CASE REPORT

Non-puerperal uterine inversion in a patient with intracranial sigmoid sinus thrombosis and facial palsy

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Accepted 17 February 2015

SUMMARY

Non-puerperal uterine inversion and cranial nerve palsies in patients with sigmoid sinus thrombosis are both extremely rare. We report a case of a patient who presented with both simultaneously. The symptoms of sigmoid sinus thrombosis resolved with subcutaneous enoxaparin, and an abdominal dissection with removal of the uterus vaginally resulted in a successful outcome for the patient.

BACKGROUND

Uterine inversion is rare and non-puerperal uterine inversion is so rare that it is impossible to estimate its incidence.¹ To date, only about a hundred cases of non-puerperal uterine inversion have been reported in the medical literature.² The incidence of thrombosis of cerebral veins and sinuses is 3–4 per million,³ and the incidence of cranial nerve palsy in a patient with sigmoid sinus thrombosis is extremely rare.⁴ We report possibly the first case in the medical literature of non-puerperal uterine inversion in a patient with intracranial sigmoid sinus thrombosis, and the clinical symptoms of both conditions evolving simultaneously. The patient was treated with low-molecular-weight heparin (LMWH; enoxaparin), and the uterine inversion was surgically managed by hysterectomy for an infected fundal leiomyoma and irreducible inversion. Informed consent was taken from the patient for publication of this case.

CASE PRESENTATION

A 42-year-old woman (para 4) attended our obstetrics and gynaecology emergency department with a red, congested, infected and irreducible prolapsed mass per vaginum with generalised headache and malaise for 2 weeks, along with left lower motor neuron facial palsy for 1 week, and progressive double vision worsening with left gaze. The headache progressively increased in severity and was associated with vomiting. Seven days after the onset of headache, the patient noticed difficulty in closing her left eyelid followed by deviation of the angle of her mouth to the right. She had four full-term vaginal deliveries, the last being 13 years earlier. She did not give any immediate history of fever, seizures, ataxia or other otological symptoms such as ear pain, drainage of fluid from the ear or diminished hearing.

She was afebrile on admission and except for mild conjunctival pallor, all her vital signs were within normal limits. Neurological examination was normal except for left peripheral cranial nerve palsy of the abducens and facial nerves (VI and VII). Her visual

acuity was normal (10/10 bilaterally), visual field testing and dilated fundus examination did not reveal any abnormality. There was no clinical evidence of myasthenia gravis or thyroid ophthalmopathy. Blood tests revealed raised inflammatory markers with neutrophilia (white cell count $15.7 \times 10^9/L$ with a neutrophil count of $12.7 \times 10^9/L$). Lumbar puncture opening pressure, cerebrospinal fluid (CSF) examination and routine blood chemistry were normal.

On per-vaginal examination, a circumferential solid mass, approximately 8 cm in diameter, was seen protruding through the introitus. The mass was red, congested and appeared infected. The cervix could barely be palpated. The uterus was not palpable on a rectovaginal examination.

The patient was started on intravenous ceftriaxone and metronidazole empirically. Ultrasound showed absence of uterus in the pelvic cavity and an empty bladder postvoiding. The provisional diagnosis of uterine inversion, possibly caused by the intra-uterine polypoidal mass, led the treating team to suggest an abdominal hysterectomy for the patient.

When a CT of her brain for persistent headache and left peripheral cranial nerve palsy of the IV and VII failed to reveal any abnormality, MRI of the patient's brain was performed, which only revealed left sigmoid sinus thrombosis.

Haemoglobin was 9.4 g/dL, with hypochromic normocytosis (mean corpuscular volume 68.6 fL, mean corpuscular haemoglobin (MCH) 21.4 and MCH concentration 31.1). Total and differential leucocyte count and platelet and reticulocyte count were normal. Antinuclear antibody, anti-double stranded DNA and antiphospholipid antibodies were negative.

The patient was started on subcutaneous enoxaparin 60 mg twice daily. Her headache started gradually resolving from the third day after starting enoxaparin and completely resolved by the 10th day. Her facial palsy also started resolving by the 3rd day, and by the 10th day there was only slight deviation of the angle of her mouth to the right, suggesting mild residual facial nerve paresis.

She was taken up for an abdominal hysterectomy under general anaesthesia on the 14th day after starting enoxaparin for sigmoid sinus thrombosis, after she was off subcutaneous enoxaparin for 24 h. The abdomen was opened by making a Pfannenstiel incision. On opening the abdomen, there was absence of the uterus in the abdominal cavity and there was no constriction ring as the cervix was also completely inverted and could not be delineated separately from the vagina (figure 1). Owing to the large infected polypoidal mass protruding outside

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To cite: Datta MR, Nag DS, Kabiraj M. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-206937

Figure 1 Absence of the uterus in the abdominal cavity.

and irreducibility of the inversion, the decision was taken to deliver the uterus vaginally without correcting the inversion. The round ligament was identified and ligated and the fold of peritoneum from that point was incised upwards towards the paracolic gutter. This was performed to trace the ureters downwards from the pelvic brim. To facilitate tracing of the ureters, the anterior leaf of the broad ligament was opened from the point where the round ligament was cut and ligated. The ovarian vessels were identified and ligated. The uterine artery was then identified at its point of crossing the ureter and was cut and ligated. As there was complete inversion of the uterus and cervix, and part of the vaginal vault, no cervical rim was identifiable intra-abdominally. The polypoid mass and the inverted uterus were pushed upwards by an assistant vaginally and this caused the appearance of an elevated rim of the vaginal wall. The urinary bladder was pushed down for an approximate distance of 1.5 cm after opening the vesicouterine pouch. An incision was then made on the elevated vaginal rim to open the vault. The incision on the vaginal vault was extended sideways and posteriorly making sure that the line of incision was above the ureters laterally. When the uterus was thus separated, it was pushed down and out through the vagina and the vault was closed ([figure 2](#)). As the vault tissues were very lax, it was postulated that an abdominal sling for vault suspension would prevent vault prolapse. So, a rectus sheath sling was put in the vault.

Figure 2 Removed uterus with polypoid mass.

When subcutaneous enoxaparin was restarted 48 h postoperatively, there was mild vaginal bleed on the third day postoperatively. Enoxaparin was stopped and the diffuse ooze gradually stopped on the fifth postoperative day. Subcutaneous Enoxaparin was restarted on the seventh postoperative day, and after the next two uneventful days (9th postoperative day), the patient was discharged, and followed up by the gynaecologist and neurophysician in the outpatient department. She was subsequently converted to anticoagulation with warfarin for her sigmoid sinus thrombosis. The pathologist found that the specimen had an elongated cervix reaching onto an inverted uterus with a large subserosal fibroid measuring $7.5 \times 6 \times 5.4$ cm on gross examination. Histopathological examination of sections from the cervix showed superficial ulceration with non-specific inflammatory granulation tissue and non-preserved epithelial lining; uterine wall showed non-secretory endometrial lining and a normal myometrium. The tumour was consistent with characteristics of a leiomyoma. No thrombus in vessels of infected fibroid polyp could be noted. Warfarin was stopped at 6 months and on follow-up for 1 year, the patient had no medical problems.

DISCUSSION

With only about 100 cases reported in the medical literature, non-puerperal uterine inversion is so rare that its exact incidence cannot be determined.^{1 2} Uterine leiomyomas are the commonest cause of non-puerperal uterine inversion, but in women under the age of 45 years, the commonest cause is malignancy.⁵ Although it can be idiopathic, there are various other causes such as uterine sarcomas, endometrial cancer and benign polyps.² In our patient, uterine leiomyoma was the cause of the uterine inversion. Although there is no pathological association reported in the medical literature, the patient presented with clinical features of sigmoid sinus thrombosis and uterine inversion simultaneously.

The incidence of single and multiple cranial nerve palsies from isolated unilateral thrombosis of the transverse/sigmoid sinus is extremely rare.⁶ The various causes of sigmoid sinus thrombosis include a wide spectrum of aetiologies consisting of infective causes including otitis, mastoiditis, sinusitis, meningitis and systemic infections, non-infective causes such as inflammatory diseases, head injury, brain tumours or anticoagulation disorders, or they are of unknown aetiology.^{4 6} Although a raised leucocyte count with neutrophilia could be due to a systemic infection, in the absence of any inflammatory cells in a CSF examination, a corresponding central nervous system infection causing the raised leucocyte and neutrophil counts seems unlikely. In our patient, although blood tests revealed raised inflammatory markers, in the absence of any fever, it would be most likely due to the localised infection of the prolapsed uterus and polypoid fibroid. The cause of the cerebral venous thrombosis remained unknown as no systemic infectious disorders or thrombophilia could be established by the diagnostic tests available at our hospital.

A lumbar puncture was performed subsequent to the absence of papilloedema on a fundus examination. The necessity of performing a CT before carrying out a lumbar puncture remains controversial, especially because a “normal CT scan does not prove that ICP (intracranial pressure) is also normal” and up to 50% of patients with elevated intracranial pressures will have a normal CT scan.⁷ Although some authors do recommend lumbar puncture in cerebral vein and dural sinus thrombosis,⁸ it does appear, retrospectively, that in the absence of a strong suspicion of meningitis, “examination of the CSF is typically not helpful”.⁹

In cerebral venous thrombosis, the cranial nerve lesions are usually caused by 'venous congestion' and the resultant oedema of the surrounding areas.⁶ As the insult is usually due to "reversible compromised oxygen and glucose consumption"⁶ and not 'venous infarction',⁶ possibly this was also the cause of the early resolution of the symptoms after initiation of therapy. Kuehnen *et al*⁶ observed that the cranial nerve dysfunction is temporary until thrombolysis occurs, either "spontaneous or accelerated by anticoagulation". Although controversial, the sigmoid sinus thrombosis was managed with anticoagulation with LMWH as a 'bridge to oral anticoagulation with a vitamin K antagonist' based on current guidelines.¹⁰ We followed the American College of Chest Physicians guidelines¹¹ on the perioperative management of antithrombotic therapy by stopping enoxaparin 24 h before surgery. The same guidelines of 'resuming therapeutic-dose LMWH 48–72 h after surgery' for patients who are at a high bleeding risk were followed in restarting enoxaparin 48 h after surgery. However, this resulted in diffuse vaginal ooze, which resolved with stopping enoxaparin.

Among the various surgical techniques that have been described for non-puerperal uterine inversion, the abdominal approach described by Huntington and Haultain, and the vaginal approach described by Kustner and Spinelli have both been successful.^{12–13} Owing to our concern for the location of the ureters and the inability to revert the uterus, we employed an abdominal approach but delivered the uterus vaginally without repositioning the uterus to its normal anatomical site. Although difficult, a successful attempt of vaginal restoration and removal has been

reported in the literature.² Sometimes, intraoperative cystoscopy may be required to ensure bladder and ureteral integrity.² We have described an approach where the inverted uterus could not be restored to its original anatomical site, but after surgical dissection abdominally, the uterus was removed vaginally without contaminating the abdominal cavity. Despite the clinical impression of a polypoidal mass being a leiomyoma, it is prudent to send the specimen for histopathology, given the fact that malignancy is the commonest cause of non-puerperal uterine inversion in women under the age of 45 years.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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Learning points

- Non-puerperal uterine inversion and cranial nerve palsies in patients with sigmoid sinus thrombosis are both extremely rare and awareness about them is the key to their diagnosis.
- Management of sigmoid sinus thrombosis with anticoagulants raises perioperative concerns about the risk of bleeding.
- For non-puerperal uterine inversion, we describe an approach where surgical dissection was performed abdominally and the uterus was delivered by the vaginal route.

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