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Case Report

An unusual case of the Amyand hernia with perforated appendicitis and a sessile serrated lesion within a recurrent inguinal hernia

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ABSTRACT

The Amyand hernia is a rare type of inguinal hernia where the appendix is located within the hernial sac. It is present in less than 1% of inguinal hernias and in 0.13% of cases, it is associated with complications including acute appendicitis and perforation. This case details an 81-year-old female with recurrent Amyand hernia with perforated appendicitis and a sessile serrated lesion. A literature review was conducted to identify existing cases of the Amyand hernia and neoplasm or recurrent inguinal hernia. Fifteen cases of neoplastic appendix within an Amyand hernia were identified in the literature. All cases found acute appendicitis, three of which were perforated. Histology varied from adenocarcinoma, goblet cell, carcinoid, fibroma, and mucinous cystadenoma. There were nine identified cases of Amyand hernia within a recurrent inguinal hernia. Seven of these cases had acute appendicitis, three of which were perforated, whilst one was gangrenous. All cases performed an appendicectomy, and various open techniques were used for hernia repair with or without mesh. The most interesting finding of the literature review was that Amyand hernia is rarely diagnosed pre-operatively, and that there was a high incidence of appendicitis when in the presence of a neoplastic appendix or a recurrent inguinal hernia. This paper highlights the first reported case of an Amyand hernia with a simultaneous neoplasm and recurrent inguinal hernia, which was managed using a laparoscopic approach without hernia repair.

Keywords: Amyand's hernia, Inguinal hernia, Recurrent inguinal hernia, Neoplasm, Appendicitis, Case report

INTRODUCTION

The Amyand hernia is a rare type of inguinal hernia, first described by the French surgeon Dr. Claudius Amyand in 1735, in which the appendix is located within the hernial sac.[1,2] It has an incidence rate of less than 1% of inguinal hernias, and in 0.13% of cases it is associated with complications including perforation, abscess, or acute appendicitis.[2,3] The Amyand hernia is three times more common in childhood, due to affiliation with persistent processus vaginalis.[3, 4] It is also more common in males, and usually occurs on right side, with mean age of 34 years.[3-5] Clinical presentation of Amyand hernia can vary widely, from asymptomatic, to acute appendicitis or even sepsis.[2,6] The diagnosis of Amyand hernia can be challenging, as it can be easily misdiagnosed as an inguinal hernia.[7] Imaging is not often successful in correctly diagnosing this condition, as most cases are diagnosed intra-operatively.[8-10] This misdiagnosis can result in inappropriate surgical intervention, leading to potential complications such as bowel perforation, sepsis, and even death.[3,6] Therefore, it is essential to understand the clinical presentation, diagnosis, and management of Amyand hernia to ensure optimal patient outcomes. This paper highlights a rare case of an 81-year-old female with an Amyand hernia within a recurrent inguinal hernia, with perforated appendicitis and a sessile serrated lesion. The existing literature of these topics is also reviewed.

CASE REPORT

An 81-year-old female presented to emergency department with fevers, lower abdominal pain, and a palpable right groin lump. She had a distant history of bilateral inguinal hernia repairs, although the timing and specific details were unknown. Further medical history included necrotising fasciitis of right lower leg, hypothyroidism, osteoarthritis, osteoporosis, depression, diverticulitis, bladder and rectal prolapse. She reported 1 week of abdominal pain, which was suprapubic and umbilical, radiating through the right flank and towards the back. It was worse post-prandial and felt similar to previous episodes of diverticulitis, but worse in severity. Her bowels had opened that morning and she denied diarrhoea, bleeding/ melena. She reported reduced appetite, nausea, and a single episode of green vomitus. She also described chronic constipation and 13 kg of weight loss over 1 month. On examination she was febrile, drowsy, and had bilateral crackles to lung bases. The abdomen was soft with scant bowel sounds. There was tenderness to right lower quadrant, with both Rovsing's and Murphy's signs positive. The patient had palpable right groin lump which was tender and the computed tomography (CT) did not initially report the appendix, but reported free fluid in right iliac fossa and fat containing right femoral hernia (Figures 1-2). Surgical specialist review of patient and CT scan identified possible Amyand hernia. Ultrasound arranged to distinguish an inguinal vs femoral hernia. Ultrasound reported an Amyand hernia with features of appendicitis and abscess formation (Figure 3). An emergent laparoscopic appendicectomy was performed without complications, and confirmed diagnosis of Amyand hernia. The appendix was reduced from direct inguinal hernia and then resected (Figures 4-5). No hernia repair was performed. There were no post-operative complications, and patient discharged after 2 days. Histo-pathology revealed perforated appendicitis and sessile serrated appendiceal lesion, with clear resection margins (Figures 6-7). There were no complications/reoccurrence reported in 12-month period following surgery. Written consent was obtained from patient for publication of de-identified case details and associated images.

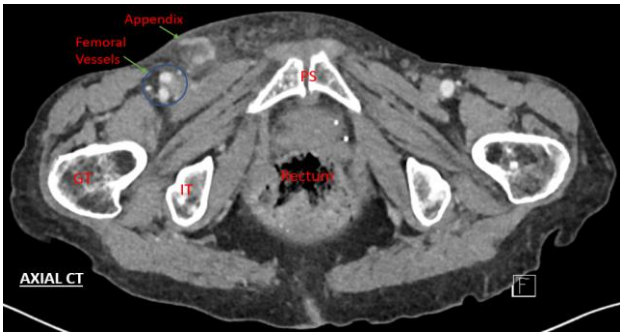

Figure 1: Axial CT scan demonstrating the appendix outside of the abdominal cavity.
GT: greater tuberosity, IT: ischial tuberosity, PS: pubic symphysis.

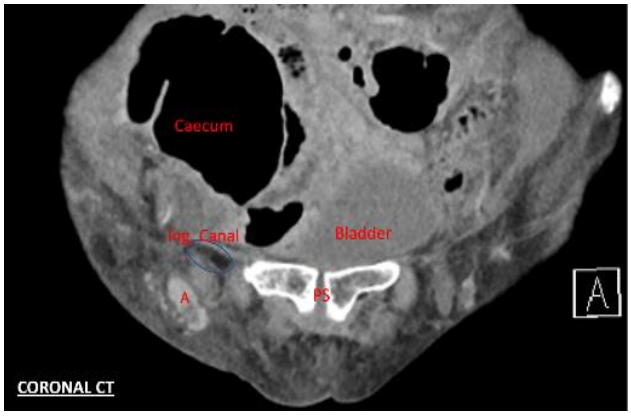

Figure 2: Coronal CT scan demonstrating the appendix herniating through the superficial ring of the inguinal canal.
Ing.: inguinal, A: appendix, PS: pubic symphysis.

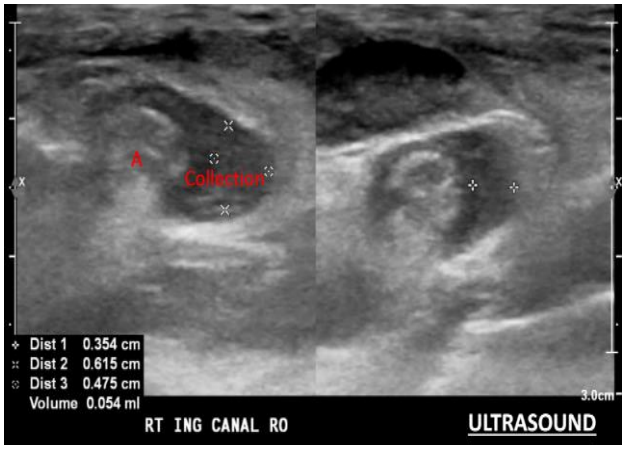

Figure 3: Axial ultrasound demonstrating the appendix with an adjacent collection measuring 6.2×4.8×3.5 mm.
A: appendix.

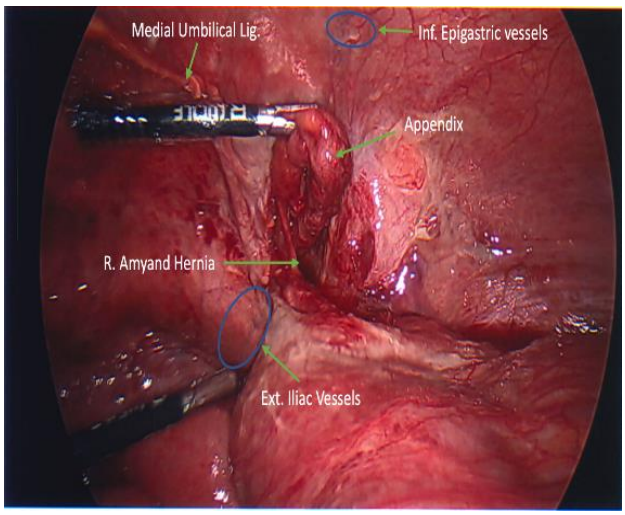

Figure 4: Intraoperative laparoscopic photograph of appendix within the inguinal canal.
Lig: Ligament, Inf: inferior, R: right, Ext.: external.

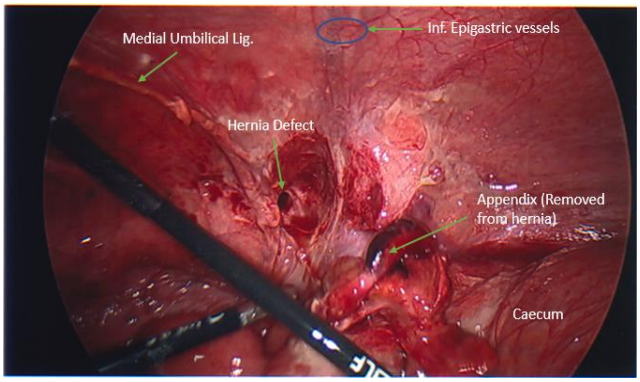

Figure 5: Intra-op laparoscopy of appendix once reduced from inguinal canal.
Lig: Ligament, Inf: inferior.

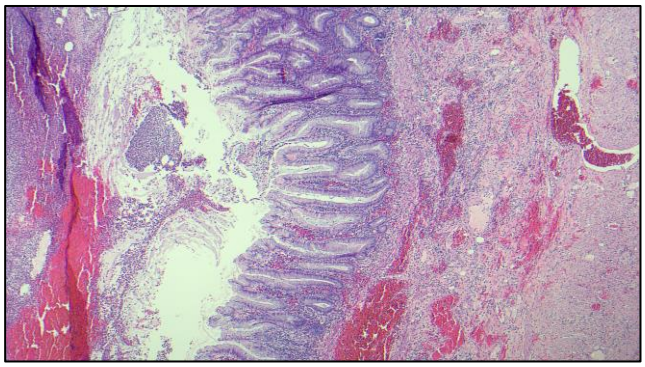

Figure 6: Acute suppurative appendicitis with perforation and sessile serrated lesion.

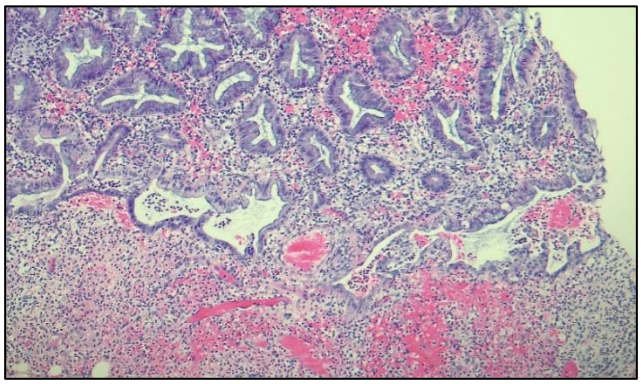

Figure 7: Histology of sessile serrated lesion demonstrating serrated crypts and basal crypt dilatation, with extension along basement membrane.

DISCUSSION

Diagnosis

The Amyand hernia is difficult to diagnose clinically, often misdiagnosed as an incarcerated inguinal hernia.[11] CT is the recommended modality for diagnosis of it and its possible complications. US and MRI should be considered in children and pregnant females.[12] This case used a combination of clinical examination and imaging to achieve accurate diagnosis. The CT initially incorrectly reported a fat containing inguinal hernia. However US confirmed that contents of hernia was in fact appendix. That is, a blind-ending tubular structure, arising from caecum and located inside inguinal canal.[12] Clinicians should consider ultrasound during their work-up of patients with suspected Amyand hernia to aid in diagnosis.

Surgical management

The majority of surgical interventions for Amyand hernia in the literature have utilised an open inguinal surgical incision. Hernia repair is commonly performed, although use of synthetic mesh is controversial when appendicitis is present, due to increased risk of infection.[2,7-9] In these cases, biological mesh could be used as alternative.[13] Appendicectomy is widely accepted as part of Amyand hernia management where appendicitis is present, however it is disputable in a normal appendix. This is because intestinal contents may contaminate the hernia repair, and it removes ability to use appendix in future for other purposes such as a urinary conduit.[7,14,15] However, performing an appendicectomy reduces the risk of future appendicitis and further surgery.[9] The laparoscopic approach has become increasingly popular over recent years for advantages of shorter hospital stay and reduced risk of infection and ileus.[16]

In this case report, a laparoscopic approach without hernia repair was performed. This was because there was no radiographic or intra-operative evidence of bowel herniation, and the hernial defect was quite small in size. No further hernia repair or bowel resection was planned due to the patient's age, comorbidities, and clear resection margins of the sessile serrated lesion.

Neoplasm

Carcinoid tumours were the most commonly reported neoplasm identified in the appendix, with incidence of about 2%.[3,17] Study by Enblad et al 2017 found that untreated appendicitis increases the risk of right-sided colon and appendiceal cancer.[18] They suggested that a local inflammatory process was a possible contributor to the pathogenesis of cancer, due to physical proximity of the appendix and associated cancers.[18] Furthermore, inflammation is well understood to contribute to development, growth, and metastasis of tumours.[19] The sessile serrated lesion identified in this case is a known precursor of colorectal cancer, most commonly occurring in proximal colon.[20]

Only a small number have reported a neoplastic appendix in association with Amyand hernia. Sarici et al (2019) and Baldinu et al (2022) have previously summarised existing case reports.[3,17] An updated literature review was conducted in this paper to capture identifiable case reports of neoplasm within an Amyand hernia. These cases are summarised in Table 1.

Fifteen cases of neoplastic appendix in Amyand hernia hernia were identified in existing literature. Interestingly, all cases identified acute appendicitis with either incarceration/ strangulation, and 3 were perforated appendixes. Difficulty of diagnosis is evident, as most cases were preliminarily diagnosed as inguinal hernia. All cases performed appendicectomy, whilst various techniques used for hernia repair with/ without mesh.

Only 2 cases used a laparoscopic approach. Carcinoid tumours and mucinous cystadenoma were most prevalent. There was no reoccurrence of neoplasm reported, although very few cases detailed patient follow-up.

Recurrent inguinal hernia

An Amyand hernia identified within a previously repaired inguinal hernia has been seldom reported. A literature review was conducted to capture all case reports of this phenomenon, as summarised in Table 2.

Table 1: Review of the literature on cases of neoplastic appendix within an Amyand hernia.[3,4,17,21-24]

S. No.	References	Year	Age (Years)	Sex	Preliminary diagnosis*	Hernia side	Hernia type	Hernia status	Intra-op findings
1.	Arenas	2022	72	M	Inguinal hernia	Right	Indirect	Incarcerated	Acute appendicitis
2.	Ashcroft	2023	82	F	Groin abscess	Right	Indirect	Incarcerated	Acute appendicitis (perforated)
3.	Christodoulid	2017	52	M	Inguinal hernia	Right	Indirect	Incarcerated	Acute appendicitis
4.	Elbanna	2015	81	M	Inguinal hernia	Right	Indirect	Incarcerated	Acute appendicitis
5.	Fiordaliso	2021	87	M	Inguinal hernia	Right	Indirect	Incarcerated	Acute appendicitis
6.	Karanikas	2015	92	F	Inguinal hernia	Right	Indirect	Incarcerated	Acute appendicitis (perforated)
7.	Nahmias	2013	50	M	Inguinal hernia	Right	Indirect	Strangulated	Acute appendicitis
8.	Oh	2019	37	M	Acute appendicitis	Right	Indirect	Incarcerated	Acute appendicitis
9.	Oyelowo	2020	28	M	Inguinal hernia	Right	Indirect	Incarcerated	Acute appendicitis
10.	Reynu	2015	70	M	Inguinal hernia	Right	NA	Strangulated	Acute appendicitis
11.	Salemis	2006	61	M	Inguinal hernia	Right	Indirect	Strangulated	Acute appendicitis (perforated)
12.	Sarici	2019	64	M	Inguinal hernia	Right	Indirect	Incarcerated	Appendix attached to mass
13.	Shabeeb	2010	62	M	Inguinal hernia	Right	Indirect	Incarcerated	Acute appendicitis
14.	Wu	2010	62	M	Inguinal hernia	Right	NA	Incarcerated	Acute appendicitis
15.	Yahya	2017	NA	NA	NA	NA	NA	NA	Acute appendicitis
Surgery		**Hernia repair**		**Approach to appendix**		**Histology**	**Tm size (mm)**	**Tm location**	**Recurrence (of neoplasm)**
16.	Open	Lichtenstein		Appendicectomy		Mucinous neoplasm	30	Tip	No (5 mo)
17.	Open	Primary		Appendicectomy		Mucinous adenocarcinoma	NA	NA	NA
18.	Open	Bassini		Appendicectomy		Goblet cell carcinoid	22	Tip	No (12 mo)
19.	Open	Bassini		Appendicectomy		Carcinoid	15	Tip	NA
20.	Open	Modified Bassini (d)		Appendicectomy		Mucinous adenocarcinoma	NA	Base	NA
21.	Open	Primary (c)		Appendicectomy		Adenocarcinoma	NA	NA	NA
22.	Laparoscopic	Internal ring closure (a)		Appendicectomy		Carcinoid	10	NA	NA

Continued.

S. No.	References	Year	Age (Years)	Sex	Preliminary diagnosis*	Hernia side	Hernia type	Hernia status	Intra-op findings
23.	Laparoscopic	Internal ring closure			Appendicectomy	Mucinous cystadenoma	2	NA	NA
24.	Open	Lichtenstein			Appendicectomy	Fibroma	30	NA	NA
25.	Open	Primary			Appendicectomy	Mucinous cystadenoma	NA	NA	NA
26.	Open	Shouldice			Appendicectomy	Villous adenoma	3	Base	No (12 mo)
27.	Open	Primary (e)			Appendicectomy	Carcinoid tumour	NA	Base	No (24 mo)
28.	Open	Lichtenstein			Appendicectomy	Mucinous cystadenoma	30	NA	NA
29.	Open	Bassini (b)			Appendicectomy	Adenocarcinoma	25	NA	NA
30.	NA	NA			NA	Goblet cell carcinoid	NA	NA	NA

*Preliminary diagnosis prior to radiological or intra-operative diagnosis, M: Male, F: Female, NA: Not Available, Tm: Tumour, mo: months, (a): transabdominal preperitoneal hernia repair with mesh was performed six weeks after the first operation, (b): Right hemicolectomy was performed a few days after the pathology report was approved, (c): Right hemicolectomy concurrently, (d): Ileocolic resection concurrently, (e): Right hemicolectomy was performed in the following operation.

Table 2: Review of the literature on cases of recurrent Amyand hernia.[8,9,13-15,25-28]

S. No.	Reference	Year	Age (In years)	Sex	Preliminary diagnosis*	Time previous hernia repair	Details previous hernia repair	Hernia side
1.	Bailon-Cuadrado	2016	89	M	Incarcerated recurrent hernia	40 years prior	NA	Right
2.	de la Hoz Rodriguez	2018	72	M	NA	15 years prior	Mesh repair	Right
3.	Grez	2018	59	M	Incarcerated recurrent hernia	10 years prior	Lichenstein	Right
4.	Kueper	2007	72	F	Incarcerated recurrent hernia	2 years prior	Shouldice repair	Right
5.	Lombardo	2012	47	M	Recurrent inguinal hernia	12 months prior	Bassini	Right
6.	McBride	2017	66	M	Incarcerated recurrent hernia	'As a child'	NA	Right
7.	Quartey	2012	71	M	Incarcerated recurrent hernia	17 months prior	Mesh repair (plug and patch)	Right
8.	Ranganathan	2011	80	M	Strangulated recurrent hernia	40 years prior	NA	Right
9.	Velimezis	2015	78	M	Incarcerated recurrent hernia	12 years prior	Primary repair	Right
Hernia type		**Hernia status**	**Intra-operative findings**		**Surgery**	**Hernia repair**	**Approach to appendix**	**Reoccurrence**
10.	Indirect	NA	Acute appendicitis (gangrenous)		Open	Shouldice	Appendicectomy	NA
11.	Indirect	NA	Acute appendicitis (perforated)		Open	Primary repair	Appendicectomy	NA
12.	Indirect	Incarcerated	Normal appendix		Open	Mesh repair	Appendicectomy	NA
13.	Indirect	NA	Acute appendicitis (perforated)		Open	Bassini	Appendicectomy	NA
14.	Indirect	Incarcerated	Normal appendix		Open	Modified Bassini	Appendicectomy	NA
15.	Indirect	Incarcerated	Acute appendicitis (perforated)		Open	Mesh repair (a)	Appendicectomy	NA
16.	Indirect	Incarcerated	Acute appendicitis		Open	Mesh repair	Appendicectomy	Nil reoccurrence at 5 mo
17.	Indirect	Incarcerated	Acute		Open	Lichenstein	Appendicectomy	NA

Continued.

S. No.	Reference	Year	Age (In years)	Sex	Preliminary diagnosis*	Time previous hernia repair	Details previous hernia repair	Hernia side
			appendicitis					
18.	Indirect	Incarcerated	Acute appendicitis		Open	Lichenstein	Appendicectomy	Nil reoccurrence at 36 mo

*: preliminary diagnosis prior to radiological or intra-operative diagnosis, M: Male, F: Female, NA: Not Available, mo: months, (a): orchiectomy performed due to herniated omentum inseparable from the right testicle.

Nine cases of Amyand hernia within a recurrent inguinal hernia were identified in the existing literature. Seven identified acute appendicitis, 3 of which were perforated, whilst one was gangrenous. No cases correctly identified an Amyand hernia prior to imaging or surgery. All cases were either incarcerated or strangulated, and all were managed with an open appendicectomy and hernia repair with or without mesh. As for previous hernia repairs, they were reported between 1 to 40 years prior to current presentation, and different repair techniques included primary and mesh repairs.

If not diagnosed pre-operatively, there is a risk of damage and perforation to the appendix during surgery. Lombardo and Pavone described an Amyand hernia with the appendix adhered to a previous mesh repair.[15] The pathophysiology of Amyand hernia in a recurrent inguinal hernia has not yet been proposed, however future research could further investigate the risk factors and clinical findings.

CONCLUSION

This study identified the first reported case of an Amyand hernia with a simultaneous neoplasm and recurrent inguinal hernia. Furthermore, the case was unusual due to the rarity of diagnosis in an elderly female, in addition to the infrequent complications of appendicitis, perforation, and abscess formation. This case defined the first report of a laparoscopic approach without hernia repair for the Amyand hernia within recurrent inguinal hernia. A review of the literature revealed most cases of neoplastic appendix or recurrent inguinal hernia to be associated with appendicitis. Furthermore, the diagnostic dilemma is evident as these cases reviewed did not correctly identified the Amyand hernia prior to imaging or surgery. A combination of thorough clinical examination and multiple imaging modalities should be considered in aiding diagnosis. Surgeons should be aware of the possibility of an appendix located within the inguinal sac and be prepared to manage this condition if it is encountered intra-operatively.

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