

Bilateral vulval emphysema: A rare complication of laparoscopic hysterectomy

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ABSTRACT

Vulvar edema can present in various obstetric and gynecological conditions in a patient. There are instances of it presenting after tocolysis of pre-term labor, secondary to ovarian hyperstimulation syndrome, as a complication of severe pre-eclampsia; however, vulvar edema presenting after laparoscopy or intra-operatively during laparoscopy is rarely reported. A 44-year-old patient, P5L5 with a previous 1 lower segment cesarean section and 4 vaginal birth after cesarean presented with abnormal uterine bleeding due to leiomyoma, for the past 8 months, which was not responding to medication. A total laparoscopic hysterectomy was planned. On cauterizing and cutting off bilateral round ligaments, a bilateral swelling was noted over both the labia Majora, which was reducible on manual compression. It was concluded to be due to the bilateral patent canal of nuck. It may be more prevalent in asymptomatic adults than currently appreciated. Our case is the only one documented in the literature so far of a bilateral patent canal of nuck, presenting in an asymptomatic adult. Only one other similar case of a unilateral patent canal of nuck has been reported. In conclusion, vulvar edema following a laparoscopic surgery is uncommon and is usually treated conservatively. However, in our case, the vulvar edema (emphysema) was evident intra-operatively, which was bilateral, due to the patent canal of nuck, which is a very uncommon presentation and difficult to diagnose pre-operatively, especially in adults who doesn't present with any signs till adulthood.

Key words: Canal of nuck, Laparoscopic hysterectomy, Vulval edema, Vulval emphysema

Abnormalities involving the canal of nuck are uncommon developmental issues affecting female genital anatomy. These defects are typically diagnosed and surgically corrected in early childhood, most often within the first 5 years of life. The first documented case was described by Anton Nuck in 1691 [1]. When the canal does not naturally close after birth or during the 1st year, it can remain patent, which may lead to either hydrocele or hernia through this channel [2]. A bilateral patent canal of nuck presenting for the 1st time intra-operatively in a previously asymptomatic adult is very rare. With one reported case thus far in the literature [3]. We are presenting this case, which is a rarity, to prompt clinicians to keep this differential in mind while coming across such a presentation in a previously asymptomatic adult.

Vulvar edema can present in various obstetric and gynecological conditions in a patient. There are instances of it presenting after tocolysis of preterm labor, secondary to ovarian hyperstimulation syndrome, as a complication of severe pre-eclampsia, and it is also reported in patients of ascites of hepatic origin [4-7]. Vulvar edema presenting after laparoscopy or intra-operatively during laparoscopy is rarely reported [8-11].

We present a case of bilateral vulvar edema (emphysema) presenting intra-operatively during laparoscopic hysterectomy due to bilateral patent canal of nuck, which may be the only reported case thus far.

CASE REPORT

A 44-year-old patient, P5L5 with a previous 1 lower segment cesarean section and 4 vaginal birth after cesarean presented with abnormal uterine bleeding, for the past 8 months.

On general physical examination, the patient had mild pallor, hemodynamically stable, with a heart rate of 90/min, SpO₂ of 98% in room air, and blood pressure of 130/82 mmHg. On per-vaginum examination, a uterus of 10 weeks was identified.

On investigating the patient, her hemoglobin was found to be of 9 g%, with a normal platelet count. Peripheral smear showed microcytic hypochromic anemia. Her other laboratory investigations were found to be normal, including her thyroid profile. An ultrasound scan was done, which revealed a leiomyoma (FIGO Type 2) of 6.2 × 5.5 × 4.8 cm³ in the posterior myometrium. She was already started on medications by a private

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gynecology specialist for the past 6 months, to which she had not responded; hence, a total laparoscopic hysterectomy with bilateral salpingectomy was planned.

The patient was operated under general endotracheal anesthesia, in dorsal lithotomy position, in steep Trendelenburg. Pneumoperitoneum was achieved by Veress needle insertion through the lower margin of the umbilicus. Intra-abdominal entry was confirmed by noticing the two-click sound and the intra-abdominal pressure in the insufflator. CO₂ was used for insufflation. Intra-abdominal pressure was set to 15 mmHg. For primary trocar entry, the skin wound of Veress needle insertion was extended to 11 mm size using an 11-number blade. A reusable trocar with a cannula was used, the primary trocar was inserted, and intra-abdominal insertion was confirmed by the whooshing sound and loss of resistance. A 10 mm telescope was inserted from the primary cannula. Omental adhesions with the anterior abdominal wall were seen at multiple sites. Two working ports were inserted on the left abdominal wall under direct visualization after ruling out any omental adhesions. To reach up to the uterus and adnexa, omental adhesiolysis was done using a hybrid energy device (ultrasonic and advanced bipolar energies; Thunderbeat Olympus). Once the uterus and adnexa were visualized, another assisting port was inserted on the right abdominal wall under direct visualization, about 10 cm away from the primary trocar. Bilateral tubes and ovarian ligaments were cauterized and cut. Bilateral round ligaments were cauterized and cut using a hybrid energy device. Before proceeding to the next steps, a bilateral swelling was noted over the labia Majora. The variation was significant compared to the patient's pre-operative status. The skin in the lower abdominal region, groin, and mons pubis did not appear swollen. There was no bleeding and hematoma was ruled out for there was no skin discoloration. The patient had stable vitals. The labia Majora was palpated and it revealed an air-filled swelling. The total reduction of the swelling was achieved by direct gentle pressure over the labia Majora. The telescope was inspecting the internal surface of the abdominal wall and pelvis, where the bladder was separated from the adhesions with the anterior abdominal wall and uterus. It did not reveal any blood or exudate ruling out any peritoneal-cutaneous fistula. The swelling reappeared within a few minutes of removal of the manual pressure over the labia Majora (Fig. 1).

Due to this finding, which was noticed immediately after cutting the round ligaments, insufflation was stopped, and the further steps of hysterectomy (with perineorrhaphy done for relaxed perineum) were done by vaginal route. Both the ovaries were preserved as they were normal and keeping in mind the patient's age. After the hysterectomy was complete, it was decided to explore the inguinal region and the region of insertion of the round ligaments again; no gross abnormality was noticed. However, externally, the emphysema appeared again, bilaterally. The conclusion of the presence of a bilateral patent tract extending from the peritoneum till the insertion site of the labia Majora was made. After ensuring hemostasis, the insufflation was stopped, trocars were removed, and the abdomen was brought back to



Figure 1: Bilateral edema (emphysema) of labia Majora (left more than right) due to bilaterally patent canal of nuck

normal pressure. Skin incisions were closed. The patient was shifted to the gynecology ward for post-operative care. The post-operative period was uneventful.

DISCUSSION

Anton Nuck, a Dutch anatomist, first described the condition in 1691, and this uncommon anatomical finding was named as canal of nuck.

It is the homologous structure of "processus vaginalis" in males. The parietal peritoneum enclosing the ligamentum rotundum or round ligament that descends through the inguinal canal during embryological development and connects to the ipsilateral labium majus remains patent in certain populations. This generally occurs between the 7th month of gestation to 1 year of age [12]. A partially obliterated canal of nuck usually presents before 5 years of age as hydrocoele or inguinal hernias [12,13]. It is a rarity of 0.76% as found in a large series of 787 inguinal surgeries [2]. A patent canal of nuck often presents as a painless vulvar swelling in adolescence or childhood, but may be more prevalent in asymptomatic adults than currently appreciated [13,14]. Description of the hydrocoele of the canal of nuck in adults seeking surgical correction for groin swelling, the condition's rarity often precludes diagnostic imaging like magnetic resonance imaging, leaving its prevalence in asymptomatic adults largely unknown [2,12,15]. Most of the case reports found online are of the unilateral patent canal of nuck [3]. However, our case demonstrates how the bilateral patent canal of nuck can unexpectedly manifest as labial swelling following insufflation during laparoscopic abdominal surgery. After a vast literature search, we have concluded that the case we are presenting is the first case documented so far, of the bilateral patent canal of nuck identified in an asymptomatic adult, intra-operatively.

Vulvar pneumatocele, a rare occurrence during laparoscopic procedures with limited documented references, shares a similar pathophysiology to scrotal pneumatocele. The proposed mechanism involves air pressure tearing weakly adherent walls of the processus vaginalis, which aligns with the observation of

pneumatocele formation during laparoscopy and its subsequent resolution after surgery [16].

While vulvar edema, as a complication of laparoscopic surgery, was first reported in 1996. All the cases were presented 24 h post-operatively. The causes, according to the investigators, were attributed to escaped Ringer's lactate fluid through some suprapubic puncture site, as a result of adhesiolysis and/or improper trocar placement, which might have escaped into subcutaneous tissue either intra-operatively or post-operatively. Moreover, vulva being the most dependent area and accessible, the edema was evident in this region [10]. All these cases were self-limiting but required admission due to the patients' inability to self-void urine. However, all were managed conservatively with an ice-pack and Foley's catheterization. These are unlikely in our case due to the resolution of swelling after discontinuing insufflation of the abdomen and the absence of post-operative edema. This case emphasizes the potential role of a patent canal of nuck in the development of unexpected labial swelling during or after laparoscopic surgeries. In another report, severe vulvar edema was noted 24 h post-operatively, which, according to the investigators, was due to the escape of anti-adhesion fluid through one of the lower 5 mm trocar sites. This patient too, was managed conservatively in the hospital with urinary in-dwelling catheterization for 9 days [9]. As no anti-adhesion fluid was used in our case and there was no improper trocar placement, as confirmed on entry into the abdominal cavity, our case of vulvar edema was due to pneumatocele, as said above.

CONCLUSION

Vulvar edema following a laparoscopic surgery is uncommon and is usually treated conservatively. However, in our case, the vulvar edema (emphysema) was evident intra-operatively, which was bilateral, reducing manual compression and self-resolving on stopping insufflation. Hence, it was a case of patent canal of nuck, which is a very uncommon presentation and difficult to diagnose pre-operatively, especially in adults who do not present with any signs till adulthood. Hence, a unilateral or bilateral patent canal of nuck should be on the differentials of cases presenting with swelling in the vulval or inguinal region undergoing laparoscopic surgery. Conservative management can be done in such cases that do not present with any complications.

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