CASE REPORT

Latent vitellointestinal duct sinus presenting with massive lower gastrointestinal bleeding in an adolescent

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SUMMARY

A 12-year-old boy with a history, at birth, of a weeping pink fleshy lesion after his umbilical cord detached, requiring repeated chemical cauterisation, presented with massive lower gastrointestinal bleeding and required resuscitation and blood transfusion. Augmented Tc99m nuclear medicine scan confirmed ectopic gastric mucosa. The lateral view suggested its attachment behind the umbilicus. At exploration, a latent vitellointestinal duct sinus with ectopic gastric mucosal mass was found. Segmental resection of the sinus and mass excision with primary anastomosis and incidental appendectomy was curative. Pink fleshy mass discharging coloured fluid at the umbilicus following detachment of umbilical cord should be considered a remnant of vitellointestinal duct unless proved otherwise. A pink lesion with yellowish discharge resistant to chemical cauterisation should raise the suspicion of embryonic structures. Latent vitellointestinal sinus is a new lesion in the spectrum of umbilical anomalies. Lateral view of the nuclear medicine scan is helpful in locating the site.

BACKGROUND

Most remnants of the vitellointestinal duct and urachus present early in infancy.^{1 2} Massive lower gastrointestinal bleeding secondary to a vitellointestinal sinus containing ectopic gastric mucosa is rare in adolescents.^{3 4} During embryonic life, the vitellointestinal duct connects the yolk sac to the intestinal tract and usually it obliterates by the sixth week of life. If the vitellointestinal duct persists, various congenital anomalies develop, leading to patent vitellointestinal duct, obliterated fibrous cord, umbilical sinus at the outer end, enterocyst in the centre and, most commonly, Meckel's diverticulum on the intestinal end.^{1 3 5} We describe a case of latent vitellointestinal duct sinus towards intestinal end, which has not been described earlier in the literature, with a unique ectopic gastric adenoma mass just below the umbilicus presenting with recurrent massive lower gastrointestinal bleeding.

CASE PRESENTATION

A 12-year-old boy presented with central abdominal pain and recurrent massive lower gastrointestinal bleeding with pallor and fainting. At the initial event of periumbilical pain and fresh bleeding, he was taken to his general practitioner and no local perianal cause was found. He and his parents were reassured that he must be wiping his bottom

hard and should not worry about it as it was self-limiting, and they were sent home.

After a few days the boy started having similar periumbilical pain associated with persistent massive fresh lower gastrointestinal bleeding and was taken to the paediatric accident and emergency department of our hospital. On arrival, he was very pale, tachycardic, hypotensive, and very anxious and apprehensive.

Abdominal examination showed scarred umbilicus with a palpable mass measuring 3×2×1 cm just below the umbilicus, which was tender to touch. The rest of the abdominal examination was normal.

Further questioning revealed that at birth he had had a pink fleshy mass lesion at the belly button site, which was oozing yellowish fluid, at times with severe excoriation around the umbilicus. The lesion required repeated silver nitrate cauterisations by a dermatology specialist, which lasted over 2 months, but then it had healed with scarring. The boy had recurrent short lasting periumbilical pain that would eventually settle down.

INVESTIGATIONS

The patient's urine dipstick, microscopy and culture were normal. Biochemical investigations including renal functions were within normal limits. C reactive protein measured 18 mg/L. Haematological investigations showed haemoglobin 58 g/L, red blood cells $2.41 \times 10^{12}/L$, haematocrit 0.179, mean corpuscular volume (MCV) 74 fl, mean corpuscular haemoglobin (MCH) 23 pg, platelets $392 \times 10^9/L$, white blood cells $5.51 \times 10^9/L$ and neutrophils $3.99 \times 10^9/L$. Abdominal and pelvic ultrasound scan showed a mixed echogenic mass just below, and attached to, the umbilicus.

A Tc99m nuclear scan showed presence of large ectopic gastric mucosa in the central abdomen on anteroposterior view and just behind the umbilicus on the lateral view, and possibility of remnant of vitellointestinal duct with ectopic gastric mucosa communicating with ileum was considered.

DIFFERENTIAL DIAGNOSIS

Bleeding Meckel's diverticulum, enterocyst, duplication cyst with ectopic gastric mucosa, inflammatory bowel conditions, etc, were considered in the differential diagnosis.

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TREATMENT

The patient was resuscitated with three boluses of normal saline and required three packed red cells to correct anaemia and hypotension. He was kept nil by mouth and started with intravenous fluids, ranitidine and paracetamol. His bleeding started coming under control.

He was taken to the operating room and examination under anaesthesia revealed a 3 cm firm mass attached to the scarred umbilicus. Supraumbilical exploration showed a dense mass of ectopic gastric mucosa with a patent rest of the vitellointestinal sinus entering terminal ileum. The mass was resected from behind the umbilicus and a segmental resection of the terminal ileum containing vitellointestinal sinus and adenomatous mass at the tip with end-to-end ileoileostomy and incidental appendicectomy concluded the operation.

OUTCOME AND FOLLOW-UP

His postoperative period was uneventful and he was discharged home on third postoperative day. The resected specimen contained a tubular piece of vitellointestinal remnant measuring 60×25 mm with an attached adenomatous mass measuring 50×20 mm. On cut section, background terminal ileum and remnant mucosa appeared unremarkable but close to the mass at the tip there was a small dark ulcerated area suggestive of the possible site of bleeding. Microscopic examination confirmed ectopic gastric mucosa in the mass and chronic inflammation in the lamina propria and one area with mucosal ulceration with marked acute inflammation. The resection margins and terminal ileum were normal. An incidental appendicectomy specimen showed normal histology of appendix. He was asymptomatic and well at 1-year follow-up and discharged from the clinic.

DISCUSSION

In the early embryo, the vitellointestinal duct provides nutrition, but once the placental nutrition is established, the duct gets obliterated by the end of the seventh week of intrauterine life. In approximately 2% of humans the remnants of the duct persist in the form of a spectrum of which Meckel's diverticulum is the commonest and patent vitellointestinal duct is the rarest.^{1 3}

Our case had an umbilical pink fleshy mass and discharge at birth and represented patent vitellointestinal duct. However, it was mistaken for umbilical granuloma and repeated chemical cauterisation with silver nitrate led to scarring and closure of the umbilical end but it remained patent internally with a communication with terminal ileum. Periumbilical excoriation at birth and palpable mass just behind the scarred umbilicus should alert to the possibility of an ectopic gastric mucosa as it is generally seen at the tip of the vitellointestinal duct remnant.

Umbilical sinus with or without a band is a very well known remnant but latent vitellointestinal sinus open at the intestinal end is a new lesion in the spectrum of umbilical anomalies and has not been described earlier. This is partly congenital due to acid irritation from the ectopic gastric mucosa very close to umbilicus and partly iatrogenic due to repeated chemical cauterisation with silver nitrate by overenthusiastic paediatric dermatologists. A high index of suspicion may help diagnose this condition in early infancy and at that age a transumbilical excision would give less morbidity and better cosmetic results.

Massive fresh lower gastrointestinal bleeding is generally seen in young infants but it can also be a presentation in adolescents. This signifies a sinister organic problem rather than a self-limiting illness and patients should be immediately referred to specialists rather than reassured and sent home, especially if

there is no local cause to explain the illness. Background detailed history and findings on clinical examination should help in considering the possibility of vitellointestinal remnant with ectopic gastric mucosa.

Vitellointestinal remnants may remain asymptomatic but rarely may present dramatically, especially in the first few years of life or, more rarely, in adolescent as in our case or in adults. The complications are generally serious and are commonest in infants and young male children.^{6 7}

H2 blockers may not only help therapeutically in controlling acute bleeding from the ectopic gastric mucosa but they also augment the nuclear medicine scan and enhance its sensitivity and specificity. Proton pump inhibitors are used to control bleeding in an emergency, thus allowing time for resuscitation and preparation.

Laparoscopy has been used for diagnostic and therapeutic purposes for these types of lesion recently.⁸ However, in our case the large palpable mass just below the umbilicus precluded this modality. The mass being larger than the umbilical ring did not allow us to use the transumbilical route, which is very easily used in infants with patent vitellointestinal duct or umbilical sinus with clear tissue planes. A periumbilical approach is reasonable, safe and cosmetic, and resection of the lesions and incidental appendicectomy can easily be performed through this approach.

Learning points

- ▶ A pink fleshy mass discharging coloured fluid at the umbilicus following detachment of umbilical cord should be considered a remnant of vitellointestinal duct unless proved otherwise and should be referred to paediatric surgical specialists.
- ▶ Although umbilical granuloma is very common, a pink lesion with yellowish discharge resistant to chemical cauterisation should raise the suspicion of embryonic structures.
- ▶ Latent vitellointestinal sinus is a new lesion in the spectrum of umbilical anomalies due to remnants of vitellointestinal duct and has not been described earlier.
- ▶ Massive fresh lower gastrointestinal bleeding is generally seen in young infants but it can also be a presentation in adolescents; patients should immediately be referred to a specialist rather than reassured and sent home.
- ▶ H2 blockers help in temporarily controlling bleeding due to ectopic gastric mucosa, and they also help in enhancing the Tc99m scan by increasing its sensitivity and specificity. The lateral view of the nuclear medicine scan is helpful in locating the site.

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