# CASE REPORT

# Management of Spontaneously Occurred Isolated Hematocervix Case: A Rare Pathology

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**ABSTRACT** Hematocervix is a very rare condition in which blood or blood clot is found in the cervix. It usually occurs after a surgical procedure involving the cervix. Hematocervix presents with lower abdominal pain and can be diagnosed with transvaginal ultrasound. A 45-year-old patient with no risk factors for hematocervix was admitted to the outpatient clinic with new-onset amenorrhea and severe abdominal pain. Ultrasonography and magnetic resonance imaging showed a fluid collection in the cervix. The patient was treated with simple dilation and drainage under general anesthesia. The diagnosis, follow-up, and treatment are presented with a review of the literature.

Keywords: Abdominal pain; amenorrhea; hematocervix; hematometra

Hematometra is defined as a rare pathology in which blood or blood clot is found in the uterine cavity. It can be acquired or congenital. Congenital hematometra, which is seen more frequently, is mostly caused by imperforated hymen and transverse vaginal septum.<sup>1</sup> Endocervical atrophy, uterine synechiae, radiation exposure, space-occupying lesions of the endocervix or cervix, and surgical interventions on the cervix are some of the acquired causes of hematometra. The incidence of hematometra varies depending on the etiology. According to the literature, congenital hematometra is seen in approximately 1 of 1,000-2,000 teenagers.<sup>2</sup> Among the acquired causes, the rate of hematometra after conization has been shown to be 1.7%.<sup>3</sup>

If the cervix is the only site where the blood collection is seen, the pathology is defined as hematocervix.<sup>1,4</sup> Hematocervix is a less common condition than hematometra, and only case reports have been described in the literature about the pathology.<sup>4</sup> Patients usually present with lower abdominal pain and ultrasonography is often sufficient for diagnosis.

There is no consensus on the effective treatment method for hematocervix cases. Incision applied to the external cervical ostium, simple dilation and drainage of the blood collection, and hysterectomy are the various treatment methods that can be applied in hematocervix cases.

In this study, we present a case with isolated hematocervix with no recent history of cervical or uterine surgery, who applied to our clinic with severe abdominal pain and was successfully treated with simple dilation. The diagnosis, follow up and treatment approaches are presented with the review of the literature.



## CASE REPORT

Written informed consent was obtained from the patient for the publication of this case report.

A 45-year-old patient (gravida 2, parity 2) was admitted to the outpatient clinic with severe abdominal pain lasting for 3 days. The patient had no systemic disease or drug use. She had had regular menstrual periods until the last 3 months but had amenorrhea ever since. There was no history of cancer in the family and no history of space-occupying lesions of the lower genital tract. The result of the cervical Pap-smear test taken 1 year ago was seen as benign cellular changes.<sup>5</sup> The patient, who had a previous history of two cesarean sections and an umbilical hernia repair, did not have a history of any intervention to the cervix.

The vital signs were stable. Abdominal examination revealed minimal tenderness in the lower abdomen without any palpable mass. On vaginal examination, the cervix was seen edematous and tight, the external cervical ostium was noted to be stenotic.

Laboratory tests showed hemoglobin 11.3 g/dL, hematocrit level 35.2%, white blood cell (WBC) count 6,500/uL, C-reactive protein (CRP) 29.8 mg/L. The negative blood pregnancy test and normal biochemical and coagulation markers were seen.

Ultrasound examination showed an adenomyotic uterus with a linear endometrium of 7 mm thickness. No pathology was detected in the bilateral adnexa. An enlarged cervix with a fluid collection of 5x6 cm in size, with mixed echogenicity and thick septations, was observed. No blood flow on Doppler was seen. Pelvic magnetic resonance imaging showed a cervical cystic lesion with septations, 5.8x5.3 cm in size (Figure 1). The lesion was interpreted in favor of a complicated nabothian cyst of the cervix. The patient was started on antibiotic therapy. Examination and drainage under anesthesia were planned.

Initially, 200 µg vaginal misoprostol (Cytotec® 200 µg, Ali Raif Ilac, İstanbul, Türkiye) was administered. 2 hours later, under general anesthesia, after the preparation of the surgical site with an antiseptic solution, a tenaculum was placed on the anterior cervix. Under ultrasound guidance, the external cervical os was passed with a hysterometer. The hysterometer was observed within the cystic structure detected by imaging in the cervical canal. Meanwhile, a blood clot, which is the probable reason for the obstructed external cervical ostium, was seen mobile with the movement of the hysterometer in the ultrasound (Figure 2). After the external cervical os had passed, a dense, chocolatelike fluid began to drain. The cervix was dilated until 4 mm with hegar dilators. The removal of the collection was done with suction curettage. In total, the amount of drained blood was 100 cc. Endocer-



FIGURE 1: Magnetic resonance image in hematocervix case.



FIGURE 2: The hysterometer can be seen within the cystic structure in the cervical canal-arrow, the blood clot, which is the probable cause for the obstructed external cervical ostium-arrowhead.



FIGURE 3: Postoperative ultrasonographic image of the hematocervix case.

vical and endometrial sampling had performed after the cervix was dilated until 7 mm. During sampling, the surrounding cervix was found to be extremely soft. No bleeding or early complications were observed. A cervix and uterine cavity with a normal appearance were observed in postoperative ultrasonography.

Antibiotic therapy was administered during the postoperative follow-up. Oral intake started on the first postoperative day. No complications were observed during follow-up. The patient was discharged with recommendations after no decrease in hemo-globin levels and no progression in WBC count was observed. CRP level was seen as 12.1 mg/L.

After 3 weeks, the patient was called to the outpatient clinic for examination. Ultrasound examination showed a normal cervix and uterus, blood collection in the cervical canal was not observed (Figure 3).

### DISCUSSION

Hematometra and hematocervix are rare conditions that should be considered in the differential diagnosis of women who present with new-onset amenorrhea and abdominal pain after previous cervical surgery or uterine ablation. Hematometra can be diagnosed with the visualization of pockets of echogenic fluid in the uterine cavity on transvaginal ultrasound.<sup>4</sup>

In the literature, data on isolated hematocervix have been reported only as case reports. In these cases, the pathology is generally seen after surgical intervention to the cervix or the uterus.<sup>6-10</sup> Hematocervix is seen more frequently secondary to cold knife conization, especially in cases where suturing is used for hemostasis. However, widespread use of electrocauterization has led to a further decrease in the incidence of hematocervix cases observed.<sup>4</sup> During our literature review, we have not encountered a case of spontaneously developed isolated hematocervix. We do not have information about what caused cervical stenosis in this case, why it occurred at this time, and why it resulted in hematocervix instead of a more common pathology, hematometra. In this respect, as far as we know, this case is the first in the literature.

Various treatment strategies have been applied in the management of hematocervix. Hysterectomy was performed as the first-line treatment in two of the previously reported cases.<sup>6</sup> Although hematocervix drainage was successfully applied, hysterectomy was the definitive treatment in two cases because of the accumulation of fluid again.<sup>10</sup> Cases in which the simple dilation procedure was sufficient to have also been reported in the literature. As another treatment method, drain placement after simple dilation followed by insertion of a copper intrauterine device (IUD) to prevent restenosis was successfully applied in some cases.<sup>4</sup>

The high rate of hysterectomy after simple dilation and drainage, and the excellent results of IUD placement for providing cervical patency indicate that this method can be a valid method for treating hematocervix. In the case reported in this study, simple dilation and drainage were successfully performed, and no IUD was used afterward. Restenosis was not observed in the subsequent controls of the patient. This shows us that this simple treatment method can be an adequate treatment approach in cases of isolated hematocervix that occur spontaneously. In this way, possible complications such as perforation or infection, which may be caused by the application of an IUD to the softened cervix and uterus, can be prevented.

In conclusion, hematocervix is a very rare condition and very few cases have been described so far. Although the cases usually occur secondary to a previous uterine or cervical surgical procedure, hematocervix can also occur spontaneously in a patient who applies to the clinic with abdominal pain; hence, it should be considered in the differential diagnosis. Simple dilation can be a successful treatment option; however, in recurrent cases, utilizing the benefits of an IUD to prevent restenosis is an option as well.

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#### **Conflict of Interest**

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

#### Authorship Contributions

Idea/Concept: Samican Özmen; Control/Supervision: Erkan Çağlıyan; Data Collection and/or Processing: Egehan Bilen; Analysis and/or Interpretation: Aslı Akdöner; Writing the Article: Samican Özmen; Critical Review: Onur Yavuz.

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