THE TRAILING APPENDIX IN A NIGERIAN INFANT: A CASE REPORT AND REVIEW OF LITERATURE.

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ABSTRACT

Introduction: **Amyand hernia is described as an inguinal hernia in which the hernia sac contains the appendix[1]. Its incidence is rare, although common in children, and it is often an incidental finding at surgery[2,3]. While the management widely depends on the Lasanoff and Basson guidelines, the treatment plan, however, remains controversial.**

Case Presentation: **A two-month-old male child with a history of progressively increasing right groin swelling noticed since birth. Examination revealed a right reducible inguinoscrotal swelling, which was separate from the right testis with a positive silk glove sign. A diagnosis of a right reducible inguinoscrotal hernia was made, and he was scheduled for a right herniotomy. Intra-operatively, after reduction of the contents of the hernia sac, the vermiform appendix was seen peeping out of a rent in the wall of the hernia sac. The appendix was returned to the peritoneal cavity, and a Marcy repair was done to complement the herniotomy. The post-operative period was uneventful.**

Conclusion: **Amyand hernia is a rare clinical condition reported more commonly in children, whose diagnosis remains fortuitous, and the treatment plan, controversial. It is often an intraoperative diagnosis, and the management plan for this condition varies, depending on the status of the appendix at surgery. In large inguinoscrotal hernias in males, we recommend that the content of the sac be examined before transection, and a Marcy's repair should be considered.**

Keywords: Amyand, Inguinal hernia, Children, Nigerian, Appendix

INTRODUCTION

Amyand hernia is described as an inguinal hernia with the hernia sac having the vermiform appendix as part of its contents[1]. The first description of this eponymous diagnosis occurred simultaneously with the first report of appendicectomy as described by French-born English Surgeon, Claudius Amyand, in 1735[4]. This was reported while operating on an 11-year-old male who had presented with a suppurating right inguinoscrotal hernia, discovering a ruptured appendix in the hernia sac [4,5].

Amyand hernias account for 0.19% to 1.7% of inguinal hernias[1,2,6]. Even rarer are related peculiar complications such as appendicitis in the inguinal sac, accounting for 0.07–0.13% of all cases of Amyand hernia, and 2% of appendectomies in children[1,7]. Amyand hernia is three times more common in children than adults[2,6]. The diagnosis is usually fortuitous because identifying the appendix as a content of the hernia sac is often an incidental finding at surgery[1,2,8]. The management is dependent on intraoperative findings. An uninflamed appendix is often reduced and a herniotomy or herniorrhaphy performed, while an inflamed appendix might require appendicectomy or intraperitoneal exploration when complicated[2,6]. The outcome depends largely on the state of the appendix at surgery. While most surgeons are aware of the eponymous diagnosis, the guidelines of management are often not well known by most surgeons, and this report hopes to serve as a reminder and guide for this condition. We present the case of an eight-week-old male managed for a right Amyand hernia with an uninflamed vermiform appendix.

Case Presentation

A two-month-old male child was brought by his mother to the Paediatric Surgery Outpatient Clinic with complaints of a right groin swelling noticed since birth.

The swelling was initially small and becomes more visible when crying. The swelling subsequently became conspicuous at all times and progressively increased in size about four weeks prior to presentation. There was no history of feeding intolerance, vomiting, inconsolable cry, abdominal distension, or constipation. There was no similar swelling in the left groin. Abdominal examination revealed a right reducible inguinoscrotal swelling, which was separate from the right testis with a positive silk glove sign. It was non-tender, had no differential warmth. Both testes were intra-scrotal. There was no abdominal distention and no palpable intra-abdominal mass. A diagnosis of a right reducible inguinoscrotal hernia was made. Parents were counselled on the diagnosis. The pre-anaesthetic review revealed an American Society of Anaesthesiologists, class I (ASA I) patient.[9] Informed consent was obtained for the procedure from the parents, and the child subsequently had a right herniotomy under general anaesthesia.

Figure 1: Intraoperative image of the ruptured right inguinal hernia sac showing the protruding vermiform appendix in the hernia sac

Following skin preparation and application of sterile drapes to isolate the right groin, a 4 cm transverse incision was made in the right groin, and blunt dissection was used to expose the right spermatic cord structures. The hernia sac was identified, the contents of the hernia sac were reduced, and the hernia sac was isolated. While attempting to isolate the hernia sac, there was an inadvertent rent in its wall, and the trailing appendix was seen as a content of the sac (figure 1).

Table 1: Losanoff and basson classification of amyand hernia[20]

Types	Features	Surgical Management
I	Normal appendix within an inguinal hernia	Depending on the patient's age, either a reduction or an appendicectomy will be performed, followed by a hernioplasty using mesh.
II	Acute appendicitis localized within a hernia sac (no abdominal sepsis)	To treat a hernia, the surgeon will remove the appendix through the same incision, and repair the hernia without using any mesh.
III	Acute appendicitis within an inguinal hernia sac along with peritoneal sepsis	After removing the appendix via a large incision in the abdomen, the surgeon will repair the hernia directly without using mesh.
IV	Acute appendicitis within an inguinal hernia along with other related or unrelated abdominal pathology	The patient will undergo an appendicectomy, and additional tests and treatments will be done based on their symptoms

Table 2: Profile of case reports of amyand hernia in Nigerian children

S/No	Author (year)	Age	Sex	Right /Left	Classification of Amyand hernia	Diagnosis	Surgery Done
1.	Nwagbara et al (2016)[13]	18 months	M	Left	Grade III	Left Strangulated Inguinoscrotal Hernia	Appendicectomy + repair of caecal wall perforation + herniotomy
2.	Ogwuche et al (2019)[14]	7 months	M	Left	Grade I	Obstructed left Inguinoscrotal hernia + reducible right inguinoscrotal hernia + meatal stenosis	Bilateral herniotomy + Appendicectomy + meatoplasty
3.	Ogbetere (2021)[28]	15 years	M	Right	Grade I	Right Reducible Inguinoscrotal Hernia	Appendix returned into peritoneal cavity + herniotomy
4.	Iluobe et al (2022)[29]	4 Weeks	M	Right	Grade II	Right Irreducible Inguinoscrotal hernia + Right Undescended Testes	Appendicectomy + herniotomy

The appendix was examined and was found to be normal. It was returned to the peritoneal cavity, and the hernia sac was transected and transfixed at the deep inguinal ring. Marcy repair was done to narrow the deep inguinal ring. The wound was closed in layers. A firm groin dressing and scrotal support were applied. The post-operative period was uneventful. The patient was discharged home three hours postoperatively. He was followed up at the surgery outpatient clinic and had no complaints.

Discussion

Amyand hernia is a rare presentation in patients with inguinoscrotal hernia. It has been reported in all age groups, with a bimodal peak observed in children and adults older than 70 years.[2] Yet, it is three times more common in children.[2,6] Its increased incidence in children and the elderly is related to factors that promote a widened internal inguinal ring[1,6,8]. In children, the presence of a patent processus vaginalis is central to this.[2,6] Amyand hernia has been reported in children as early as the third week of life.[1,8,10] The patient reported here is a two-month-old infant, but the swelling had been noticed from the first week of life. As in this case, Amyand hernias are usually on the right, as the vermiform appendix has its normal anatomical position on the right side of the body, and inguinal hernias more commonly occur on the right side.[6,11] However, left-sided Amyand hernias have been reported in some patients, and this has been attributed to the presence of conditions like situs inversus, intestinal malrotation, and mobile caecum.[6,7,12-14]

Patients with Amyand hernia may suffer any of the known complications of inguinal hernia, and in addition, can also develop acute appendicitis in the hernia sac which may be complicated, presenting as a ruptured appendix or an appendiceal abscess.[15] Soilage from complicated acute appendicitis in an Amyand hernia may be limited to the inguinal canal, due to the presence of the inguinal ring[7] or it may extend into the peritoneal cavity.[6] The exact pathophysiology leading to an inflamed appendix within the hernia sac remains unknown; however, several studies have proposed the repeated occlusion of the appendiceal lumen by the deep inguinal ring with increased intraluminal pressure, stasis, and bacterial proliferation.[2,6] Also, repeated microtrauma due to recurrent herniation and reduction of the hernia with associated adhesions between the contents of the hernia sac and the hernia sac.[7,12]

Pre-operative clinical diagnosis is practically impossible, but has been reported via ultrasonography, magnetic resonance imaging (MRI), or computed tomography (CT) scan as a tubular blind-ended structure originating from the caecal wall and extending into the hernia sac.[6,8,16] An ultrasound scan, MRI, or CT scan is, however, not required for diagnosis in most patients with inguinal hernias, as diagnosis is clinical.[17-19] Consequently, the likelihood of reaching a pre-operative diagnosis of an Amyand hernia is rare, and so it is usually diagnosed intraoperatively.[6,12] In this patient, a scrotal ultrasound scan was done prior to presentation at our outpatient clinic, but it did not identify the appendix as a content of the hernia sac.

Losanoff and Basson, in 2007, categorized Amyand's hernia into four subtypes depending on the status of the appendix in addition to their surgical approach (Table 1).[20] While Losanoff and Basson, in their classification of Amyand hernia, provided a standardized approach to its management, the treatment plan, however, remains controversial, particularly that of Type 1 as seen in our patient.

Many surgeons do not recommend prophylactic appendicectomy where a non-inflamed vermiform appendix is encountered within the hernia sac.[6] This is proposed because performing an appendicectomy in this category of patients increases the risk of surgical site infection complicating an otherwise clean surgical procedure.[12,20] Additionally, recent findings reveal that the vermiform appendix plays a useful role in modulating the immunity of the gut, especially in children, and a missing appendix may result in an increased incidence of gastrointestinal illnesses.[21,22] Furthermore, it has been found useful in the treatment of other conditions, such as the Mitrofanoff procedure.[23] However, for reasons, including high risk of recurrence of hernia, and a future likelihood of appendicitis presenting with atypical features, other surgeons favour a prophylactic appendicectomy, particularly in cases of a left-sided Amyand hernia.[6,12,24] In our patient, the appendix was not inflamed and was existing in a non-obstructed right inguinal hernia. This was treated as a case of Type 1 Amyand hernia, and appendicectomy was not done. The appendix was returned into the peritoneal cavity, a herniotomy was done, and narrowing of the internal ring was done as well. Narrowing of the internal ring in patients with a widened internal inguinal ring has been shown to reduce recurrence[19,25-27], however, concerns of strangulation of spermatic cord structures exist.[25]

A literature search on reported cases of Amyand hernia in children in Nigeria was conducted. A total of four cases were found to have been reported so far. The summary is presented in Table 2. In 2019, Ogwuche *et al.* noted that only two cases of Amyand hernia in Nigerian children had been reported in the literature (from 35 case reports of left Amyand hernia identified worldwide). From our literature search, two

more cases of Amyand hernia in Nigerian children were identified (Table 2). This report is thus the fifth reported case in a Nigerian child. A reporting system for uncommon pathologies, such as Amyand hernia, should be put in place to understand the true incidence of these clinical entities.

CONCLUSION

Amyand hernia is a rare clinical condition reported more commonly in children, whose diagnosis remains fortuitous, and the treatment plan, controversial. It is often an intraoperative diagnosis, and the management plan for this condition varies, depending on the status of the appendix at surgery. In large inguinoscrotal hernias in males, we recommend that the content of the sac be examined before transection, and a Marcy's repair should be considered.

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