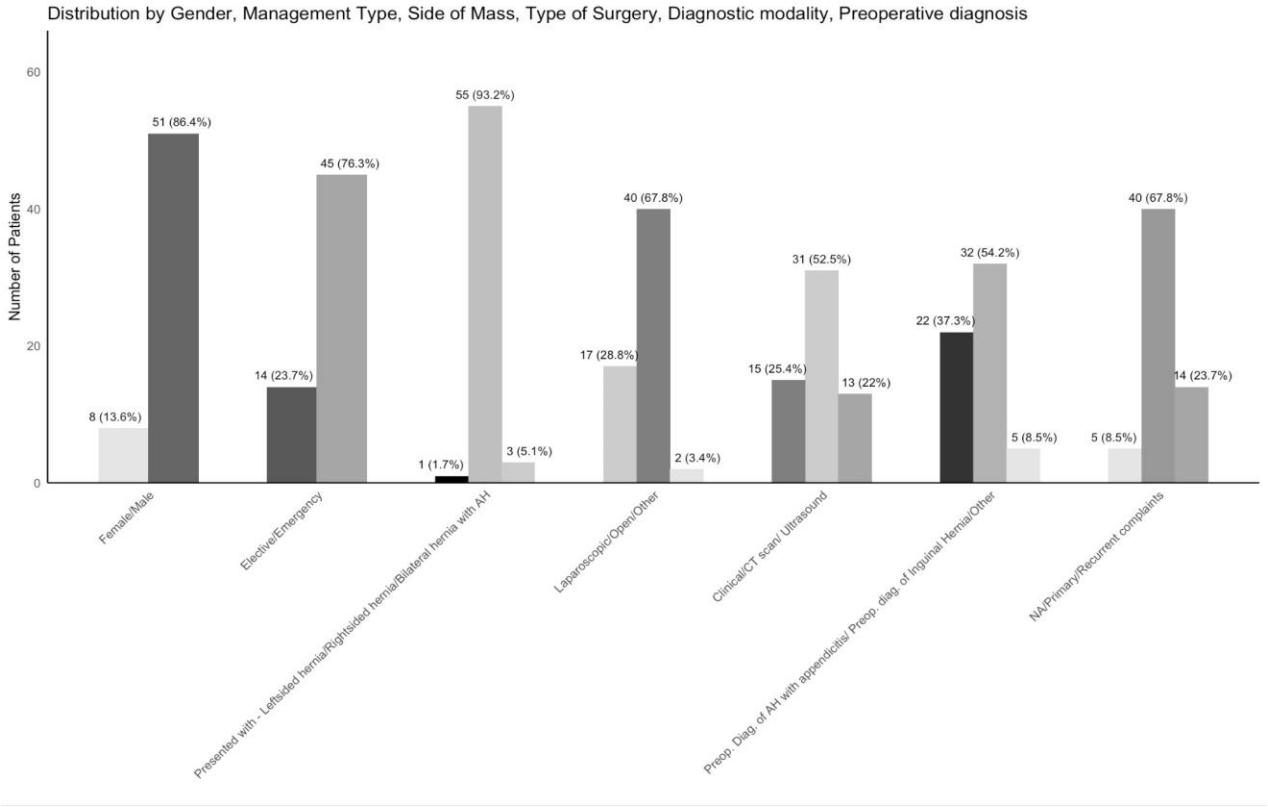

Figure 6: Patient Characteristics

As per the data in Table 4, use of CT scan in 22(48.9%) was used more frequently out of 45 cases of emergency management with no statistically significant difference (p-value >0.05) compared to Use of Clinical in 13(28.9%) and use of Ultrasound in 10(22.2%). Ultrasound in 3 (21.4%) patients, clinical diagnosis in 2(14.3%) and CT scan in 9(64.3%) are used for diagnosis of Amyand's Hernia with appendicitis is statistically not significant difference with p value>0.05 in 14 cases with elective management with surgery.

Based on the data in the Table 5, Out of 45 cases with Emergency surgery management type, 12 (26.67%) patients had Laparoscopic approach utilization while 33 (73.33%) patients had Open approach utilization which is also statistically significant in Emergency surgery management (p-value < 0.001). According to the Table 5, we found that in 12 cases with elective surgery management type, there were 5 (41.67%) cases with Laparoscopic approach utilization and 7 (58.33%) cases with Open approach utilization which is statistically not significant as p-value >0.05. Thus, we can conclude that the utilization of Open approach is generally more

than utilization of laparoscopic approach in emergency management. No significant difference in utilization of laparoscopic approach compared to open approach in elective management group.

Based on the data in the Table 6, we found that Herniorrhaphy which suturing of the fascias is more commonly done i.e. 30 (66.67%) cases compared to mesh utilization seen in 10 (22.22%) patients and No repair i.e. letting the defect open till reduced inflammation and then surgery with herniorrhaphy which is done in 5 (11.11%) patients out of 45 cases with Emergency Management type with statistically significant difference (p-value < 0.05). While in 12 patients with elective management, there was statistically significant difference between Mesh repair vs Herniorrhaphy (50% vs 50%) with p value N/A.

According to the studies in Table 1, Chagam et.al. Showed 1 case of Mesh infection which was managed by laparoscopic approach, Riojas-Garza et.al. Reported a case of death due to organ failure unrelated to management by Open approach and Sun et.al. Showed that the patient had Acute oligoanuric renal failure after the Open approach management of the patient. For the 2 patients out of 59 patients included which were treated by Supportive management and I&D in case of discharge and not in the analysis, showed that one patient in O'Connor et.al. Had an uneventful postoperative outcome after 12 days of supportive treatment and Loberant et.al. Had reported the death of the patient. The data for the no. of studies published about AH with appendicitis is also given in Figure 6.

DISCUSSION

Amyand's Hernia is defined to be the presence of Vermiform appendix in an inguinal hernia sac with the term given by Claudius Amyand when he performed the first Appendectomy [8]. This may be explained in pediatric patients because of the possibility of patent processus vaginalis [9]. Diagnosis of AH is considered uncommon as it is less than 1% of cases with inguinal hernia but for cases of AH with Appendicitis is rare even with other types of groin hernia containing inflamed appendix such as DeGarengot's hernia and Giant Inguinoscrotal Multivisceral Hernia[10,7]. It is due to the inflammation of the appendix within the hernia sac of Amyand's Hernia which is seen in the range of 0.07% to 0.13% in cases of inguinal hernia [22]. Herein, we reported a case of Amyand's Hernia which got complicated with Appendicitis while being diagnosed as Acute appendicitis. AH is recognized as cases with the most incarcerated but with low inflammation [11]. We also did a systematic review on the available literature of AH with appendicitis determining the utilization rates, clinical complaints and complication of the treatment.

Even though the condition AH has been known since 1734, the cases of AH with appendicitis are very rare to come across, leading to less interest in this topic. The interest peaked in the decade 2014-2024 with more studies after 2019 as seen in the Figure - 5. Most of the studies were case reports or small patients case series with only two studies being retrospective review [38,66,46,68] from which only one review and one case series has multiple patients diagnosed as AH with appendicitis or with complications of Inflammation such as Pus formation, enterocutaneous fistula, etc. This accounts for 7.33% of the studies of the entire study with a patient pool of only 13.56% of cases which explains the lack of large patient series, and maybe possibility of underreporting of AH with appendicitis.

From the studies, we determined that the mean age of patients with AH with Appendicitis is 63.54 containing male to female ratio of 6.375:1 which is higher than cases of inguinal hernia [13]. As we noted from the table - 3, the cases of AH with appendicitis is most commonly associated with right sided swelling due to anatomical proximity of the appendix to the inguinal canal. Interestingly, 1.7% case presented with left sided Amyand's Hernia with Appendicitis which may be associated with high cecal mobility, intestinal malrotation and situs inversus [55].

The presentation of Inguinal mass is most commonly seen in the right side which is around 93.2% of the included study cases. The clinical complaints of patients of AH with appendicitis ranges from non-specific symptoms such as Fever, constipation, scrotal discharge and diffuse abdominal pain to Painful right inguinal mass which is seen in 66.1% of patients. These overlaps of symptoms are often the cause for making preoperative diagnosis of AH with appendicitis very unlikely for the doctors. There were complicated cases of discharge from scrotum seen in some cases [25,34,59]. From our data, the preoperative diagnosis of AH with appendicitis seen in 37.3% of cases. It may be explained due to the advent of CT scan as symptoms of appendicitis also present in these patients which can show us an inflamed appendix within hernia sac, thus not getting shadowed by the hernia complaints. The superiority of CT scan is the ability to delineate the anatomic structures [13,14]. This also explains that the use of CT scan for diagnosis is more common seen in 52.5% of cases in the study. Ultrasound is a cheaper option which is also highly unreliable in these cases [24]. The use of CT scan was more commonly seen in studies after 2019 as seen in the Table 1. The second most common modality for diagnosis in the patients was Clinical diagnosis which is preferred by the surgeons due to differential diagnosis of strangulated hernia complaints which also prompt for immediate surgery.

Patients generally present commonly as Primary complaints as seen in 67.8% of cases with patients having no symptoms or history of similar complaints or history of previously treated hernia. These may be explained due to acute presentation of the complaints which lead to prompt surgical intervention by the physicians. We discovered that most of the studies' cases presented in an Emergency setting seen in 76.3% of the total pooled patient's cohort were treated with Emergency surgery while some cases presented in the clinic and treated elective after antibiotic therapy. There was one case which was treated supportively due to the patient's refusal for surgery [42] and one case which was treated with supportive management along with Incision and drainage of the abscess [65]. Interestingly, both cases were patients of Octogenarian age groups. Thus, our data showed that the majority of patients are treated in emergency settings and require immediate surgery. The guidelines for Amyand's Hernia types are noted in Losanoff and Basson's classification which promotes treatment as a form of Open approach [2]. This explains the utilization of open approach is seen to be more than laparoscopic approach in emergency cases (Table - 5). There are concerns of utilization of Mesh in cases of AH with appendicitis as a definitive treatment is contemplated [7]. We found from our data that the utilization of Non-Mesh repair such as Herniorrhaphy and No repair is more compared to Non mesh repair seen in 66.6% and 11.1% vs 22.2%. No repair is used frequently in cases with necrotic infection or very catarrhal inflammation as seen in Table 1. Mesh repair was generally avoided and herniorrhaphy was preferred in most cases. This is explained by WSES and Losanoff and Basson's guideline for treating Amyand's hernia as explained by Papaconstantinau et.al [7] which states the management for grade 2C type which is managed with

herniorrhaphy. Even though the use of Mesh is limited, the cases of AH with appendicitis are mostly underreported due to small sample sizes of the studies. There is still debate on the use of Mesh in hernia repair in AH with appendicitis even with acute settings which can be with controllable morbidity, surgical site infection and mesh infection [15,16]. Overall, there were 2 cases with postoperative death seen, but with small patient size in study. Thus, due to publication bias being highly likely, the cases are mostly seen with no postoperative complications. Ebaugh et.al. Reported with prolonged ileus and pulmonary embolism. Sun et.al. Reported acute oligoanuric renal failure and Chagam et.al. Presented Mesh infection post operatively.

The main limitations of this study are potential publication bias, selection bias, small sample size of the patient, Methodological bias and reporting bias. The publication bias is because the majority of the study is heavily relied on Case reports, Case series and retrospective reviews which leads to over-representation of successful or notable cases causing skewed conclusions. The selection bias is due to exclusion of pediatric and Non English cases which was because of pediatric cases having different mechanisms of having Amyand's Hernia [9]. This can limit the generalizability of the data to be for all age groups. Small sample size also reduces the statistical power of the study. Due to study being limited to Pubmed, it leads to restriction of comprehensiveness of the study. Reporting bias is seen in some cases for Primary and recurrent complaints as it was not available.

According to the data, we propose the management of AH with appendicitis should be individualized and stratified according to the Centers for Disease Control and Prevention (CDC) surgical wound classification system and the Ventral Hernia Working Group (VHWG) grading platform [17]. It states that the cases of AH with appendicitis should be managed with emergency surgical intervention with Appendectomy and Herniorrhaphy of the patient. Use of mesh should be avoided in case of perforated appendix or periappendiceal abscess. While in cases of severe necrotic infection, No repair should be preferred.

CONCLUSION

A case report of Amyand's Hernia complicated by appendicitis can be treated in a laparoscopic approach which can be a safe and feasible approach in elective setting. We also report the pooled patient analysis which reports that the method of hernia repair post appendectomy in cases of Amyand's Hernia should be individualized and stratified according to the extent of inflammation of the appendix. More consistent reporting of cases of Amyand's hernia complicated by Appendicitis should be encouraged to optimize treatment and improve patient outcomes.

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