

Ultrasonographic diagnosis and minimally invasive treatment of a patent urachus associated with a patent omphalomesenteric duct in a newborn

A case report

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Abstract

Rational: Patent urachus (PU) is due to an incomplete obliteration of the urachus, whereas patent omphalomesenteric duct (POMD) is due to an incomplete obliteration of the vitelline duct. These anomalies are very rarely associated with one another. We describe a case of a newborn with a PU associated with a POMD, who was diagnosed by an abdominal ultrasound (US) and laparoscopy, and managed with a minimally invasive excision.

Patient concern: A 28-day-old male neonate was referred to our hospital to investigate a delay in umbilical healing, with blood-mucinous material spillage for 3 weeks prior to the referral. The baby had no symptoms and was in good general health.

Diagnosis: After a thorough cleaning of the umbilical stump, a clear granuloma with a suspected fistula was evident under the seat of the ligation of the stump. An abdominal US examination revealed the formation of a full communication, starting below the umbilical stump and developing along the anterior abdominal wall that connected with the bladder dome. The US also revealed a tubular formation containing air, which was compatible with POMD, in the deepest portion of the same umbilical stump. Considering these findings, the rare diagnosis of a PU associated with a POMD duct was suspected.

Interventions: The child was then hospitalized for an elective laparoscopy that confirmed the US picture, and a minimally invasive excision was performed.

Outcome: The postoperative course was favorable and uneventful.

Lessons: Our case underlines the importance of evaluating all persisting umbilical lesions without delay when conventional pharmacological therapies fail. Using a US as the first approach is valuable and should be supported by laparoscopy to confirm the diagnosis; a minimally invasive excision of the remnants appears to be an effective therapeutic approach.

Abbreviations: GA = gestational age, POMD = patent omphalomesenteric duct, PU = patent urachus, URs = urachal remnants, US = ultrasound.

Keywords: patent omphalomesenteric duct, patent urachus, ultrasound, urachal remnants

1. Introduction

Urachus is an embryological remnant of the allantois.^[1] Between the fourth and fifth month of gestation, the fetal bladder tends to gradually descend, and its apical portion creates the urachus. The urachus obliterates itself and becomes the median umbilical

ligament. Its persistence after intrauterine life can manifest as different pathologies called urachal remnants (URs).^[2] Patent urachus (PU) is due to an incomplete obliteration of the urachus.

The vitelline duct is the narrow embryological channel that connects the yolk sac to the intestine. During the fetal life, this structure tends to obliterate itself until it disappears.^[3] The failure to achieve a full obliteration can cause the development of a duct, a ligament or a diverticulum, known as Meckel's diverticulum. Patent omphalomesenteric duct (POMD) is due to the incomplete obliteration of the vitelline duct.^[4] PU and POMD anomalies are very rarely associated with one another. We describe a case of a newborn with a PU associated with a POMD, who was diagnosed by an abdominal ultrasound (US) and laparoscopy, and managed with a minimally invasive excision.

2. Case

2.1. Presenting concerns

A 28-day-old male neonate was referred to our hospital to investigate a delay in umbilical healing with blood-mucinous material spillage. The primary care pediatrician referred the baby to the pediatric surgeon after 3 weeks of failed treatment with silver nitrate.

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The patient's parents provided their written informed consent for the publication of this study.

The authors report no conflicts of interest.

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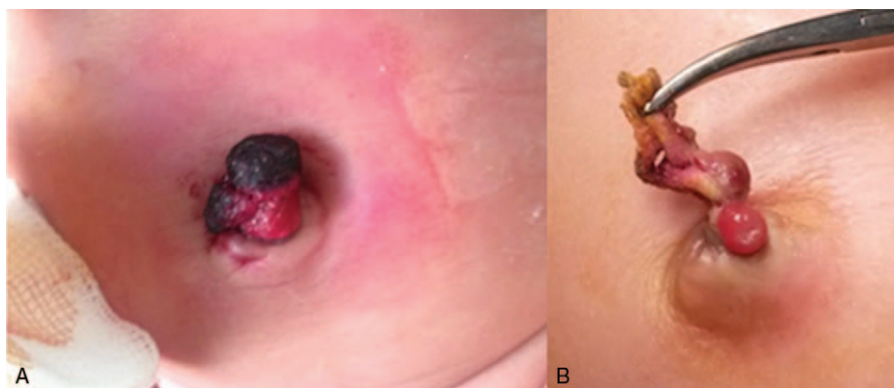


Figure 1. (A) The umbilicus of the newborn upon his arrival. (B) The granuloma with a suspected fistula after the cleaning of the umbilical stump.

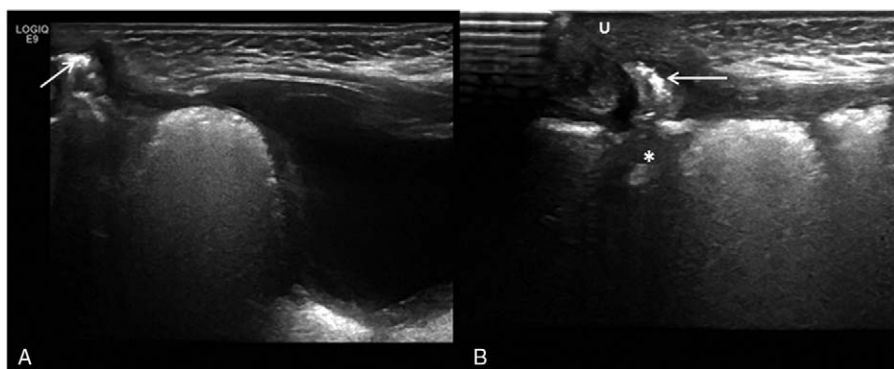


Figure 2. (A) Sagittal scanning: US revealed the formation of a tubular hypoechoic full communication, connecting the bladder dome to the umbilical stump, developing along the anterior abdominal wall, which is compatible with PU. Cranial to PU, in the deepest portion of the same umbilical stump, we could also observe a tubular formation containing air (hyperechoic) that was in apparent continuity with the intestinal loop, converging toward the umbilicus, which is compatible with POMD. The two findings are not on the same plane and, therefore, are not viewable by the same scan. (B) Paraxial right scanning: details of the umbilical stump, which is thickened and hyperemic, inside of which is an appreciable tubular formation, with hyperechoic content and laminated walls, in continuity with the intestinal loop placed more deeply, and also with air content (hyperechoic), which is compatible with POMD. To the right of this structure, the PU can be observed.

2.2. Clinical findings

In the clinical examination, the baby had no symptoms and was in good general health. A granuloma was evident below the mummified umbilical stump (Fig. 1A) with slight persistent bleeding.

2.3. Diagnostic focus and assessment

Hematological tests showed lymphocyte leukocytosis (white blood cells count, 12,230/ μ L, 67.4% lymphocytes). After a thorough cleaning of the umbilical stump, a clear granuloma with a suspected fistula was evident under the seat of the ligature of the stump (Fig. 1B). An abdominal US examination revealed the formation of a full communication, starting below the umbilical stump and developing along the anterior abdominal wall that connected with the bladder dome, and, first, a diagnosis of PU was made (Fig. 2A). The US revealed a tubular formation containing air, which is compatible with POMD, in the deepest portion of the same umbilical stump (Fig. 2B).

Considering these findings, the rare diagnosis of a PU associated with a POMD duct was suspected. The child was then hospitalized for elective laparoscopy that confirmed the US picture (Fig. 3).

2.4. Therapeutic focus and assessment

A minimally invasive excision was performed by removing the entire PU with a small cuff of the bladder dome through a 10 mm subumbilical incision. The removal of the POMD was performed

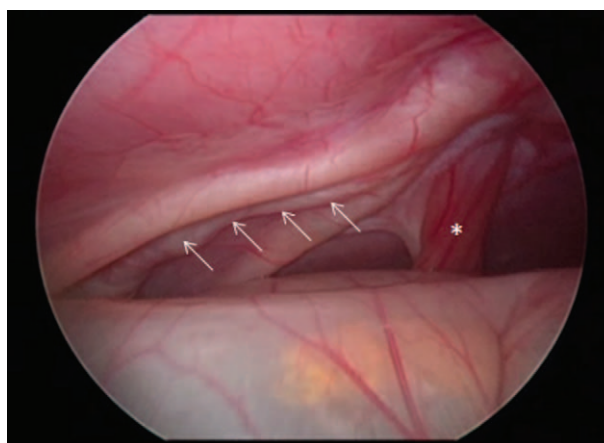


Figure 3. Laparoscopy showed a definitive diagnosis of a PU (arrows) associated with a POMD (asterisk).

Table 1

Cases with a PU associated with a POMD.

Pt	Authors	Age	Gender	GA	First attempt treatment	Signs at physical examination	Specific signs	Exams	Preoperative diagnosis	Surgical approach	Diagnosis of PU associated to POMD
1	Lexer et al, 1898 ^[10]	1.5 years	M	Not reported	Salves and plasters	Umbilical fistula in previous reddish umbilical tumor	Umbilical spillage of urine	None	PU	Elliptic incision around the navel	Histopathological
2	Cullen et al, 1916 ^[11]	14 months	M	Not reported	Not reported	Umbilical bleeding	Umbilical spillage of feces and urine	None	None	Elliptic incision around the navel	Incidental intraoperative
3	Davis et al, 1926 ^[12]	1 year	M	Not reported	Silver nitrate	Elevated umbilical pouch covered with red mucous membrane and bled easily	Umbilical spillage of feces and urine	None	None	Elliptic incision around the navel	Incidental intraoperative after bleeding from cutting the omphalomesenteric artery
4	Nerdrum et al, 1963 ^[13]	17 days	M	Full term	Not reported	Nonhealing navel flesh colored tumor protruding from the umbilical opening	Not reported	None	Omphalocele	Umbilical circumcision	Incidental intraoperative
5	Griffith et al, 1982 ^[14]	1 month	M	Full term	Not reported	Umbilical granuloma	Umbilical spillage of liquid, mucus, and soft stools	None	None	Elliptic incision around the navel	Incidental intraoperative
6	Alessan-dirini et al, 1992 ^[15]	Not reported	M	Not reported	Not reported	Not reported	Not reported	None	None	Elliptic incision around the navel	Incidental intraoperative
7	Fujiwara et al, 1997 ^[16]	8 months	M	Full term	Povidone iodine	Umbilical granuloma	Umbilical spillage of mucus and yellow liquid	IV Indigo carmine	PU	Pfannenstiel incision	Incidental intraoperative
8	Lizerbram et al, 1997 ^[17]	6 days	M	Not reported	Not reported	Not reported	Umbilical spillage of yellow fluid	Cystogram fistulography MR	Suspected association of PU and POMD	Subumbilical transverse incision	Confirmed intraoperatively
9	Sharma et al, 2011 ^[18]	21 days	F	Full term	Not reported	Intestine prolapsing through the umbilicus	Previous reported spillage of feces and clear fluids	None	POMD	Subumbilical transverse incision	Incidental intraoperative
10	Chawwada et al, 2013 ^[19]	1.5 months	M	Full term	Not reported	Swelling in the umbilical region	Previous reported spillage of feces and clear fluids	None	POMD	Subumbilical transverse incision	Incidental intraoperative
11	Gupta et al, 2014 ^[20]	2 months	M	Not reported	Antibiotic cream and silver nitrate	Umbilical induration	Serous umbilical spillage	US, cystogram	Urachal anomaly	Subumbilical transverse incision	Incidental intraoperative
12	Present case	28 days	M	Full term	Silver nitrate	Nonhealing navel	Granuloma with slight persistent bleeding spillage	US	Suspected association of PU and POMD	Semicircular subumbilical incision	Confirmed laparoscopically

GA= gestational age, POMD= patent omphalomesenteric duct, PU= patent urachus, US = ultrasound.

through a limited (1.5 cm) ileal resection, and an end-to-end anastomosis was performed through the same subumbilical incision.

2.5. Follow-up and outcomes

The postoperative course was uneventful, and the child was then discharged 6 days after the surgery. Six months after the diagnosis, he is in an optimal clinical condition without any complications.

2.6. Timeline

The observation of the clinical data led to the suspicion of an anomaly related to URs. The diagnosis of a PU associated with a POMD duct was confirmed by an immediate abdominal US examination and an elective laparoscopy. During the laparoscopy, a successful minimally invasive excision of the associated anomalies was performed.

3. Discussion

A partial or total failure of the obliteration of the urachus gives rise to various anomalies, which can be discovered in children and adults. The incidence of these anomalies is 1: 5000 live births.^[5] URs can manifest as PU, urachal cysts, urachal sinus, and bladder diverticulum.

Omphalomesenteric duct remnants are the most common anomalies in the gastrointestinal tract and are often asymptomatic.^[6] These anomalies may range from POMD to the more common Meckel's diverticulum. Omphalomesenteric duct malformations may become symptomatic at any age,^[7] and the common symptoms include abdominal pain, intestinal bleeding, intestinal obstruction, umbilical drainage, and umbilical hernia. Infections and tumors are the two main complications of urachal anomalies. In the case of infections, the drainage of the infectious fluids can occur in the bladder, umbilicus, or both. Tumors may be either benign or malignant. Urachal carcinomas are typically silent because of their extraperitoneal location, and, therefore, most of the patients present a diagnosis of local invasion and/or metastasis, which are detected by US and computed tomography (CT).

The diagnosis of these anomalies is rarely based only on US. CT is often the preferred method for a definitive diagnosis. Widni et al affirmed that the use of a US in the diagnosis of URs lacks accuracy for distinguishing true negatives from false positives.^[8] Nevertheless, we think that US can play an important diagnostic role for these types of anomalies if performed by expert physicians. Indeed, in our case, we have highlighted very well the coexistence of the two anomalies.

In the reported case, our patient has been subjected to a treatment with silver nitrate for several days, without any clinical improvement, leading the pediatrician to suggest an examination by a pediatric surgeon. At the surgical outpatient visit, a delay in the healing of the umbilicus was observed. Below the mummified umbilicus, a granuloma with mild bleeding spillage was observed. The possible presence of UR was suspected and was definitively confirmed by US, leading to a diagnosis of a PU, with the very rare simultaneous presence of a POMD. The pediatric surgeons proceeded with a laparoscopic exploration, confirming the suspected diagnosis, and then an excision of both anomalies was performed using a minimally invasive approach with a very small subumbilical incision.^[9]

A review of the literature regarding the association between PU and POMD was performed to confirm the different clinical management and surgical approaches.

We found only 12 cases from 1898 in the English literature,^[10–20] including our last case (Table 1). All cases, except one,^[13] were males. When reported (6/12), all newborns affected by this association were full term. The specific signs reported were the spillage of different materials, such as urine, mucus, soft stools, and/or clear fluid. In all cases, a nonhealing navel was reported in the physical examination in different manners, except for two cases in which one had an evident fistula and the other had a bowel prolapse through the umbilicus. Nevertheless, the possible association between PU and POMD was not suspected in any of these patients and was only evidenced during the surgical excision. The first attempt treatment was reported in 5 cases and consisted of the local application of silver nitrate in 2 cases and application of silver nitrate associated with an antibiotic cream, povidone iodine, and salves, and plasters in 3 cases. The diagnostic examination was performed with i.v. indigo carmine in one case; cystogram, fistulography, and magnetic resonance imaging in another case; US and cystogram in another case; and US only in our case.

The approach to a nonhealing navel in newborns has changed considerably during the last century due to the reduced importance of invasive methods, such as cystography and fistulography. The improving resolution of US over recent decades has led to the consideration of US exams as a first step attempt to define umbilical anomalies. However, since an association of a PU and a POMD is extremely rare, it is also difficult to diagnose. From this perspective, the US approach should aim to evaluate both the URs and other possible associated malformations to better define the anatomy of the region before considering a surgical approach. A laparoscopy can confirm the suspected diagnosis, and, as it is a minimally invasive technique, could be considered a possible gold standard approach to avoid further invasive diagnostic exams with the advantage of having the correct diagnosis and the ability to proceed to the excision of both remnants with a minimal incision.

In conclusion, our case report underlines the importance of evaluating any persisting umbilical lesions without delay when conventional pharmacological therapies fail. A US exam is valuable as a first approach and should be supported by laparoscopy to confirm the diagnosis; a minimally invasive excision of the remnants appears to be an effective therapeutic approach.

4. Patient's parents' perspective

We are very grateful to the surgeons for their diagnostic and therapeutic approach, which was minimally invasive and associated with a favorable clinical evolution.

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