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·临床研究·

成人Amyand疝患者临床特征的系统评价

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摘 要 **背景与目的：**Amyand疝是一种特殊类型的腹股沟疝，目前并无统一的处理规范。本研究旨在通过系统评价其临床特征及相关手术决策，为降低该病的并发症和改善患者预后提供依据。

方法：计算机检索多个国内外数据库，收集自2000年1月1日至今公开发表的成人Amyand疝的相关文献。根据纳入与排除标准，由2名研究员独立筛选文献、提取相关资料、评估偏倚并交叉核对后行系统评价。

结果：最终纳入141篇文献，共计184例成人Amyand疝患者。中国为发文量最多的国家，而刊文量最多的期刊为Hernia。在临床特征方面，男性占91.8%，其中位发病年龄为60岁，而女性中位发病年龄为69岁；89.1%的Amyand疝发生于右侧，8.9%的患者是复发疝；75例患者未报告相关信息，余109例患者中有104例斜疝，4例直疝，1例马鞍疝；25.7%（45/175）的患者于术前诊断，其中超声准确率为23.1%（6/26），CT准确率为75.0%（33/44），超声联合CT的准确率为62.5%（5/8），1例通过灌肠诊断。治疗方面，42例患者行择期手术，122例行急诊手术，20例未获得相关信息；45.6%的患者行无张力疝修补术，50%的患者行传统疝修补术，4.4%的患者未做修补或择期手术修补；80.9%的患者切除阑尾，其余患者均保留阑尾；140例术中表现为Losanoff和Basson 1、2型，35例为3、4型，其中12例合并阑尾肿瘤。结局方面，术后中位住院时间为4 d，125例患者获随访，中位随访时间为6个月，17例出现并发症（肺部感染或栓塞性疾病6例，手术部位感染5例，血清肿3例，复发3例，尿潴留1例），均为急诊手术患者。

结论：成人Amyand疝是一种特殊类型的腹股沟疝，与嵌顿性或绞窄性疝难以鉴别，常需急诊手术，术中应根据具体情况个体化治疗，以不引起植入的补片感染为基本原则。应鼓励更多Amyand疝的基础和临床研究，以提高Amyand疝的诊疗水平及改善患者预后。

关键词 疝，腹股沟；Amyand疝；疝修补术；阑尾切除术；系统评价

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Clinical characteristics of adult Amyand's hernia: a systematic review

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Abstract **Background and Aims:** Amyand's hernia is a special type of inguinal hernia, and there is currently no

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uniform treatment standard. This study was conducted to provide a basis for reducing complications of this condition and improving prognosis of the patients by systematically evaluating its clinical characteristics and related surgical strategies.

Methods: The studies concerning Amyand's hernia in adults published since January 1, 2000 to date were collected by comprehensively searching several domestic and foreign online databases. After literature screening, relevant data extraction, bias assessment and crosschecking by two independent reviewers according to the inclusion and exclusion criteria, a systematic review was performed.

Results: A total of 141 studies were included involving 184 adult patients with Amyand's hernia, and the most majority of papers was contributed by the authors from China and the highest number of papers was published by the journal of *Hernia*, respectively. In terms of clinical features, male patients accounted for 91.8%, with a median onset age of 60 years, and the median onset age in women was 69 years; 89.1% of them had a right hernia and 8.9% had a recurrent hernia; a total of 75 patients failed to access the relevant information; the remaining 109 patients consisted of 104 indirect hernias, 4 direct hernias, and 1 saddle hernia; 25.7% of the patients were diagnosed before surgery, and the diagnostic accuracy of ultrasound, CT and their combination were 23.1% (6/26), 75.0% (33/44), and 62.5% (5/8), and one case was diagnosed by enema. With regard to the treatment, 42 patients underwent elective procedures, 122 cases received emergency operation, and 20 cases had no treatment information; 45.6% of cases underwent mesh repair, 50% of cases underwent endogenous repair, and 4.4% of cases did not undergo repair or delayed repair; 80.9% of cases underwent appendectomy and the appendix was preserved in remaining of them; there were Losanoff and Basson type 1 or 2 lesion in 140 cases and type 3 or 4 lesion in 35 cases verified by intraoperative findings, and 12 cases had combined appendix tumors. As for the outcomes, the median length of postoperative hospital stay was 4 d, a total of 125 patients were followed up with a median time of 6 months, and 17 cases had complications (6 cases of pulmonary infection or embolic disease, 5 cases of surgical site infection, 3 cases of seroma, 3 cases of recurrence, 1 case of urinary retention), all of them were emergency surgery patients.

Conclusion: The adult Amyand's hernia is a rare type of inguinal hernia, which is difficult to distinguish from the incarcerated or strangulated hernia. It usually requires an emergency surgery. The treatment of Amyand's hernia should be individualized based on intraoperative conditions, with the fundamental principle of avoiding the infection of the implanted mesh. Further basic and clinical research on Amyand's hernia should be encouraged for improving the treatment efficacy and the prognosis of the patients.

Key words Hernia, Inguinal; Amyand's Hernia; Herniorrhaphy; Appendectomy; Systematic Review

CLC number: R656.2

男性患腹股沟疝的风险为 27%~43%，女性为 3%~6%，而腹股沟疝修补术也是普外科最常见的手术之一[1-2]。Amyand 疝是一种特殊类型的腹股沟疝，因纪念英国学者 Claudius Amyand 而命名，其疝内容物为阑尾。Amyand 疝临床罕见，发病率约为 1%，合并阑尾炎的概率更低，约为 0.07%~0.13%[3-4]。Amyand 疝缺乏特异性的临床症状、体征及相关影像学特征，很难与嵌顿性或绞窄性疝相鉴别，极易造成术前误诊和术中漏诊，影响进一步治疗和预后。目前有关 Amyand 疝的研究多为病例报道或病例系列，缺乏高质量的系统评价证据，对于最佳的管理办法仍然没有共识。手术医生需要了解 Amyand 疝的相关知识，但分散的文献使得这一过程变得十分耗时费力。因此，本文通过系统综述领域内重要文献，归纳其流行病学、临床表现、诊疗过程和预后特点，重点关注其手术决策，以期为临床诊治提供帮助。

1 资料与方法

1.1 纳入与排除标准

纳入标准：(1) 研究对象为成人（年龄>18岁）Amyand疝确诊病例；(2) 手术治疗为主要干预措施；(3) 具有较为完整的临床相关特征及结局指标；(4) 研究类型，如病例报道或病例系列、横断面研究、队列研究、病例对照研究。排除标准：(1) 重复发表的文献；(2) 非中、英文文献。

1.2 文献检索策略

计算机检索PubMed-Medline、Embase、中国知网、维普和万方数据库，检索时限均为2000年1月1日至2021年5月20日。采用主题词与自由词相结合的方式进行，中文检索词包括：Amyand疝、疝、腹股沟、阑尾、阑尾炎；英文检索词包括：Amyand、Amyand Hernia、Appendix、Appendicitis、Inguinal Hernia。

1.3 文献筛选与资料提取

由2名研究员独立进行文献筛选、资料提取和交叉核对工作，如遇分歧，则由第三方仲裁。去除重复文献后，通过阅读标题和摘要初筛，排除明显不相关的文献。根据纳排标准进一步阅读全文，以确定是否最终纳入。如有必要，通过电话、邮件等方式联系作者予以补充相关信息。资料提取内容主要包括：(1) 纳入研究的基本特征，包括研究类型、题目、第一作者、出版年份、样本量、样本来源等；(2) Amyand疝患者相关的临床特征及结局指标，如：人口学特征（年龄、性别、疝病分型），术前实验室及影像学检查（白细胞计数、超声及CT检查结果），手术相关变量（手术指征、术中Losanoff和Basson分类[5]、手术方式），结局变量（术后病理结果、并发症、病死率、术后住院日、随访时间）；(3) 偏倚风险评价的相关要素。

1.4 纳入研究的偏倚风险评价

使用澳大利亚循证护理中心（Joanna Briggs institute，JBI）评价工具评估病例报告和病例系列的偏倚风险[6]。该工具从纳入研究的人口统计学特征、病史、临床表现、诊断及评估方式、干预或治疗措施、干预后的情况、不良事件及是否对类似病例有启示等8个方面进行评价，每回答一个“是”计1分。

1.5 统计学处理

使用EndNote X9、Excel 2010和SPSS 25.0软件进行相关文献管理、数据收集整理和统计分析工作。采用均数±标准差（$\bar{x} \pm s$）和中位数（四分位间距）[M（IQR）]来分别描述正态分布和非正态分布的计量资料，用频数（百分比）[n（%）]来描述分类资料。用两独立样本t检验或秩和检验进行计量资料的组间比较，χ^2检验或Fisher确切概率检验行分类资料的组间比较。P<0.05为差异有统计学意义。

2 结 果

2.1 文献筛选流程图

检索相关数据库后共获得文献1 965篇，剔除重复文献后获得1 582篇，阅读标题和摘要去除明显不相关文献后获得428篇，进一步阅读全文后，最终纳入定性分析141篇（病例报道128篇[7-134]，病例系列10篇[3-4, 135-142]，病例评价3篇[143-145]）。文献筛选流程图及结果见图1。

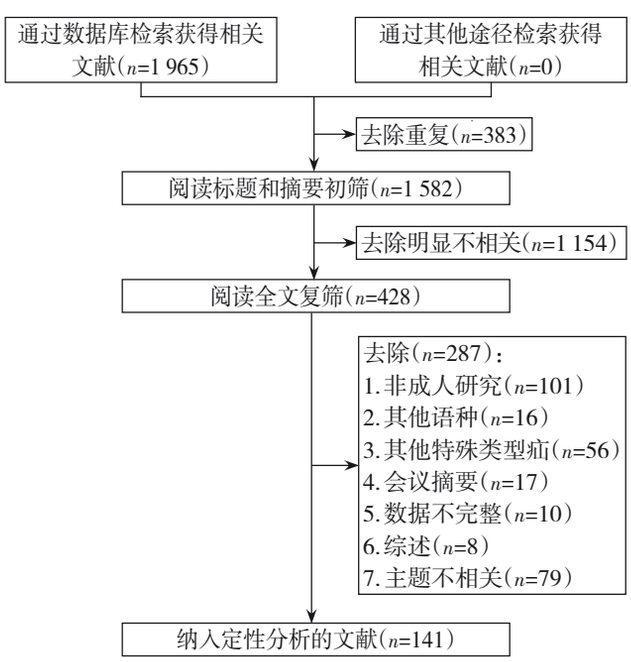

图1 文献筛选流程图

Figure 1 Literature screen process

2.2 纳入研究的偏倚风险评价结果与基本特征

各研究的偏倚风险评价结果显示：51%的（n=72）文献评分为8分，31%的（n=44）文献评分为7分，13%（n=18）的文献评分为6分，5%（n=7）的文献评分不小于4分，均为高质量研究。文献基本特征分析结果显示：(1) 发文量最多的国家是中国（26篇），

其次是美国（20 篇）、印度（17 篇）、希腊（14 篇）、土耳其（13 篇）和英国（11 篇），其它国家发文量均在 10 篇以下；(2) 发文高峰在 2011—2020 年，以 2015 年（19 篇）和 2020 年（17 篇）为最多；(3) 刊文量最大的期刊是 Hernia（19 篇）和 International Journal of Surgery Case Reports（15 篇）（图 2）。

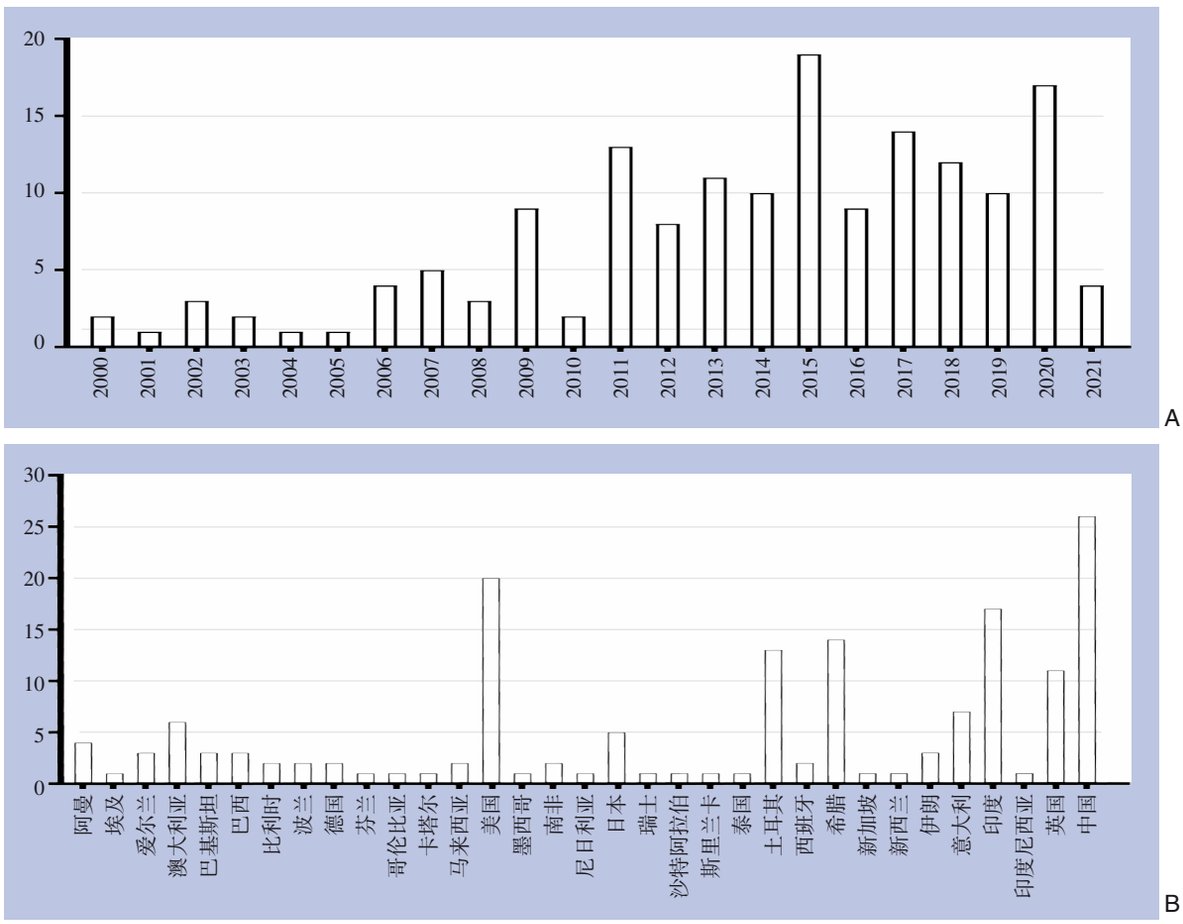

图 2　Amyand 疝研究文献的分布情况　A：时间分布；B：地区分布

Figure 2　Distribution of studies concerning Amyand's hernia　A: Temporal distribution; B: Regional distribution

2.3　Amyand 疝患者的临床特征分析

2.3.1　临床特点　本研究共纳入 184 例成人 Amyand 疝患者，其临床特点：(1) 男性占比为 91.8%（169/183），男女比例为 12∶1（1 例未报告性别）；(2) 中位发病年龄为 61 岁（年龄跨度为 20~92 岁），男性发病年龄为 60 岁，女性发病年龄为 69 岁；(3) 89.1%（164/184）的 Amyand 疝发生于右侧；8.9% 的患者（16/179，5 例未报告）是复发疝；75 例患者未报告相关信息，余 109 例患者有 104 例斜疝，4 例直疝，1 例马鞍疝。

2.3.2　辅助检查　(1) 27.7% 的患者（51 例）术前白细胞计数正常，而 33.2% 的患者（61 例）白细胞计数升高，72 例患者未提及相关数据；(2) 25.7%（45/175）的患者于术前确诊为 Amyand 疝，其中 CT 准确率为 75.0%（33/44），超声准确率为 23.1%（6/26），超声联合 CT 的准确率为 62.5%（5/8），灌肠的准确率为 100.0%（1/1）。

2.3.3　治疗和结局　归纳如下：(1) 42 例患者行择期手术，122 例行急诊手术，20 例未获得相关信息；(2) 择期患者手术指征包括可复性疝 16 例（38.1%），难复性疝 17 例（40.4%），术前怀疑或诊断为 Amyand 疝 7 例（16.7%），急诊患者手术指征包括难复性疝 7 例（5.7%），嵌顿性或绞窄性疝 62 例（50.9%），Amyand 疝 37 例（30.4%），肠梗阻或肠穿孔 7 例（5.7%），阑尾炎或腹膜炎 6 例（4.9%），脓毒症 2 例（1.6%），急性睾丸扭转 1 例（0.8%）；(3) 82.9% 的患者（150/181）行开腹手术（119 例经腹股沟切口，5 例经腹部切口，10 例经腹股沟切口联合腹部切口，16 例未获得相关资料），21 例完全经腹腔镜手术，10 例行腹腔镜联合开腹手术治

疗（9例联合腹股沟切口，1例联合腹部切口）；⑷ 45.6%的患者（83/182）行无张力疝修补术，50%（91/182）的患者行传统疝修补术，4.4%的患者（8/182）未做修补或择期手术修补；80.9%的患者（148/183）切除阑尾，余35例保留阑尾；⑸ 140例术中表现为Losanoff和Basson 1、2型，35例为3、4型；⑹ 术后病理提示56例为正常阑尾，113例为急性阑尾炎，14例合并腹部其他病理改变（阑尾类癌5例，阑尾良性腺瘤2例，阑尾腺癌1例，阑尾黏液性肿瘤1例，阑尾纤维瘤1例，盲肠腺癌1例，盲肠管状腺瘤1例，坏死性筋膜炎1例，坏疽性盲肠1例）；⑺ 125例患者获随访，中位随访时间为6个月，术后中位住院时间为4 d，17例出现术后并发症（肺部感染或栓塞性疾病6例，手术部位感染5例，血清肿3例，复发3例，尿潴留1例），均为急诊手术患者；⑻ 择期手术和急诊手术的治疗特点不同，急诊手术患者具有术前白细胞计数异常、术前诊断率高、术中补片放置率低、阑尾切除率高、开腹手术比例高、术中Losanoff和Basson 1型比例低、术后平均住院日时间长、术后并发症多等特点（均$P<0.05$）（表1）。

表1 急诊和择期手术患者的治疗特点

Table 1 Comparison of treatment characteristics between patients undergoing emergency and elective operation

项目	择期	急诊	P
男性[n(%)]	40(95.2)	111(91.7)	0.454
年龄[岁,M(IQR)]	60.0(34.8~71.8)	62.0(42.0~77.5)	0.206
白细胞数升高[n(%)]	1(5.0)	59(67.0)	<0.05
右侧疝[n(%)]	36(85.7)	112(91.8)	0.251
斜疝[n(%)]	34(97.1)	61(95.3)	>0.05
复发疝[n(%)]	3(7.1)	13(10.7)	0.508
术前已确诊[n(%)]	6(14.3)	37(31.4)	0.032
超声	0(0.0)	6(5.1)	
CT	5(11.9)	26(22.0)	
超声+CT	1(2.4)	4(3.4)	
其他方式	0(0.0)	1(0.9)[1)]	
疝修补方式[n(%)]			
无张力疝修补术	34(81.0)	34(28.3)	
传统疝修补术	7(16.7)	79(65.8)	<0.05
未做疝修补术	1(2.4)	7(5.8)	
阑尾切除[n(%)]	27(64.3)	107(88.4)	<0.05
手术方式[n(%)]			
腹腔镜手术	10(23.8)	10(8.2)	
开腹手术	30(71.4)	104(85.2)	0.028
腹腔镜+开腹手术	2(4.8)	8(6.6)	
术中Losanoff和Basson分型[n(%)]			
1型	24(57.1)	20(17.7)	
2型	15(35.7)	62(54.9)	<0.05
3型	1(2.4)	18(15.9)	
4型	2(4.8)	13(11.5)	
术后阑尾病理结果[n(%)]			
正常	24(57.1)	20(16.7)	
急性阑尾炎	17(40.5)	86(71.7)	<0.05
合并其他病理结果	1(2.4)	14(11.6)	
术后并发症[n(%)]	0(0.0)	17(20.5)	0.006
术后平均住院日[d,M(IQR)]	2(1~4)	5(2~7)	0.004

注：1）1例患者是通过灌肠术前确诊Amyand疝

Note: 1) One case of Amyand's hernia diagnosed by enema before operation

3　讨　论

Amyand 疝最早报道见于 1735 年，根据疝入阑尾的状态不同可分为 4 型：1 型为正常阑尾；2 型为局限于疝囊内的急性阑尾炎；3 型为急性阑尾炎合并腹膜炎；4 型为急性阑尾炎合并其他腹部病变[4]。本研究发现，对其集中报道主要出现在 2011—2020 年，以病例报道和病例系列为主，仅有 3 篇单中心的回顾性研究[4,135,142]，临床证据较为有限。

本文中 Amyand 疝的男女比例为 12∶1，中位发病年龄为 61 岁且女性好发于绝经后，和其他研究一致[4]。据报道，Amyand 疝呈现以婴儿和老年人为主的双峰分布趋势，这主要是因为刚出生时内环最大，以后逐渐缩小，中年以后又随着年龄再次扩大[146]。Amyand 疝好发于右侧，这和阑尾更靠近右侧腹股沟管相关，本文中有 10.9% 的患者发生于左侧，这或许跟盲肠的高度活动或阑尾过长、肠旋转不良和内脏转位有关。

Amyand 疝多表现为上腹或脐周疼痛，局限于右下象限，并伴有腹股沟区可复性或不可复性肿物。其病情的严重程度与阑尾疝入的时间及状态密切相关，严重者可合并急性附睾炎、睾丸炎，阴囊或阴道脓肿，坏死性筋膜炎，甚至肠穿孔而需要急诊手术干预[8,28,62]。术中需高度警惕合并滑疝，Richter 疝和 Littre 疝的可能。

腹部 X 线几乎没有诊断意义，相较于超声，CT 的诊断效能更高，可直接显示位于腹股沟管内的阑尾，而盲肠接近疝囊也可作为 Amyand 疝的一个间接征象[36]。但 CT 并不是常规检查手段，只有在需要排除更为严重的腹腔内病变或并发症时才应用。因此，本文中只有 25.7% 的患者获得了术前诊断。实际上，Amyand 疝与嵌顿性或绞窄性腹股沟疝难以鉴别，常需急诊手术治疗，而进一步的影像学评估很少改变治疗策略，故这一数字之低是意料之中的。

对于 Amyand 疝的治疗至今仍未形成共识，主要的争议在两点：是否应用补片和是否切除阑尾。本文中择期患者比急诊患者具有更高的补片使用率，可能是由于正常阑尾的占比较高。进一步研究发现，对伴有急性阑尾炎的 Amyand 疝患者，更多采用传统疝修补术或推迟修补而替代补片的植入，这主要遵循了避免将补片放置在潜在清洁污染切口的临床共识。但越来越多的学者[81, 113, 147–150]提出，嵌顿性或绞窄性疝（即使需要手术切除肠管时）的急诊处理中使用补片是安全、可行、有效的，具有可接受的手术部位及补片感染率，且能显著降低术后复发。目前国内外已有多例应用补片成功治疗嵌顿性 Amyand 疝或伴有严重阑尾炎症的 Amyand 疝的报道，均预后良好，未出现严重并发症[36,113,133]。对于伴有严重阑尾坏疽穿孔伴明显脓肿形成的嵌顿性 Amyand 疝，在组织严重水肿的条件下，若采用经典的 Shouldice 或 Bassini 术缝合将极为困难，复发率高且缝线也易诱发感染。Torino 等[113]通过术中行术区抗生素灌洗+腱膜下引流+术后甲硝唑联用哌拉西林他唑巴坦抗感染等措施，用聚丙烯补片成功修补，预后良好。笔者认为，随着新型生物补片的出现、做好局部冲洗和通畅引流、术后抗生素的连续应用等一系列举措，不必将阑尾炎看做是使用补片修补的绝对禁忌，但伴有明显坏疽或脓肿形成的阑尾炎，在应用补片时仍需慎重。腹腔镜在 Amyand 疝中的应用并非十分广泛（择期 23.8%，急诊 8.2%），但随着腹腔镜技术日渐成熟，其在 Amyand 疝的探查和治疗中具备一定的优势，尤其在诊断不明的病例中[12,151]。腹腔镜可直观地观察腹腔内情况，既可可视化游离阑尾减少局部感染风险，又避免了开腹手术疝囊颈部的扩张或疝缺损的扩大，降低了疝复发的可能性。而是否应该预防性切除正常的阑尾也是争论的焦点所在，大部分学者并不赞成此项操作，一方面可规避潜在的伤口感染风险，另一方面保留的阑尾将来也可留作它用[3]。相反，有文献[3, 152–153]指出切除是有必要的，一是对正常阑尾的牵拉可能会引起继发性阑尾炎，二是阑尾很可能会再次嵌顿引起病情反复，尤其在年轻群体中。Michalinos 等[139]介绍了 2 例因共病而不得不预防性切除的情况，有效的避免了潜在的并发症和病死率。而对于左侧 1 型 Amyand 疝，大多数学者赞成手术切除正常阑尾[153]。综上，笔者认为需结合患者的年龄、预期寿命、罹患阑尾炎的风险和术中探查情况具体研判。

本文中有 17 例患者术后出现不同程度的并发症，其中 5 例为传统疝修补术后[4, 16, 45, 109, 137]出现手术部位感染，经对症治疗后好转。这似乎和常规腹股沟疝修补术后的感染率相似[154]，但笔者认为在存在选择偏倚且缺少补片相关感染数据的情况

下，这一数据可能被低估。2例患者[4,48]因合并盲肠穿孔行Bassini修补术后疝复发。3例患者[68, 99, 138]出现手术部位血清肿。仅有1例患者[4]在术后第9天死于肺炎。

Amyand疝合并肿瘤的情况就更为罕见，很难收集大量的数据去做临床决策的依据，需要临床医生遵循肿瘤学的原则个体化处理。本文中的12例相关患者，均无特异性症状，无1例术前诊断[19, 22, 26, 37, 44, 52, 56, 63, 72, 74, 105, 117]。据文献[45]报道阑尾类癌好发于远端1/3，约占阑尾肿瘤的80%，平均发病年龄为40岁，本文5例男性阑尾类癌发病年龄较高（65岁），这可能和较有限的病例数有关。一般认为，当符合以下一项或多项时，应考虑行右半结肠切除术[105]：⑴ 肿瘤>2 cm；⑵ 淋巴结转移；⑶ 阑尾基底部受累及盲肠壁浸润；⑷ 有丝分裂活动增加；⑸ 未分化癌。而对于女性杯状细胞类癌患者，由于较高的腹膜种植率，推荐行双侧输卵管-卵巢切除术[155]。与既往研究[22]相一致，笔者认为病变超过黏膜的阑尾腺癌应行右半结肠切除术，对于局限于黏膜内的原位腺癌，右半结肠不比单纯切除阑尾更具优势。

据笔者所知，本文是关于Amyand疝最详尽的系统评价。本研究将检索时间限制为近20年，以减小诊断与手术方法的异质性，但仍存在一些局限性。首先，由于纳入的原始研究以病例报道和病例系列为主，存在难以规避的选择偏倚和发表偏倚。其次，关于新型生物补片应用的数据较少，仍有很大的发展前景。第三，本文的中位随访时间为6个月，对于感染、慢性疼痛和复发等并发症缺乏长期客观的随访数据，因而也缺少对这些结果的定性分析。最后，由于Amyand疝的发病率低，很难有大规模的随机对照试验去作为治疗决策的依据。但笔者认为，开展多中心的临床研究以及前瞻性大数据库的建立，能为Amyand疝的诊疗提供帮助。到目前为止，Amyand疝的谜底尚未完全揭开，未来仍需要普外科、儿科、病理科和放射科医生的密切合作。

Amyand疝是一种特殊类型的腹股沟疝，临床医生应该对其有所认识。Amyand疝好发于男性，与嵌顿性或绞窄性疝难以鉴别，常需急诊手术。Losanoff和Basson分类有助于指导术中决策，笔者认为在手术野较为干净的清洁污染切口，补片修补是可行有效的，而当阑尾坏疽穿孔时，应用补片应慎重，以不引起植入的补片感染为基本原则行个体化治疗。不常规对正常阑尾行预防性切除看似是合理的，还应考虑到患者的年龄、身体状态和预期寿命。应鼓励更多Amyand疝的基础和临床研究，提高Amyand疝的诊疗水平和患者预后。

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