Cutaneous presentations of omphalomesenteric duct remnant: A systematic review of the literature

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Background: Disorders of the umbilicus are commonly seen in infancy, including hernias, infections, anomalies, granulomas, and malignancies. Meticulous inspection of the umbilicus at birth might reveal a persisting embryonic remnant, such as an omphalomesenteric duct (OMD), manifested by a variety of cutaneous signs, such as an umbilical mass, granulation tissue, or discharge.

Objective: To systematically review the available data regarding the presence and management of OMD remnant with cutaneous involvement to suggest a practical approach for diagnosis and treatment.

Methods: A systematic review of the literature evaluating OMD anomalies presenting with cutaneous symptoms was performed. In addition, an index case of an 11-month-old patient is presented.

Results: We included 59 publications reporting 536 cases; 97% of the patients whose age was noted were infants (mean age 11 months). In 7.5% of the cases, diagnosis was established only after treatment failure. In 6.4% of patients, nonlethal complications were reported, and in 10.3%, the outcome was death, partly due to delayed diagnosis or mismanagement.

Limitations: Limited quality of the collected data, reporting bias.

Conclusion: OMD is relatively rare; however, the clinician must consider this remnant while examining patients with umbilical abnormalities because mismanagement could cause severe morbidity and mortality. (J Am Acad Dermatol 2019;81:1120-6.)

Key words: Meckel's diverticulum; omphalomesenteric duct; pyogenic granuloma; umbilical anomalies; umbilical polyp; umbilicoileal fistula; vitelline duct.

Disorders of the umbilicus are most commonly seen in infancy and include hernias, infections, congenital anomalies, granulomas, and rare malignancies. The umbilical cord develops after the fourth week of gestation, forming the umbilical vessels, urachus, and omphalomesenteric duct (OMD). The OMD connects the yolk sac to the fetal gut. Toward the end of the first trimester, both the OMD and the urachus regress, leaving no fetal remnants. However, in some cases, complete or partial failure of involution leads to a variety of birth anomalies.[1,2] A completely patent

Abbreviations used:

MeSH: medical subject headings
OMD: omphalomesenteric duct

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OMD results in a direct connection from the umbilicus to the gut in the terminal ileum region. A remnant at the umbilical end that has no connection to the gut results in aberrant tissue, also referred to as an umbilical polyp or closed OMD. A remnant of the duct located in the ileum region that has no connection to the umbilicus is known as Meckel diverticulum. An OMD cyst is created when both ends (the umbilical and ileal ends) of the duct are closed but the mid-duct remains patent. A fibrous cord develops when fibrous tissue connects the umbilicus and the ileum, potentially leading to bowel obstruction.[1,2]

After birth, the most common clinical manifestations of an OMD remnant include an umbilical mass, a protruding nodule resembling a pyogenic granuloma, granulation tissue, and various types of umbilical discharge, eg, fecal, bloody, or nonspecific secretions. Patients occasionally present with signs and symptoms that prompt urgent care, such as abdominal pain, vomiting, bloody stools, fever, tachycardia, or respiratory distress due to sepsis.[1-6]

CAPSULE SUMMARY

- Omphalomesenteric duct remnant is rare; however, its mismanagement could cause severe morbidity and mortality. A systematic review of the literature regarding such anomalies presenting with cutaneous symptoms has yet to be performed.
- Our findings emphasize red flags and suggest an approach for clinical diagnosis and treatment.

The diagnosis of OMD anomalies is aided by imaging studies, such as ultrasonography, a Meckel scan, or computed tomography. Tissue biopsies might be obtained to rule out other entities, such as pyogenic granulomas, hemangiomas, or malignancies.

Treatment often requires surgical intervention for resection of the redundant tissue to limit potential complications, such as strangulated hernias, bowel perforation, hemorrhage, or tissue infections.[1,2] An erroneous diagnosis made on the basis of cutaneous findings could lead to mismanagement, potentially resulting in morbidity or even mortality due to complications, such as bowel obstruction, perforation, or sepsis.[1,2,6-14]

An 11-month-old healthy boy with a pinkish-red umbilical nodule was brought to our clinic (Fig 1). An initial clinical diagnosis of pyogenic granuloma led to a bed-side surgical resection of the nodule. Surprisingly, the pathology report described bowel tissue with squamous epithelium and smooth muscle fibers at the depth of the specimen. These findings were compatible with an OMD remnant. Additional treatment and imaging studies were not required because the surgical wound healed well and the patient did not develop any complications. This patient prompted us to initiate a systematic review of the literature to better understand and prevent misdiagnosis of OMD remnants. Our objective was to systematically review the available data regarding the clinical presentations and management of OMD remnants with cutaneous involvement to develop a practical approach for diagnosis and treatment.

METHODS

A systematic review was conducted and reported in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement and was registered with the PROSPERO international prospective register of systematic reviews (listing no. CRD42018104571).

Search strategy

In March of 2018, we searched PubMed and Google Scholar for studies written in English without date limits. The following search criteria were used "omphalomesenteric" [MeSH terms] OR "omphalomesenteric" [all fields] OR "vitelline" [MeSH terms] OR "vitelline"[all fields] AND "remnant" [MeSH terms] OR "remnant" [all fields] OR "duct" [MeSH terms] OR "duct" [all fields] OR "Meckel's" [MeSH terms] OR "Meckel's" [all fields] AND "diverticulum" [MeSH terms] OR "diverticulum [all fields] OR "umbilicoileal" [MeSH terms] OR "umbilicoileal" [all fields] AND "fistula" [MeSH terms] OR "fistula" [all fields] OR "umbilical" [MeSH terms] OR "umbilical" [all fields] AND "polyp" [MeSH terms] OR "polyp" [all fields].

Eligibility criteria

We included relevant original studies of any design that described cases of OMD remnant presenting with cutaneous symptoms. Patients of all ages and of either sex were included if OMD remnant was diagnosed histologically or clinically and postsurgically.

Data extraction and quality

Two reviewers (Drs Solomon-Cohen and Zidan) independently screened titles and abstracts and then the full texts of studies considered potentially eligible. The first author (Dr Solomon-Cohen) extracted data onto an electronic form, and the second author (Dr Zidan) checked the pertinent data, including first author name, year of publication,

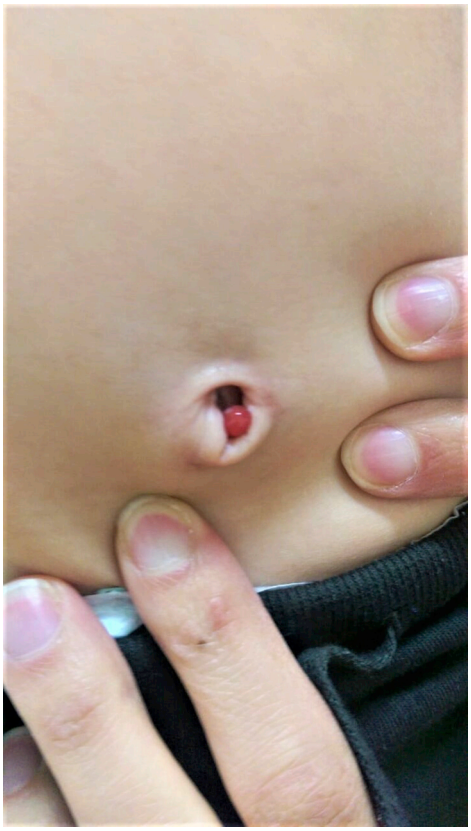

Fig 1. An 11-month-old healthy boy with a pinkish-red nodule protruding through his umbilicus that was later diagnosed as omphalomesenteric duct remnant.

number of participants, age, sex, clinical manifestation, time to diagnosis, treatment, and outcome. Discrepancies were resolved by a third reviewer (Dr Levi). Studies were graded according to their level of evidence: A for prospective, B for retrospective, and C for case reports and series.

Data were summarized with descriptive statistics. Categorical variables are presented as proportion or percentage and continuous variables as mean or median (range).

RESULTS

The study flow chart is described in Fig 2. Our search yielded 4139 publications. In addition to the present report, 59 publications fulfilled our inclusion criteria,[15-61] for a total of 60 publications dating back to 1942. These publications included 12 retrospective cohorts (grade B), 3 case series (grade C), and 45 case reports (grade C). The results are summarized in Table I.

A total of 536 patients with OMD remnants were included. Study sizes ranged 1-105 patients. Age was reported for 348 cases. Most cases (97.5%) were reported in the pediatric population; patient age was a mean of 11 months and a median of 5 weeks (range 1 day-8 years). Only 9 cases (2.5%) were reported in adults, with a median age of 34 (range 20-61) years. Sex was reported for 374 cases; of these, 283 (75.6%) were male and 91 (24.4%) were female.

Overall, 51 publications (111 patients) addressed the clinical presentations of an OMD remnant. The most common presentations were umbilical mass (60 cases, 54.1%), nonspecific or white umbilical secretion (33 cases, 29.7%), fecal umbilical secretion (19 cases, 17.1%), bloody umbilical secretion (10 cases, 9%), or umbilical granulation tissue (6 cases, 5.4%). Less common clinical presentations (which presented along with other cutaneous signs) included sepsis, rectal bleeding, respiratory distress, or umbilical prolapse of ileal loops (3 cases [2.7%] each); vomiting or a thick umbilical cord (2 cases [1.8%] each); and dehydration or an umbilical hematoma (1 case [0.9%] each). A few reports described various other clinical presentations (eg, abdominal pain, acute abdomen, and bowel obstruction), but exact numbers were not provided. The list of diagnoses is presented in Table II.

Treatment

The most common treatment modality was surgical excision of the lesion, which was performed in 99% of the cases in which outcome was reported. Two patients were inoperable[6] and eventually died of sepsis. In 1 patient, the remnant was reported to undergo spontaneous regression.[15] In 29 (7.5%) cases, diagnosis was established only after treatment failure with silver nitrate cauterization (25 patients), ligation (2 patients), laser treatment (1 patient), or oral medications (nonsteroidal anti-inflammatory drugs and antibiotics, 1 patient). Persisting symptoms required additional investigation, resulting in surgical resection and diagnosis. Two patients were diagnosed with malignancies arising in an OMD remnant (umbilical adenocarcinoma) and were treated with chemotherapy. Umbilical cord hemangioma arising in an OMD remnant was diagnosed in 2 patients, and 1 of these patients was treated with oral propranolol.

Outcomes and mortality

Outcome was reported in 515 cases; 33 (6.4%) patients experienced nonlethal complications either before diagnosis (12 cases, 36%) or after treatment (21 cases, 64%). These complications included evisceration of the small bowel or prolapse, intussusception, bowel obstruction, volvulus, infections, peritonitis or bowel perforation, peptic ulcer perforation, and hemorrhage. Fifty-three patients (10.3%) died either due to complications related to the basic

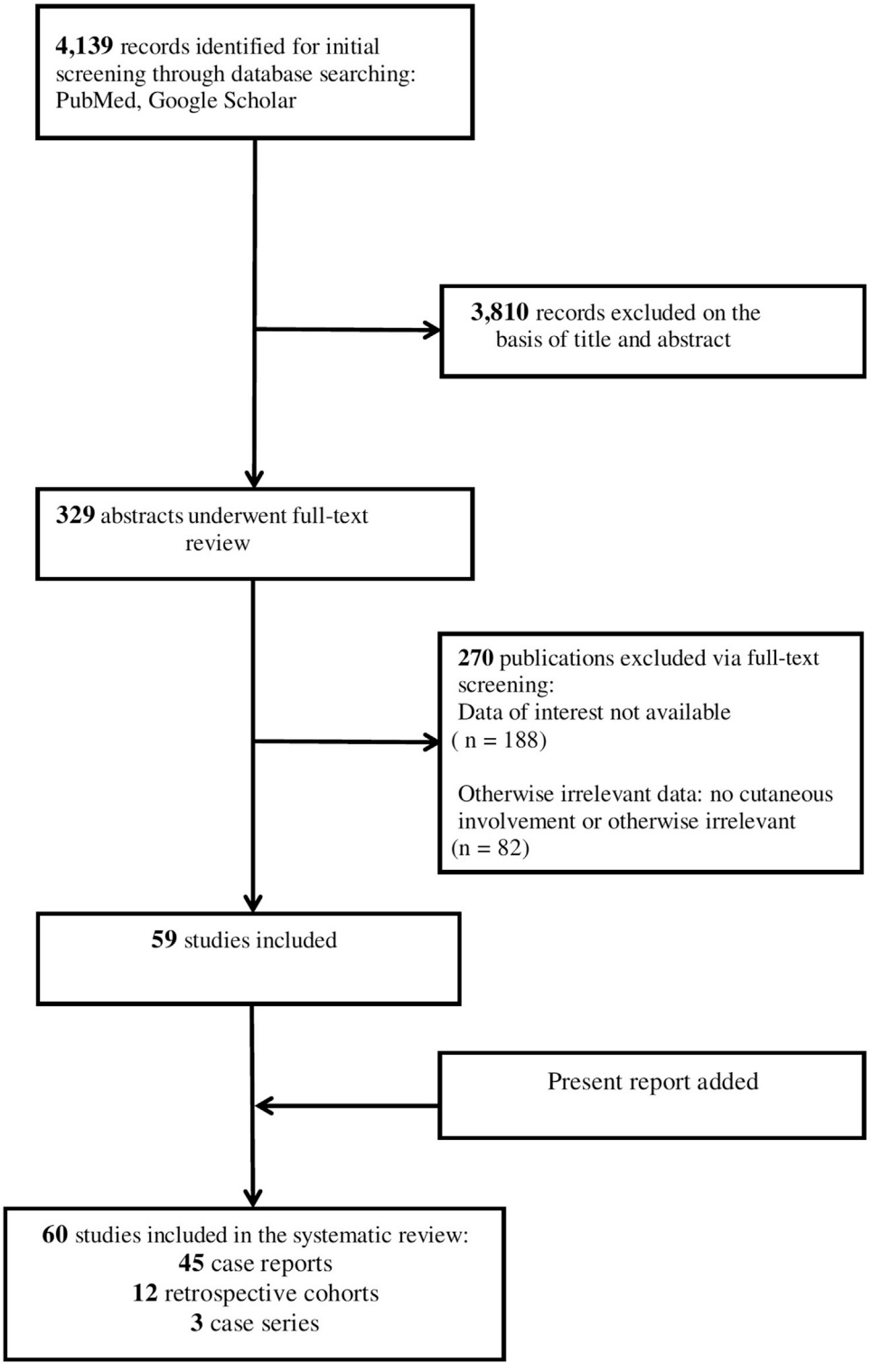

Fig 2. Systematic literature search according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines.

condition or the treatment. Causes of mortality included sepsis, peritonitis, bowel obstruction, ileal prolapse, intussusception, peptic ulcer hemorrhage, as well as other postsurgical complications (infections and anesthetic complications). It is noteworthy that 40% of these reported deaths (22 cases) occurred

Table I. Summary of the results of the systematic review

Parameter	Value
Total patients, N	536
Children, n (%)*	339 (97.5)
Median age (range)	5 weeks (1 day-8 years)
Adults, n (%)*	9 (2.5)
Age, y, median (range)	34 (20-61)
Male, n (%)*	283 (75.6)
Clinical presentation, n (%)*†	
Umbilical mass	60 (54.1)
Nonspecific umbilical secretion	23 (29.7)
Fecal umbilical secretion	19 (17.1)
Bloody umbilical secretion	10 (9)
Umbilical granulation tissue	6 (5.4)
Sepsis	3 (2.7)
Rectal bleeding	3 (2.7)
Respiratory distress	3 (2.7)
Umbilical prolapsed of ileal loops	3 (2.7)
Vomiting	2 (1.8)
Thick umbilical cord	2 (1.8)
Dehydration	1 (0.9)
Umbilical hematoma	1 (0.9)
Previous treatment failure, n (%)*	29 (7.5)
Silver nitrate cauterization	25 (86.2)
Ligation	2 (6.9)
Laser	1 (3.45)
Oral medications	1 (3.45)
Outcome, n (%)*	
Nonlethal complications before diagnosis	12 (2.3)
Nonlethal complications after treatment	21 (4.1)
Death	53 (10.3)
Spontaneous resolution	1 (0.2)
Treatment resulting in recovery	428 (83.1)

*Of cases providing data.
†Some patients had >1 clinical presentation.

Table II. List of diagnoses found in the systematic review

Diagnosis	Value, N = 343, n (%)
Closed OMD remnant (umbilical polyp)	125 (36.4)
Ileal-umbilical fistula (patent OMD)	80 (23.3)
Patent Meckel's diverticulum	64 (18.7)
Umbilical cyst	22 (6.4)
OMD sinus	20 (5.8)
Mesodiverticular band	16 (4.7)
Urachal remnant	7 (2.0)
Umbilical hernia	6 (1.7)
Adenocarcinoma of umbilicus	2 (0.6)
Umbilical cord hemangioma	2 (0.6)
Epidermoid cyst	1 (0.3)
Fibroepithelial polyp	1 (0.3)
Umbilical skin sinus	1 (0.3)

Some patients had >1 diagnosis.
OMD, Omphalomesenteric duct.

before 1942,[14] whereas 4 deaths were reported in the 21st century in developing countries.[6,9]

DISCUSSION

Although an OMD remnant is a relatively rare condition, mismanagement could cause severe morbidity and mortality, as demonstrated by our systematic review. The patient is especially prone to complications when the OMD remnant is associated with visceral malformations such as ileal-umbilical fistula or a Meckel diverticulum. OMD remnants are often clinically mistaken for other more benign lesions (eg, pyogenic granuloma). The clinician must consider OMD remnants in the differential diagnosis of umbilical abnormalities and recognize when further investigation (eg, imaging or biopsy) is needed before performing bedside treatments, such as silver nitrate cauterization or ligation.

Although the overall historical mortality rate is 10.3% in reported cases of OMD remnant, the rate is substantially lower today because of advances in medical imaging and treatment. However, serious morbidity remains a major concern, as reports of intestinal obstruction, bowel necrosis, intussusception, evisceration, and significant hemorrhage were published in recent years.[8-10]

There are no current guidelines for the diagnostic approach to umbilical disorders presenting with cutaneous findings. We hereby suggest a practical approach for diagnosis and treatment (Fig 3). A thorough history and physical examination are of utmost importance, with special emphasis on identification of red flags, such as a history of discharge, gas or urine secretion through the umbilicus, signs of peritonitis, and fever. Any of these red flags should lead to further investigation or surgical consultations.

As demonstrated by our systematic review, OMD remnant presenting in an adult is rare, with merely 2.5% of cases reported in patients >18 years of age. In a few cases, the previous cutaneous symptoms were initially misdiagnosed and led to delayed treatment, and 2 cases presented with malignancies. Most of these reports lack information regarding the exact cause for the delayed diagnosis.[16-23]

Although most umbilical conditions in infants (eg, umbilical granulomas and hernias) resolve spontaneously or with simple bedside treatments, physicians need to be aware of more hazardous disorders with similar presentations. Multiple reports of misdiagnosis, including our index case, and inappropriate management highlight the potential complications of

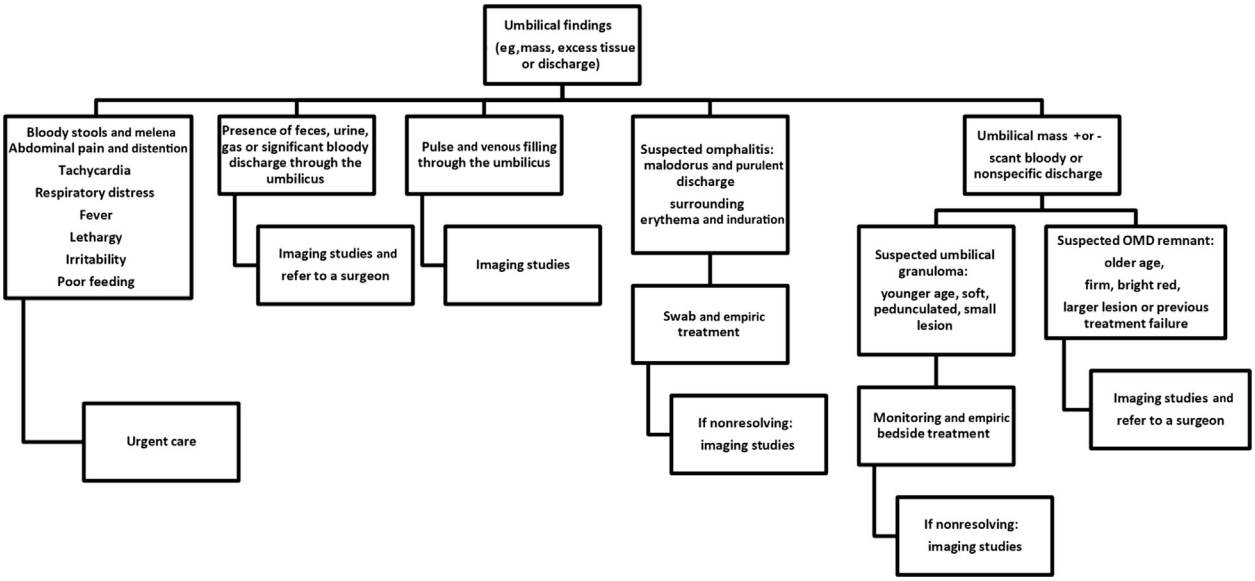

Fig 3. Suggested approach to umbilical lesions. *OMD*, Omphalomesenteric duct.

mishandled OMD remnants.[6,11,24,25] Our study emphasizes the importance of considering OMD remnant as part of the differential diagnosis in the proper clinical settings.

Study limitations

The quality of data in reports on OMD remnants with cutaneous manifestations is low, consisting of retrospective cohorts, case series, and case reports only. There is significant reporting bias, with overrepresentation of unusual or complicated cases that do not necessarily reflect the most common occurrences. In addition, some publications had missing data on the number of cases with each diagnosis and their outcomes or complications.

CONCLUSION

This systematic review sheds light on an uncommon but potentially dangerous cause of umbilical lesions, a relatively frequent clinical scenario among pediatric patients. Clinician awareness of suspicious features should prevent misdiagnosis and prompt appropriate evaluation of umbilical lesions, thereby ensuring early and proper treatment of OMD remnants.

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