Case Report

**Obstructed Left Amyand's Hernia in a Seven Month
Male Infant with Meatal Stenosis: Case Report and
Review of Literature**

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ABSTRACT

Left Amyand Hernia (AH) is a very rare form of inguinal hernia. It presents commonly as an incarcerated inguinal hernia and intraoperative diagnosis is usually the rule. This is a report of a 7 months old male infant presenting with bilateral inguinal hernia, including a complicated left AH, with associated mobile caecum and meatal stenosis. He had bilateral herniotomy, transhernial appendectomy and meatotomy and did well post-operatively. A comprehensive comparative review of all 35 reported cases of left AH in the literature was undertaken alongside this report.

Keywords: Amyand's hernia, Left sided, Mobile caecum.

INTRODUCTION

Amyand's hernia is the presence of the appendix in an inguinal sac. This rare eponymous disease was first described in 1735 by Claudius Amyand, a surgeon working as surgeon to King George II¹ The appendix has also been found incarcerated in the femoral hernia in the rare case of De Garengot hernia.² The incidence of right Amyand's hernia stands at 1%.^{3, 4, 5} even rarer is a left Amyand's hernia. In a review of hernias in 2,159 in childhood, Mehmet Hanifi Okur *et al.*, reported an incidence of 0.97% (21) with all cases reported on the right.⁸ Amyand's hernia could occur in any age group as it has been reported in neonates and 92 year old.³ It is said to be three times more likely to be diagnosed in children than in adults. This has been ascribed to the patency of the processes vaginalis in this age group.⁶ The Appendix within the hernia sac may become inflamed, infected or perforated or

entirely healthy despite being incarcerated.^{7,8, 9,10} Pre-operative diagnose of Amyand's hernia is usually serendipitously at surgery.⁶ We report the 2nd case of left AH in sub-saharan Africa, and comparatively review the 35 reported cases of left AH in world literature.

CASE REPORT

A seven month old male infant presented with a six hours history of painful left hemi scrotal swelling associated with vomiting. Parents had noticed bilateral groin swellings that appear bigger when the child cries since birth. It was also noticed that the baby strains to urinate since birth. They had sought medical help in several facilities where he was treated

2018 Journal Impact Factor: 1.10
Print ISSN: 2636-7378 | Online ISSN: 2651-5865

for recurrent urinary tract infection and were reassured that his swelling will disappear as he grows older. Examination revealed an irritable dehydrated baby who was not pale or febrile. An irreducible tender left inguino-scrotal swelling was noted and reducible right groin swelling was noted. The phallus was circumcised and external urethral meatus was stenosed. No VACTERL associated anomaly was detected. He was resuscitated and had bilateral groin exploration and meatotomy. The findings were serous fluid within the left hernial sac which contained the caecum and an appendix that appeared grossly healthy measuring 7cm long (Fig 1). A transhernial appendicectomy and bilateral herniotomy was performed.

Fig 1: Appendix and caecum displayed on the left side at operation

He did well post operatively and was discharged on the third post-operative day. Follow up at two weeks, one month and three months later revealed a healthy baby with well healed groin scars and a patent external urethral meatus.

DISCUSSION

The first reported case of an appendix within an inguinal hernia sac is credited to the British surgeon Claudius Amyand¹ who in 1735 performed an appendectomy and herniorrhaphy on an 11year boy, with this rare form of hernia. It is appropriately named after him. Though the appendix in this pioneer case was perforated, the term Amyand hernia in current usage describes the presence of the appendix, (normal inflamed, perforated or

gangrenous), within the sac of an inguinal hernia.^{6, 11} The incidence of AH is placed at 0.19% - 1.7% of reported inguinal hernia cases.⁵ They are far commoner on the right, due to the normal anatomical position of the Appendix on the right, and the fact that inguinal hernias are twice more likely to be on the right.¹² A thorough search of literature revealed only 35 reported case of left Amyand hernia including ours (Table 2). However some details were not accessible in all the reported cases. A left sided Amyand's hernia occurs as a result of situs inversus, malrotation of the intestine or a very large mobile caecum or appendix.^{2,3,4,13,14} In our review of the 35 cases, mobile caecum seen in 19(59.4%) patients was the commonest identifiable anatomical predisposition. While one patient had situs inversus, predisposing factors in the rest could not be ascertained despite retrospective investigations with ultrasonography, chest and abdominal x-rays. Six patients however were not investigated. The mobile caecum as part of the mobile caecum syndrome, is characterized by an abnormal or non-fixation of the caecum and ascending colon to the posterior abdominal wall. This condition is reported to be present in 10-20% of the population.¹⁴ The displaced appendix facilitates an AH, and more so a left-sided AH. Another documented surgical complication of a mobile caecum is caecal volvulus.^{15, 34} The mobile caecum was the identified predisposing factor in our case. Reports indicate that AH is three times commoner in children because of the higher incidence of a patent processus vaginalis in these individuals.⁵ Our review of left-sided AH revealed an age range of 2 months – 81years. Nineteen patients (57.6%) were less than 16 years, giving a child to adult ratio of 1.36:1. All patients in the cases reviewed were males.

AH usually present with clinical features in keeping with incarcerated inguinal hernia.^{6,33} Being a clinical diagnosis, diagnostic imaging studies is not routinely employed preoperatively. This invariably makes intraoperative diagnosis the rule.^{11, 13, 23,31,34,38} In all but two cases reviewed, the diagnosis of left AH was made intraoperatively. One of the pre-operative diagnoses was made by ultrasonography and the other, incidentally by barium enema study. There are however reports of pre-operative diagnosis of AH by some investigators in the literature.^{6,17}

Lasanoff and Basson classification of AH based on the pathologic state of the appendix and associated structures, also proposed appropriate treatment options for the various types (Table 1).^{18,19} Sixteen appendices (51.6%) in this review were normal (Type 1 AH), with consequent seven appendectomies.

The incongruence between the pathological description and consequent treatment option suggested by Lasanoff and Basson, may be due to one or a combination of; an equivocal

Table 1: Classification of Amyand's hernias after Losanoff and Basson^{15, 16}

Classification	Description	Surgical management
Type 1	Normal appendix within an inguinal hernia	Hernia reduction, mesh repair, appendectomy in young patients
Type 2	Acute appendicitis within an inguinal hernia, no abdominal sepsis	Appendectomy through hernia, primary endogenous repair of hernia, no mesh
Type 3	Acute appendicitis within an inguinal hernia, abdominal wall or peritoneal sepsis	Laparotomy, appendectomy, primary repair of hernia, no mesh
Type 4	Acute appendicitis within an inguinal hernia, related or unrelated abdominal pathology	Manage as types 1 to 3 hernia, investigate or treat second pathology as appropriate

Table 2: Summary of reported cases of Left-sided Amyand hernia.

	Author	Year	Sex	Age	Diagnosis	Type	Predisposing condition	Treatment	Outcome
1	Carey LC ²¹	1967	N/A	N/A	N/A	N/A	N/A	N/A	N/A
2	Bakhshi GD ²²	2004	M	35yrs	IO	2	Not Identified	Appendectomy /Herniorrhaphy	Uneventful
3	Gupta S ²³	2005	M	9m	IO	1	None Identified	Herniotomy	Uneventful
4	Breitenstein S ²⁴	2005	N/A	N/A	IO	N/A	None Identified	Appendectomy/Herniorrhaphy	Uneventful
5	Gupta N ¹¹	2007	M	11m	IO	1	None Identified	Appendectomy/Herniotomy	Uneventful
6	Tayade MB ²⁵	2008	M	34yrs	IO	4	Mobile caecum	Hemicolectomy/ Anastomosis	Uneventful
7	Johari HG ²⁰	2009	M	70yrs	IO	1	Mobile caecum	Appendectomy/Herniorrhaphy	Uneventful
8	Kaymakci A ²⁶	2009	N/A	Paed.	IO	1	Mobile caecum	Herniotomy	Uneventful
9	Kaymakci ²⁶	2009	N/A	Paed.	IO	2	Mobile caecum	Appendectomy/Herniotomy	Uneventful
10	Kaymakci A ²⁶	2009	N/A	Paed.	IO	1	Mobile caecum	Herniotomy	Uneventful
11	Ravishankaran P ²⁷	2010	M	35yrs	IO	1	None identified	Appendectomy/Herniorrhaphy	Uneventful
12	Malik KA ²⁸	2010	M	64yrs	IO	4	Mobile caecum	Hemicolectomy/Herniorrhaphy	Uneventful
13	Cankorkmaz ²⁹	2010	M	4m	IO	1	Mobile caecum	Appendectomy/ Herniotomy	Uneventful
14	Cankorkmaz ²⁹	2010	M	2m	IO	1	Not known	Appendectomy/ Herniotomy	Uneventful
15	Khan RA ³⁰	2011	M	10m	IO	2	Mobile caecum	Appendectomy/Herniotomy/cecopexy	Eventful
16	Singh K ³¹	2011	M	1.5yrs	IO	4	Mobile caecum	Herniotomy/Repair of caecal perforation.	Uneventful
17	Singh K ³¹	2011	M	1.5yrs	IO	1	Not known	Herniotomy	Uneventful
18	Ghafour A ³²	2012	M	60yrs	IO	1	Not known	Hernioplasty	Uneventful
19	Pun A ³³	2013	M	18m	IO	2	Mobile caecum	Appendectomy/ Herniotomy	Uneventful
20	Mewa Kinoo ⁶	2013	M	60	PO	3	Not known	Appendectomy/ Herniorrhaphy	Uneventful
21	Al-Mayoof ³⁴	2013	M	4m	IO	2	Situsinversus	Appendectomy/ Herniotomy	Uneventful
22	Al-Mayoof ³⁴	2013	M	10m	IO	1	Mobile caecum	Appendectomy/ Herniotomy	Uneventful
23	Gupta R ³⁵	2013	M	11m	IO	2	Mobile caecum	Appendectomy/ Herniotomy	Uneventful
24	Noubar K ³⁶	2013	M	59yrs	IO	1	Mobile caecum	Hernioplasty	Uneventful
25	Khalid H ¹³	2014	M	40yrs	IO	2	Mobile caecum	Appendectomy/ Herniorrhaphy	Uneventful
26	Dong B ³⁷	2014	M	63yrs	IO	2	Mobile caecum	Appendectomy /Hernioplasty	Uneventful
27	Maeda K ¹⁶	2014	M	62yrs	PO	1	Mobile caecum	Hernioplasty	Uneventful
28	Mongardni M ³⁸	2015	M	68yrs	IO	4	Mobile caecum	Appendectomy/ Hernioplastynon mesh.	Uneventful
29	Cigsar E ³⁹	2015	N/A	Paed.	N/A	N/A	N/A	N/A	N/A
30	Cigsar E ³⁹	2015	N/A	Paed.	N/A	N/A	N/A	N/A	N/A
31	Sharma D ⁴⁰	2015	M	60yrs	IO	4	Not known	Hemicolectomy/ Herniorrhaphy	Uneventful
32	Yoneyama ⁴¹	2015	M	8m	IO	1	Not Identified	Herniotomy	Uneventful
33	Al-Maksoud A. ⁴²	2015	M	81yrs	IO	1	Mobile caecum	Hernioplasty	Uneventful
34	Nwagbara ⁷	2016	M	1m	IO	4	Not known	Appendectomy/Caecal repair/ Herniorrhaphy	Uneventful.
35	Our case	2016	M	7m	IO	1	Mobile caecum	Appendectomy/ Herniotomy/ meatotomy	Uneventful

Key: M = Male, m = months, yrs = years, IO = intraoperative, PE = Preoperative, Paed = Paediatric (age not specified), N/A = Not Accessible.

macroscopic appearance of the appendix and possible future diagnostic dilemma due to atypical clinical presentation of an abnormally situated appendix.

Consequently, Johari suggested appendectomy even in the presence of a non-inflamed appendix for all left sided AH.²⁰ The subject of our case report, who had a Lasanoff-Basson Type 1 left AH, had appendectomy. Despite the presence of local sepsis in more than half (i.e. Types 2, 3 and 4) of the reported cases, the outcome of treatment was overwhelmingly uneventful.^{6,7,11,14,15,21-26,28-37,39-42} This is probably due to perioperative antibiotic use and adherence to the surgical principle, which discourages the use of prosthetic mesh for hernioplasty in the presence of sepsis.

Meatal stenosis is mostly an acquired condition seen in 9-10% of circumcised males.⁴³ It is a known cause of bladder outlet obstruction, associated with straining and raised intraabdominal pressure.

This, in the presence of a patent processus vaginalis, would probably contribute to the bilateral inguinal hernia seen in our report. The incidence of bilateral inguinal hernia is otherwise low, reportedly 10%.⁴⁴ Meatotomy as performed in our reported case is therapeutic for this condition and eliminates a known risk factor for recurrence of hernia.

CONCLUSION

Left sided AH remains a very rare condition, with a reported 100% male predominance and occurring in approximately equal proportions between children and adults. Its diagnosis is invariably intraoperative and has mobile caecum as the commonest predisposing factor. The appendix is inflamed in about half of the cases, and Losanoff-Basson type 1 is the commonest class. Appendectomy for type 1 left-sided AH is an acceptable option, as it guards against future diagnostic dilemma arising from an atypical clinical presentation.

Conflict of Interest

None declared.

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