Case Report

Meckel's Diverticulum Causing Ileal Volvulus and Peritonitis after a Recent Appendectomy: Case Report and Review of Literature – Should We Resect an Incidental One?

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Posted Date: 18 September 2023

doi: 10.20944/preprints202309.1185.v1

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Review

Meckel's Diverticulum Causing Ileal Volvulus and Peritonitis after a Recent Appendectomy: Case Report and Review of Literature—Should We Resect an Incidental One?

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Abstract: Meckel's diverticulum (MD) is the most common congenital anomaly of the gastrointestinal tract with a 1-3% prevalence in general population. The surgical management of symptomatic MD is well described in the literature, but there is still no consensus on the indication for prophylactic resection of incidental asymptomatic MD. To address this issue, we extensively reviewed the current literature and report our experience with laparoscopic management of an unusual case of MD causing ileal volvulus and acute peritonitis two weeks after a laparoscopic appendectomy for acute gangrenous appendicitis performed in another hospital. A 50-year-old man presented to the emergency department with acute and severe abdominal pain, vomiting and constipation. He had undergone a laparoscopic appendectomy for acute appendicitis two weeks before in another hospital. The patient was apyretic, distressed, and sought an antalgic position. The abdomen was mildly distended, tender, and the Blumberg sign was mildly positive in the central quadrants. The clinical picture deteriorated with fever, peritonismus, and leukocytosis. A CT scan showed an ileo-ileal adhesion near the ileocolic junction and dilatation of the upstream loops with air-fluid levels. Through an urgent laparoscopy, a necrotic mass, the MD, was wedge-resected and the surrounding ileal volvulus derotated. The postoperative course was uneventful. There is no definitive consensus on the appropriate management of incidental asymptomatic MD, although several studies have attempted to identify guiding criteria. Features of the MD, patient's risk factors, clinical presentation, and surgical approach need to be considered to establish definitive guidelines for the management of incidental asymptomatic MD. In the absence of definitive guidelines, personal expertise and judgement are the main resources for the surgeon approaching an incidental asymptomatic MD.

Keywords: Meckel's diverticulum; laparoscopy; peritonitis; emergency surgery; volvulus; appendectomy

1. Introduction

Meckel's diverticulum (MD) is the most common congenital anomaly of the gastrointestinal (GI) tract, with a prevalence of 1-3% in the general population [1–5]. It is the result of incomplete obliteration of the vitelline (omphalomesenteric) duct, which connects the primitive intestine to the yolk sac in early fetal life [2,6]. By definition, MD is a true diverticulum as it is surrounded by all the layers of the small bowel wall [7]. The right vitelline artery, which arises from the superior mesenteric artery, supplies the MD [8]. When present, the MD is located along the antimesenteric margin of the ileum at a mean distance of 52.4 cm (range, 7 - 100) from the ileocecal valve (ICV) and has a mean

 doi:10.20944/preprints202309.1185.v1

length and diameter of 3 and 1.58 cm, respectively [1]. In about half of cases, MD contains heterotopic mucosa within its wall: most commonly gastric mucosa (60%), followed by pancreatic acinar tissue (16%), and the remainder Brunner's glands, pancreatic islets, colonic mucosa, hepatobiliary tissue, or a combination of these [3,9]. The presence of ectopic tissue within, especially the gastric type and at younger age, tend to increase the risk of clinical manifestation from MD, mostly in the form of bleeding [1,10,11]. According to the literature, MD becomes symptomatic in 4-71% of cases, predominantly in the first three to four decades of life, and up to four times more frequently in men [1,12,13]. Symptomatic adult patients usually present with intestinal obstruction (35.6%), inflammation (29.4%) or intestinal bleeding (27.3%) [1]. In paediatric symptomatic patients, these clinical manifestations occur in 46.7%, 19.5% and 25.3% of cases, respectively [1]. Elderly patients most commonly develop neoplastic degeneration within the MD tissue [1,14,15], often with a poor prognosis [16]. Complications of MD are hemorrhage, intestinal obstruction, volvulus, inflammation, and perforation [14]. Perforation or inflammation of the MD is occasionally misdiagnosed as acute appendicitis [1,17]. Mortality of MD is around 0.1% [18]. We report our experience of laparoscopic management of an unusual case of MD causing ileal volvulus and acute peritonitis only 15 days after laparoscopic appendectomy for acute appendicitis. The surgical management of MD has been widely described in the literature, but there is no definitive consensus regarding its prophylactic resection. To address this issue, we conducted a comprehensive review of the literature.

2. Case presentation

A 50-year-old man presented to the emergency department (ED) of our hospital (Ospedale di Circolo e Fondazione Macchi, Varese, Italy) with acute, cramping, diffuse and severe (Numeric Pain Rating Scale 8) abdominal pain, associated with vomiting and constipation. He had undergone a laparoscopic appendectomy (LAE) for acute appendicitis two weeks before in another hospital. His past medical history includes an ischaemic stroke and a patent foramen ovale, currently treated with low-dose aspirin. The patient was apyretic, had a distressed and anxious appearance and sought an antalgic position. On physical examination, scars from previous surgery were physiologically healed; peristalsis was present on auscultation; the abdomen was mildly distended, tender all over, and the Blumberg sign was mildly positive in the central quadrants. The patient reported the passage of gas, but there was no fecal material on digital rectal examination. Initial laboratory tests were within normal limits (leukocytes 6.3*109 units/L, PCR 1.0 mg/dL), except for a slight elevation in transaminases. A plain thoraco-abdominal x-ray showed a dilated stomach, coprostasis, no free air in the abdomen, and metallic clips from previous surgery. Computed tomography (CT) scan showed adhesion between two distal ileal loops near the ileocolic junction, thickening of the surrounding peritoneum, dilatation of the upstream intestinal loops with internal air-fluid levels, no free fluid in the abdomen [Figures 1–3]. Initial conservative management (analgesic, antispasmodic, antiemetic and gastroprotective drugs with parenteral hydration) failed as the clinical condition rapidly deteriorated. The patient developed a fever and his abdominal pain worsened with onset of diffuse peritonismus. Blood tests showed significant leukocytosis (12.53*109 units/L). An urgent abdominal ultrasound (US) showed free fluid in the lower abdominal quadrants. An urgent laparoscopy was performed. The trocars were placed in the same positions as the previous LAE. On exploration, there was abundant serosanguinous fluid in the abdominal cavity. Approximately 30 cm proximal to the ileocolic junction, there was an ileal volvulus around a necro-hemorrhagic mass originating from the ileal wall [Figure 4]. The surgeon derotated the volvulus and performed a wedge-resection of the necro-hemorrhagic mass [Figure 5]. On anatomopathological examination, the mass was identified as a MD measuring 3 x 2.5 x 2 cm, with ischemic and infarcted areas and no evidence of heterotopic tissue. No neoplastic cells were found in an intraperitoneal fluid sample. The postoperative course was uneventful, and the patient was discharged on postoperative day (POD) 7. Follow-up was unremarkable.

 doi:10.20944/preprints202309.1185.v1

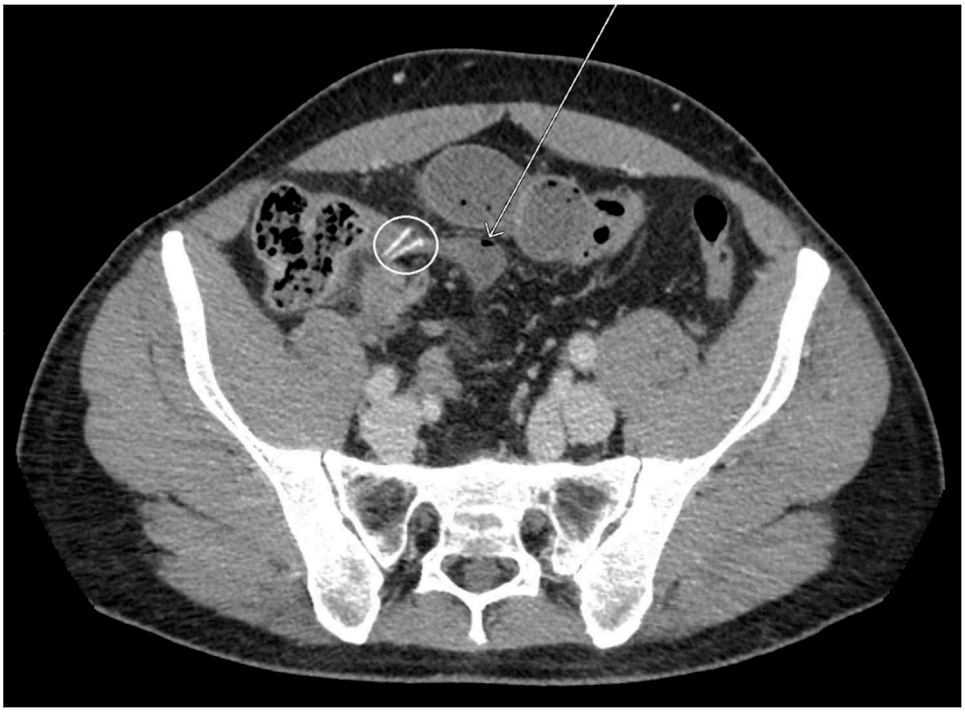

Figure 1. CT scan: MD (arrow) and suturing clips of previous appendectomy (circle).

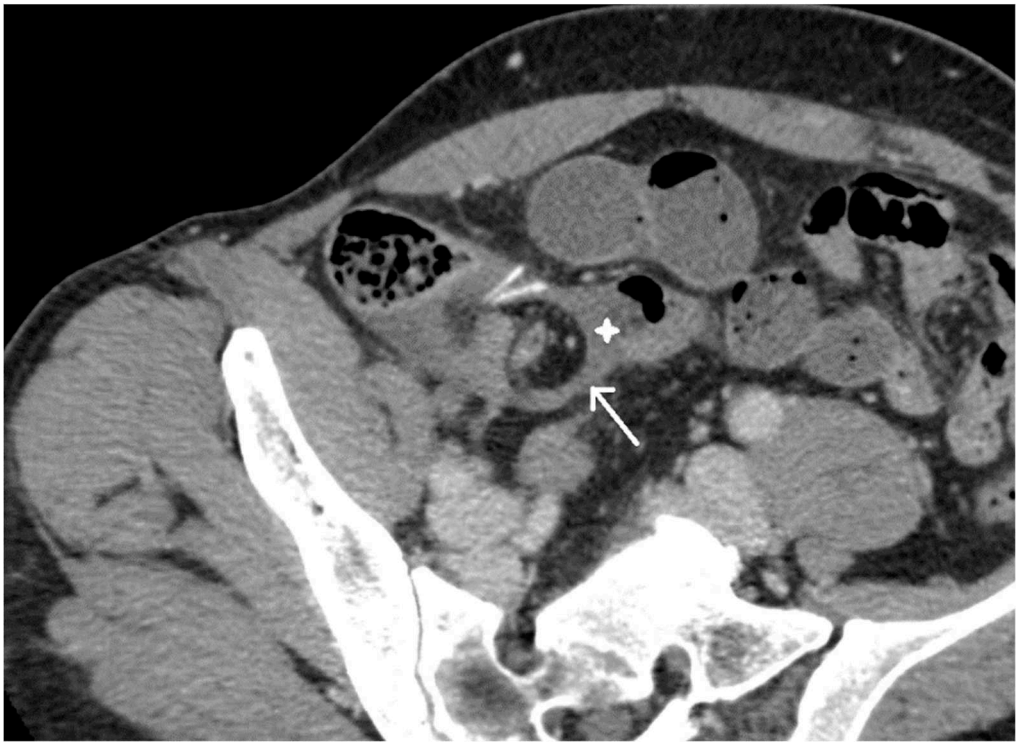

Figure 2. CT scan: Pedunculated base of the MD (arrow) and MD (star).

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Figure 3. CT scan: pre-op coronal view, congested mesentery (circle).

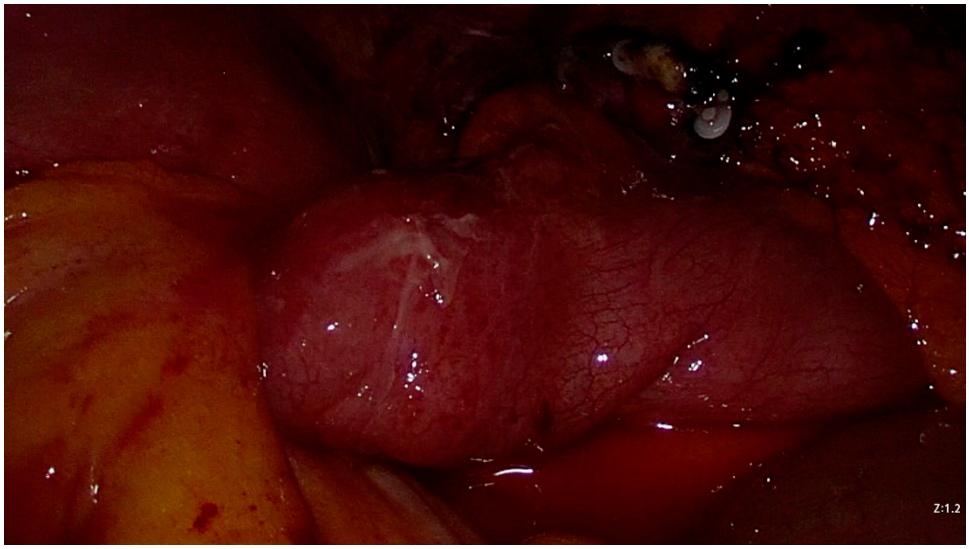

Figure 4. Intraoperative image: ileal volvulus and clips of previous laparoscopic appendectomy.

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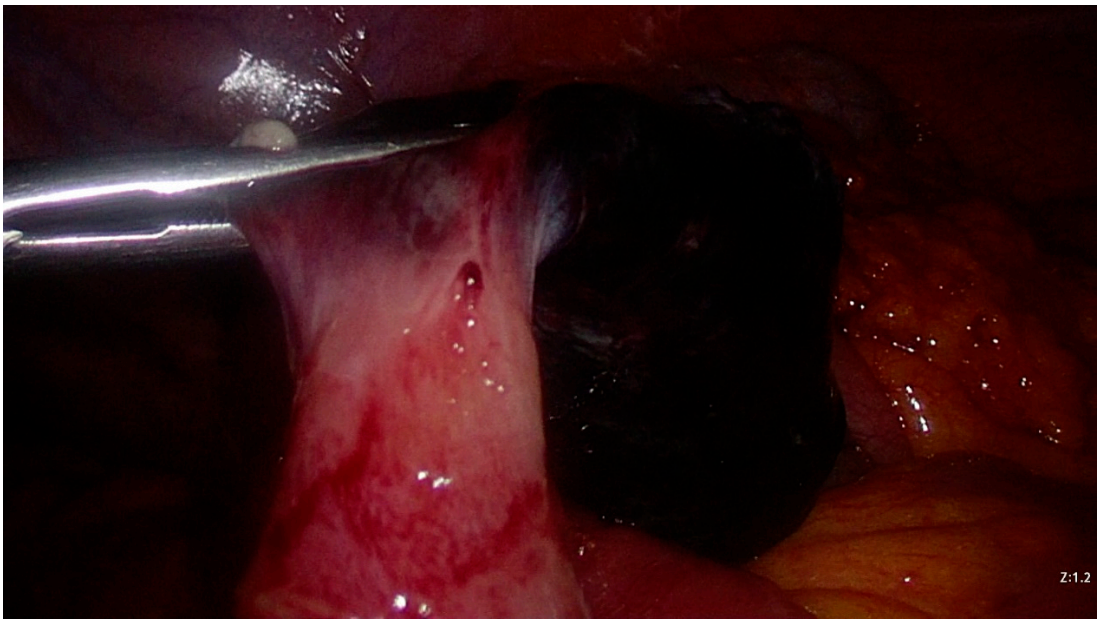

Figure 5. Intraoperative image: mechanical stapler on MD base (necrotic mass).

3. Materials and Methods

We extensively examined past publications using the Database PubMed – United States National Library of Medicine. We considered 58 publications, including case series, case reports, and reviews, ranging from 1976 to 2023.

4. Discussion

Meckel's diverticulum is a diagnostic challenge for clinicians because of its non-specific clinical manifestations. Meckel's diverticulum is frequently discovered incidentally or during work-up for clinical conditions initially attributed to another cause (e.g., during videolaparoscopy). According to the literature, surgical resection, either open or laparoscopic, is the treatment of choice for a symptomatic MD, when appropriate [1,19]. The extent of resection should be guided by the clinical presentation and intraoperative findings, ranging from a stapled diverticulectomy or a limited wedge resection with closure of the ileal defect [20], to a more extensive bowel resection with subsequent loop anastomosis [21]. Meckel's diverticulectomy has been demonstrated to be a safe method in the management of symptomatic MD in both adult and paediatric cohorts [19,22]. Morbidity was found to be higher after bowel resection than after simple diverticulectomy [23,24]. However, some advocate segmental bowel resection followed by anastomosis to ensure that no ectopic mucosa is left behind [25]. Taking a middle ground, Brungardt et al. in 2021 stated that diverticulectomy and small bowel resection are both likely to be appropriate approaches to the management of symptomatic MD when used in conjunction with surgeon discretion, as there is no difference in outcomes between the two techniques [26]. Laparoscopic diverticulectomy is technically safe, cost-effective, and efficient in both emergency and elective settings [20,22,27], avoiding the morbidity associated with both open surgery and small bowel resection, with the added benefits of precise operative diagnosis and shorter recovery time [24,28].

However, despite decades of research, there is no consensus on the management and indication for possible prophylactic resection of incidental asymptomatic MD [29]. Most of the evidence on MD comes from retrospective studies. There are only a few randomized studies, mostly single center, on complications, and none on incidental MD.

Early studies about asymptomatic MD were not in favor of prophylactic surgical resection. In 1962, Weinstein et al. reviewed 560 incidental MDs, of which 158 were left in situ on the basis that wide-mouthed diverticula (average width and length 1.92 cm x 2.99 cm, respectively) were not

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considered dangerous, and therefore their resection would only add to the risk of surgical complications and not provide any benefit [30]. In 1976, Soltero et al. estimated the lifetime risk of complications from MD to be 4.2%, eventually falling to 0% in old age; from then published data, they calculated that it would be necessary to resect approximately 800 asymptomatic MDs to save one patient's life [31]. In 1986, considering that the calculated lifetime risk of complications from MD is very low and the risk of complications after diverticulectomy is not negligible, Leijonmarck et al. concluded that an incidental asymptomatic MD in adults should be left in situ [12]. In addition, they reported 28 cases of MD left in situ with no further clinical manifestations after an average of almost 8 years of follow up [12]. In 1993, DiGiacomo et al. stated that any incidental MD whose resection with a stapler would be unsafe and difficult because it is wide and short, should be left in situ because of the low risk of complications [32]. In 1995, Peoples et al. stated that incidental diverticulectomy in adults should be abandoned, because the lifetime risk of developing complications does not significantly outweigh the surgical morbidity and mortality of resection [33].

In 1994, Cullen et al. concluded that incidental MDs should be removed to avoid future potential complications unless an additional condition (e.g., generalized peritonitis) would make resection hazardous [34]. In 1995, Matsagas et al. studied a population of 2074 patients undergoing AE with intraoperative evaluation for MD; they concluded that resection of the unexpected MD can be performed safely with a low complication rate, regardless of the patient's age [35].

In 2004, Bani-Hani et al. stated that resection of incidentally found MDs does not increase operative morbidity and mortality and was therefore worthy, especially in young patients with long and narrow (diameter ≤ 2 cm) MD [36]. Since then, many authors have advocated prophylactic resection of asymptomatic MD [11,37–46], also, but not exclusively, to prevent potential future neoplastic degeneration [39–42,44,45]. A recent multi-center retrospective study by Tree et al. supports laparoscopic stapled resection of incidental asymptomatic MD, considering the overall low complication rate and the potential for malignant transformation [24].

On the other hand, some other authors in the 2000s still prefer to leave an asymptomatic MD in situ. According to a 2004 observational study of 47 patients by Stone et al., removal of any asymptomatic MD should not be supported because complications of such a procedure, although uncommon, are often life-threatening [47]. In a 2008 review of the literature, Zani et al. did not support prophylactic resection of an incidental asymptomatic MD, noting that MD resection carried a 5.3% morbidity rate, mostly wound infections, while leaving MD in situ had an overall lower morbidity rate and long-term risk of complications [18].

A risk-based approach was first proposed in 1983 by Mackey et al.: they reviewed over 400 symptomatic MDs and identified some risk factors for complications such as age ≤40 years, length of the diverticulum ≥2 cm, presence of heterotopic tissue, and male sex [48].

In 2005, Park et al., after reviewing 1476 patients found to have an MD during surgery from 1950 to 2002, could neither definitively support nor reject the recommendation that every incidental asymptomatic MD should be removed, but recommended removal of any incidental MD that had any of the four features (patient age younger than 50 years, male sex, diverticulum length greater than 2 cm, and the presence of histologically abnormal tissue) most commonly associated with clinical manifestations [14]. In the same year, after reviewing 233 cases of MD incidentally discovered during an appendectomy, Ueberrueck et al. stated that in cases of gangrenous or perforated appendicitis, an incidentally discovered MD should be left in place, whereas in an only mildly inflamed appendix it should be removed [49]. In 2006, Robijn et al. proposed a scoring system based on four risk factors (male sex, age <45 years, diverticula >2 cm, and the presence of a fibrous band), to guide the decision for surgery in incidental asymptomatic MDs [50]. This risk-based approach has been supported by a growing number of authors [25,51–58]. According to Mackey [48], Park [14], Robijn [50] and other more recent authors, some characteristics of the patient and the MD were clear indications to perform a prophylactic diverticulectomy. According to DiGiacomo, only the intra-operatively tested fit of the MD within the mechanical stapler could lead to a decision [32]. Considering the work of Hansen, although the size of the MD was an indication for prophylactic resection due to the risk of future complications, the severe intercurrent gangrenous appendicitis validly discouraged such an

adjunctive surgical act [1]. According to Leijonmack, the size of our reported MD was not an indication for prophylactic diverticulectomy [12]. According to the conclusions of both Cullen [34] and Ueberrueck [49], gangrenous appendicitis with peritonitis was a contraindication for incidental removal of the MD, as the surgical act would have been riskier than leaving it in situ. Looking a posteriori at our reported case, not removing the MD during the previous LAE seems to be a mistake, but an a priori decision regarding its resection at such a time would have been controversial and could only have been based on the judgement, expertise, and competence of the treating surgeon.

5. Conclusions

In conclusion, MD occurs in a minority of the population and its clinical manifestations are largely non-specific. We managed life-threatening small bowel volvulus and peritonitis in a patient who had undergone LAE two weeks earlier, with the MD left in situ. There is no definitive consensus on the appropriate management of incidental asymptomatic MD. Over the decades, several studies have attempted to identify criteria that would allow a decision to be made whether to resect or leave in situ an incidental asymptomatic MD. After a thorough review of the literature, we could not definitively state whether the MD of our reported patient should have been resected or left in situ at the time of the previous LAE. According to our analysis, MD characteristics, patient's risk factors and the clinical presentation, and the surgical approach (open VS laparoscopic) need to be considered to establish definitive guidelines for the management of incidental asymptomatic MD. Considering the surgical approach, complications that might discourage open surgery and resection are less common with laparoscopic diverticulectomy. In the absence of definitive guidelines, expertise and common sense remain the main resources for the surgeon approaching an incidental asymptomatic MD.

Author Contributions: M.Z.: planned and wrote the original draft; D.I.: involved in the planning and revision of the manuscript; V.P.: involved in the planning and revision of the manuscript; N.G.: revised the manuscript; M.M.: revised the manuscript; G.S.: revised the manuscript; G.C.: involved in the planning and revision of the manuscript, supervised; G.I.: involved in the planning and revision of the manuscript, supervised. All authors have read and agreed to the published version of the manuscript.

Funding: This research received no external funding.

Informed Consent Statement: The patient verbally agreed and gave consent to publish this paper.

Data Availability Statement: Patients' data registry of Ospedale di Circolo e Fondazione Macchi (VA).

Conflicts of Interest: The authors declare no conflict of interest.

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