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REVIEW

Perforation of the Cervix by the Strings of an Intrauterine Device (IUD): a Novel Case and Systematic Review of the Literature

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ABSTRACT

Purpose: *Since their introduction, intrauterine devices (IUDs) have been associated with various complications. A very rare complication is the perforation of the cervix by the strings of the IUD. The purpose of the current study is to present a novel case of cervical perforation by the strings of a copper IUD and to perform a systematic review of the literature.*

Materials and methods: *A 43-year-old female patient attended the gynecology clinic in order to remove her copper IUD. Speculum examination revealed that both strings of the IUD had perforated the anterior lip of the cervix. Management options were offered and the patient opted for a hysteroscopic removal. We searched several electronic databases, including MEDLINE, Cochrane Library, Google Scholar, and EBSCO, in order to find similar cases.*

Results: *The electronic search yielded 1 821 articles, of which eight were selected for inclusion. The mean age of women was 35.37±7.781 (range 26-47) years. One woman (12.5%) was nulligravida, and three women were multigravida (37.5%). Seven women (87.5%) were asymptomatic. Of all IUDs, three (37.5%) were LNG-IUS and five (62.5%) contained copper.*

Conclusions: *Cervical perforation by the strings of the IUD is an extremely rare clinical entity. It is generally asymptomatic and, in most cases, the strings of the IUD may be returned back to the endocervical canal after surgical maneuvers.*

Keywords: intrauterine device, cervical perforation, hysteroscopy, copper, strings.

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INTRODUCTION

The intrauterine device (IUD) is a widely used method of contraception. It is reliable when inserted by professional health providers (1). It is widely accepted that the use of IUD is increasingly recommended and growing worldwide (2). Since their introduction, intrauterine devices (IUDs) have been related with severe but infrequent complications such as uterine perforations, pelvic infections and migration into the peritoneal cavity or adjacent organs (3). Cervical perforations have been mainly caused by the vertical arm of the IUD, which can be seen protruding through the cervical mucosa into one of the vaginal fornices (3, 4). Strings are essential components of the IUDs which ensure verification of their presence in the uterus by inspecting or palpating the external cervical os. Strings assist in the safe removal of the devices in office environment. The most commonly encountered problems related to strings are the missing or lost threads. The most frequent reasons for strings to be missing include their retraction into the cervix or uterus, translocation of the device by perforation and expulsion of the device (5).

Cervical perforation by the strings of an IUD is a very rare complication, with only scarce reports of cases caused by Copper and Lippes loop devices being found in the medical literature since the end of the 1970s and early 1980s (6, 7).

The aim of the current study is to present a novel case of cervical perforation by the strings of a copper IUD and to perform a systematic review of the available literature regarding this rare clinical condition. ❑

CASE REPORT

The patient, a 43-year-old gravida 3 para 3, attended the family planning clinic in order to remove her copper intrauterine device (IUD)-Monalisa NTCU 380 Normal manufactured by Mona Lisa NV Belgium 2019 that had been inserted 16 months ago, after having had a first IUD inserted. The patient had a history of three previous Cesarean sections and used a copper IUD for four years. She complained from headaches and galactorrhea, blood analysis exhibited high prolactin levels. Thus, the endocrinologist recommended a MRI (magnetic resonance imaging) of the hypophysis in order to exclude a possible prolactinoma. The patient expressed the desire to have her coil removed in order to undergo MRI, despite the fact that there was no complication associated with undergoing MRI and having an IUD at the same time. She attended our gynecology clinic 14 days prior to examination and underwent an annual check, including colposcopy-due to LGSIL (low grade squamous intraepithelial lesion) detected in pap smear, cervical swabs and transvaginal scan. A colposcopy was accomplished and a cervical biopsy was obtained (it revealed LGSIL in pathology analysis); transvaginal scan showed an IUD *in situ* with the strings protruding normally through the cervical os (Figure 1). Two weeks later, at the time of presentation for IUD removal, the patient reported no symptoms. On speculum examination, the observation of the cervix revealed that both strings of the IUD had perforated the anterior lip of the cervix at 2 o'clock, 15 mm left from the cervical os (Figure 2). Transvaginal ultrasound revealed an IUD located properly in the uterine cavity. The patient reported that she did not experience sexual contacts neither had heavy physical duties during the past two weeks. Management options were offered to her and she opted for a hysteroscopic removal under general anesthesia. The patient was placed in a lithotomy position and local disinfection of the vagina with povidone iodine was accomplished. Diagnostic hysteroscopy with a 3 mm rigid hysteroscope and 30o oblique view (Karl Storz, Tuttlingen, Germany) revealed the presence of an IUD located correctly within the uterine cavity, while the direction of the strings was noticed to deviate left towards the cervical wall. A dilation of the cervix was performed and removal of the IUD with the use of a Foerster sponge forceps was achieved. The IUD was intact and further cavity observation revealed no signs of uterine perforation; a small shallow gap on the anterior wall due to IUD placement was observed. The IUD was sent for culture analysis, which was negative. Final transvaginal ultrasound examination exhibited a normal uterus. The woman recovered successfully with no further complains. A written and signed informed consent for reporting this case was obtained from the patient. The case report was written in accordance with CARE (CAse REport) guidelines for writing a case report https://www.care-statement.org/ (8). ❑

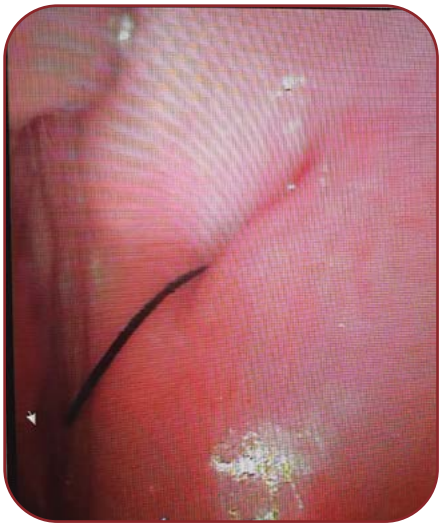

FIGURE 1. Normal positioning of the IUD strings through cervical os two weeks prior to identification of strings through the cervical tissue

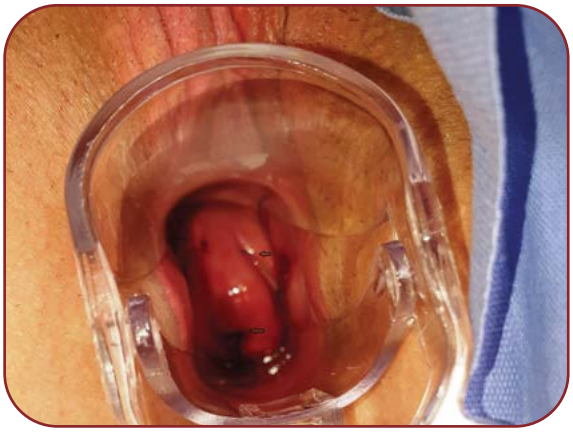

FIGURE 2. Perforation of cervix by the IUD strings cervix at 2 o'clock, 15 mm left from the cervical os

MATERIAL AND METHODS

Search strategy

We searched several electronic databases, including MEDLINE (1950–2022), Cochrane Library (2004–2022), Google Scholar (2000–2022), EBSCO (1984–2022), and no year or language restrictions were imposed. The search included the following medical subject headings (MeSH) or keywords: 'Cervical perforation' AND 'IUD'AND 'Intrauterine device' AND 'Strings'AND/OR 'Threads'. The last search was updated on 22/02/2022. The flowchart diagram of the present systematic review was drawn according to the Preferred Reporting Items for Systematic Reviews and Metaanalyses statement (PRISMA-2020) http://prisma statement.org/prismastatement/flowdiagram.aspx (9). The references of selected articles were scrutinized for studies not revealed by the initial electronic search.

Eligibility criteria

Only case reports that described perforation of the cervix by the strings or threads of IUDs were included in the present review. Studies reporting perforation of the cervix or uterus and any other pelvic organ by IUDs, and articles not written in English as well as those that appeared as grey literature were all excluded from our review.

Data extraction

Two authors (PP and CE) have independently extracted the clinical data, while DS and SZ checked and tabulated the obtained information. PT checked the results and approved the study. From each eligible study, we extracted and tabulated on an Excel spreadsheet (v16.0-2016) the following clinical items of the patients included in the analysis: author and year, country and city, setting of the study, patient's age, gravidity, patient symptoms, type of inserted IUD, the time since IUD insertion, location of the area of cervical perforation, performance of ultrasound, interventions that were used and outcomes of the case reports. We performed a quality assessment of case reports according to CARE guidelines, as previously mentioned (8). The purpose of the CARE guidelines is to support an increased accuracy, transparency and usefulness of case reports as well as a decreased risk of bias. The CARE checklist comprises 13 primary items: title, key words, abstract, introduction, patient information, clinical findings, timeline, diagnostic assessment, therapeutic intervention, follow up and outcomes, discussion, patient perspective and informed consent. A series of histograms were drawn to exhibit the quality assessment of the 13 items that should be included when writing a case report. The quality assessment of each item was evaluated with a percentage scale from 0 to 100%.

Statistical analysis

We conducted a calculation of frequencies of the extracted clinical items, mean numbers, standard deviations (SD) and ranges. The analysis was performed with the statistical package

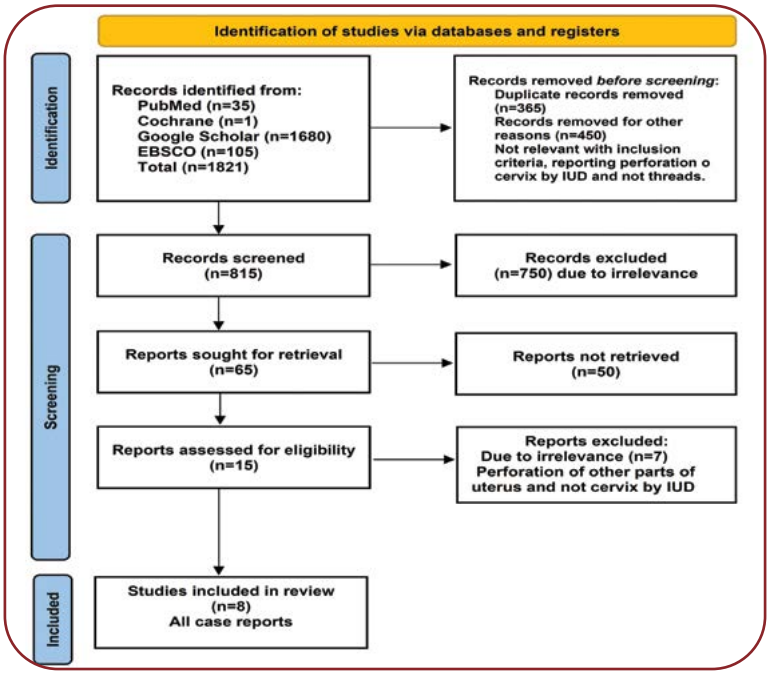

FIGURE 3. Flowchart diagram of study selection in the systematic review according to PRISMA 2020 statement: an updated guideline for reporting systematic reviews

SPSS 23 (Statistical Package for the Social Sciences-Chicago 2015). ❑

RESULTS

The initial electronic search yielded 1 821 relevant articles which were extracted from PubMed (n=35), Cochrane (n=1), Google Scholar (n=1680), EBSCO (n=105). The flowchart diagram of study selection is illustrated in Figure 3. Further assessment and removal of duplicate results led to the retrieval of 815 studies. The extracted studies were scrutinized for relevance and further assessment led to exclusion of 750 studies due to irrelevance with inclusion criteria, language restrictions and grey literature. Finally, eight articles were selected for inclusion in the review, all case reports. In total, eight patients were enrolled in the review. Articles dated from April of 1977 to October of 2018 and all of them were written in English (6, 7, 10-15). ❑

TABLE 1. Summary of clinical data from the case reports included in the systematic review

Author year	Country City	Setting	Age	Gravidity	Symptoms	Type of IUD	Time since insertion	Area of perforation	Ultrasound IUD position	Interventions	Outcomes
Sparks [6]	UK	University	32	multigravida	Asymptomatic	Lippes loop	72 mts	Anterior 7.5 mm	No	Removal	Uneventful
1978	Southampton	Hospital			routine check	Copper		from cervical os			removal
Silverman [7]	USA	University	33	G1	Asymptomatic	Copper	1 mts	Anterior 6 mm	No	Removal	Uneventful
1980	Los Angeles	Hospital			routine check			from cervical os			removal
Savardekar [10]	India	University	26	G3	Asymptomatic	Copper	15 mts	Posterior 20 mm	Yes	Removal	Uneventful
2005	Mumbai	Hospital			routine check			from cervical os	in situ		removal
								at 5 o'clock			
Oswal[11]	UK	University	35	G3	Asymptomatic	Gynefix	12 mts	Anterior left	Yes	Spontaneous	IUD
2008	Luton	Hospital			routine check	Copper		from cervical os	in situ	return of strings to	remained
										cervical os	
Gbolade[12]	UK	University	44	G2	Asymptomatic	LNG-IUS	48 mts	Anterior 10 mm	No	Removal	IUD removed
2010	Leeds	Hospital			routine check			from cervical os		with forceps	and replaced
Gonenc [13]	Turkey	University	47	multigravida	Asymptomatic	LNG-IUS	36 mts	Anterior 5 mm	Yes	Strings removed	IUD
2013	Istanbul	Hospital			routine check			from cervical os	in situ	to cervical os with	remained
								at 3 o'clock		Novak instrument	
Norman[14]	Canada	University	26	nulligravida	Symptomatic	LNG-IUS	12 mts	Anterior 10 mm	Yes	Removal	Uneventful
2014	Vancouver	Hospital						from cervical os	in situ	with ring forceps	removal
Boog [15]	UK	District	40	multigravida	Asymptomatic	Copper T	12 mts	Anterior 10 mm	Yes	Removal	Uneventful
2018	Leicester	Hospital			routine check			from cervical os	in situ	with Spencer-Wells	removal
								at 1 o'clock		forceps under GA	

IUD=intrauterine device, LNG-IUS=levonorgestrel-intrauterine system, mts=months, GA=general anesthesia

CASE REPORTS

The clinical data from the case reports included in the present systematic review are summarised in Table 1. Four of the case reports were from the UK (6, 11, 12, 15), one from the USA (7), one from India (10), one from Turkey (13) and one from Canada (14). All case reports were conducted in university hospitals (6, 7, 10-14), excepting one which was carried out in a district hospital (15). The mean age of all women enrolled in the review was 35.37±7.781 SD (range 26-47) years. All subjects were premenopausal; one woman (12.5%) was nulligravida (14); three women were reported as multigravida (37.5%) (6, 13, 15). Seven women (87.5%) were asymptomatic and scheduled for routine check (6, 7, 10-13, 15); one woman (12.5%) attended the clinic for IUD removal due to hormone-related acne possibly elicited by LNG-IUS (levonorgestrel-intrauterine system) (14). Of all IUDs, three (37.5%) were LNG-IUS (levonorgestrel-intrauterine system) (12-14), five (62.5%) contained copper (6, 7, 10, 11, 15), one IUD (12.5%) was Lippes loop (6) and another one (12.5%) Gynefix (11).

The mean number of months since IUD insertion in the eight selected cases (100%) was 26.00±24.00 SD (range 1-72). The area of perforation was located in the anterior fornix in seven (87.5%) cases (6, 7, 11-14) and in the posterior fornix in one case (12.5%) (10). The distance between cervical os and the area of string perforation was reported in seven case reports; the mean distance was 9.78±4.94 mm SD (range 5-20 mm) (6, 7, 10, 12-15).

In five cases (62.5%), transvaginal ultrasound (TVS) showed that the IUD was *in situ* (within the uterine cavity) (10, 11, 13-15). In six (75%) cases, the IUD was removed after cervical perforation

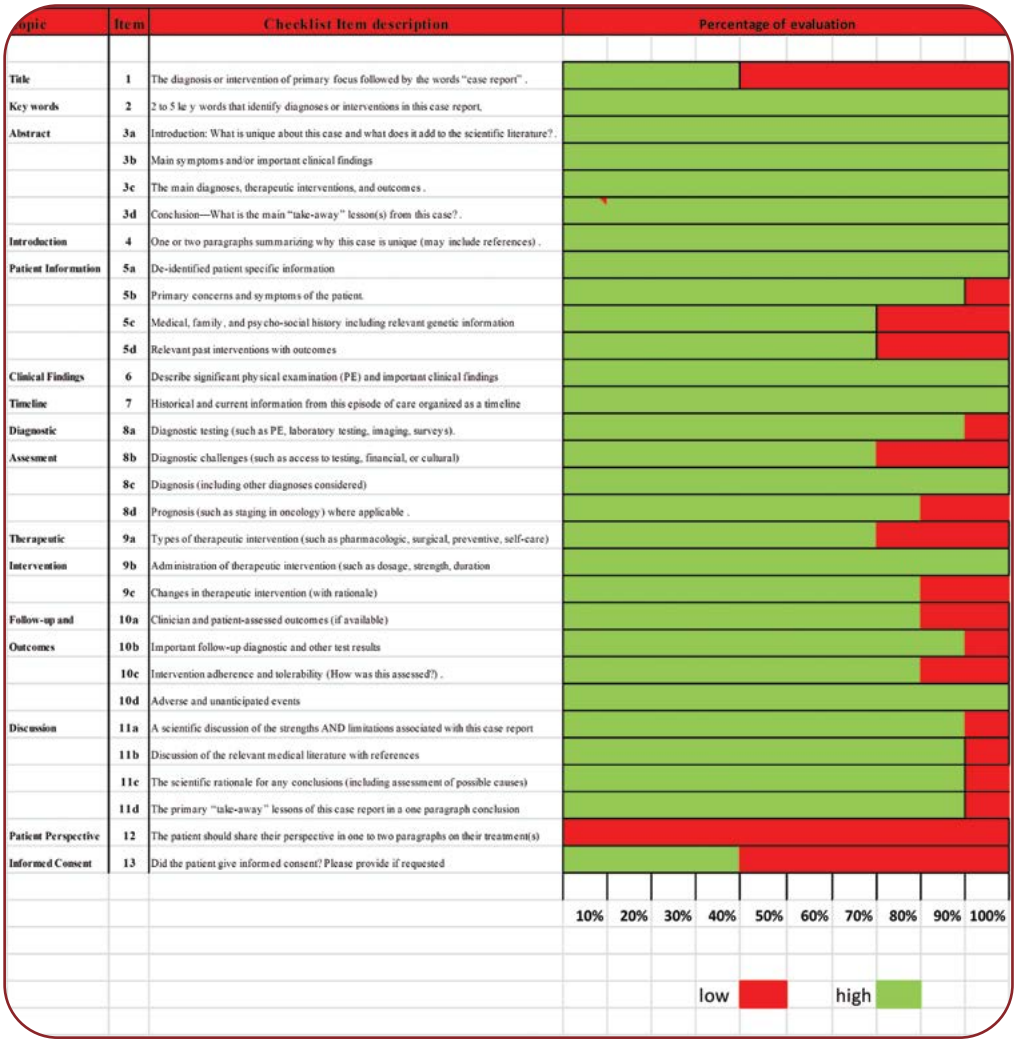

FIGURE 4. Poor quality was detected in item 12 (Patient perspective) with a percentage of 100%, followed by items 1 and 13 (60%). Moreover, a low quality (30%) was detected in the items 5c, 5d (Patient information), 9a (Therapeutic intervention). In items 8d (Diagnostic assessment), 9c (Therapeutic intervention), and 10a, 10c (Follow up and outcomes), a low quality of only 20% was found. A high quality (100%) was noted in the following items: Key words, Abstract, Introduction, Patient information, Clinical findings, Timeline, and Diagnostic assessment

(6, 7, 10, 12, 14, 15) and in three (37.5%) cases, the IUD remained *in situ* with the strings returned back at the cervical os (11-13). In one case (12.5%), the IUD was removed and replaced with a new one (12). General anesthesia and cervical dilation were performed in one case (12.5%) (15).

Quality of case reports

The quality of case reports was performed according to the criteria of CARE guidelines (8). CARE criteria consist of 13 items, as shown in Figure 4.

Strengths and limitations

The current study is the first systematic review of this condition. We endeavored to present this study in a detailed manner. However, we have to support the fact that every study may exhibit strengths and limitations. The strength is the systematic review which offers descriptive clinical information about this rare condition from different studies. The limitation is the small number of studies included in the present review, which is limited to eight, and of course their quality, all of them being case reports. Another limitation is the lack of study homogeneity, with different types of IUDS being reported in each case – this may lead to bias regarding to which IUD may lead to incidental cervical perforation by its strings. Another limitation is the lack of clinical information in the section of patient perspective on their treatment, as shown after CARE assessment of case reports. ❑

DISCUSSION

This is the ninth reported case of cervical perforation by the strings of IUD in the international literature and the first to be removed with the use of hysteroscopy. Hysteroscopy was used for diagnostic purposes. Our systematic review shows that the phenomenon of cervical perforation by the strings of IUD is an asymptomatic clinical entity without local irritating effects.

Cervical perforation was reported more often in Copper IUDs (62.5%) (6, 7, 11-14) *vs* LNG-IUS (37.5%) (12-14). Perforation of the anterior cervical fornix is the most common place where the strings have been located (6, 7, 11-14). In our case, it was incidentally diagnosed two weeks after the last clinical examination. Cervical perforation may be diagnosed 26 months after insertion as it was found in our analysis. The incidence of cervical perforation from IUDs ranges from 0.2 to 1.3% (16).

In a study conducted by Barnett et al (17) on a cohort of 61,448 women who were followed for 12 months, the overall perforation rate was 2.1 per 1 000 insertions (95% CI 1.6–2.8) for LNG-IUS users (40 + 19 perforations/27,630 insertions) and 1.6 per 1 000 insertions (95% CI 0.9–2.5) for copper-IUD users (14 + 4 perforations/11,379 insertions).

We tried to find answers to the following questions: what is the pathophysiological mechanism that leads to this phenomenon, how are the strings transferred from the cervical os and how do they perforate the cervical tissue?

According to a hypothesis reported by Gbolade (13), it is unlikely that the iron oxide and polyethylene composition of the strings of levonorgestrel-releasing intrauterine systems makes them sufficiently rigid to perforate the cervical lip. Thus, the two most plausible explanations for this occurrence are the development of a fistulous tract caused by the single-toothed tenaculum used at the time of insertion, providing a canal for the string(s), or a cervical laceration (13). The author suggests that less traumatical forceps such as such as Teales curved vulsellum (3:4 teeth) should be used for the insertion of IUDs; however, he mentioned that no trauma was caused in the cervix during the insertion in his case (13); similarly, in our case we did not record any trauma at the time of insertion.

Another scenario is the following: the mechanism whereby the threads of an intrauterine contraceptive appear to have perforated the cervix may result from their penetrating the epithelium (and the stroma) like setons (which have been used for more than 2 000 years to treat fistulae) and then working their way through the tissue. When a thread or a thin tube is passed through a fistulous tract and then tied to form a loop which is tightened at intervals, it will progressively cut through tissue while, simultaneously, the divided tissue behind it heals, resulting in the release of the seton (13).

What is the management of this condition? In general, it is advisable to return the strings back to the endocervical canal via the use of several instruments. Clearly, ultrasound will define the correct positioning of IUDs in the uterine cavity,

so every intervention should be done under sonographic guidance.

Our review showed that in five cases (62.5%) the IUD was correctly positioned. When there is no feasible return of the strings into the canal and there is a suspicion of penetration by the IUD, removal should be utilized via office or conventional hysteroscopy.

Our review has limitations due to the small number of included case reports. However, it is the first systematic review of the literature addressing this particular issue. ❑

CONCLUSION

Cervical perforation by the strings of an IUD is an extremely rare clinical entity. In general, it is asymptomatic and affects the anterior lip of the cervix. It mainly occurs in multigravida women and may be diagnosed after two years since the initial insertion. It may cause frustration to both the clinical professional and patient. The pathogenesis of this phenomenon is not completely explained and more studies are further needed. In most cases, the strings may be successfully returned back to the endocervical canal. ❑

Conflicts of interest: none declared.

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Institutional Review Board Statement: This study was approved by the Institutional Review Board of Rea Hospital.

Informed consent statement: Written informed consent has been obtained from the patient(s) to publish this paper if applicable.

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Author contributions: P. P was responsible for conceptualization and original draft preparation, C. E. for the project administration, P. P, C. E. and D. S. were involved in writing—review and editing, and S. Z. and P. T. in supervision. All authors have read and agreed the published version of the manuscript.

References

1. **Glasier A.** Contraception. In: Edmonds KD, ed. *Dewhurst's Text-book of Obstetrics and Gynaecology*, 7th edition. Maldon, MA: Blackwell Publishing, 2007, pp 299-317.
2. **Dunn S, Anderson GM, Bierman AS, POWER Study.** Temporal and regional trends in IUD insertion: a population-based study in Ontario, Canada. *Contraception* 2009;80:469-473.
3. **Paulson JD, Kao MS.** Cervical perforation by the copper-7 intrauterine device. *Obstet Gynecol* 1977;50:621-622.
4. **Koltan SO, Tamay A G, Yildirim Y.** Chronic cervical perforation by an intrauterine device. *J Chin Med Assoc* 2010;73:325-326.
5. **Johnson BA.** Insertion and removal of intrauterine devices. *Am Fam Physician* 2005;71:95-102.
6. **Sparks RA.** Problems with IUCD tails. *Br Med J* 1978;1:1141.
7. **Silverman I, Rubinstein LM.** Perforation of the exocervix by an IUD tail. *Contracept Deliv Syst* 1980;1:369-370.
8. **Riley DS, Barber MS, Kienle GS, et al.** CARE guidelines for case reports: explanation and elaboration document. *J Clin Epidemiol* 2017;89:218-235.
9. **Page MJ, McKenzie JE, Bossuyt PM, et al.** The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. *BMJ* 2021;372:n71.
10. **Savardekar LS, Shah RS, Vandana W.** Asymptomatic cervical perforation by intrauterine copper bearing contraceptive device: two unusual cases. *Eur J Obstet Gynecol Reprod Biol* 2005;120:236-237.
11. **Oswal A, Loizides S, Robinson C.** Gynefix tales: cervical perforation and repeated late expulsion with the Gynefix device. *Eur J Contracept Reprod Health Care* 2008;13:215-217.
12. **Gbolade BA.** Perforation of the cervix by the threads of a levonorgestrel-releasing intrauterine system: a case report. *J Womens Health* (Larchmt) 2010;19:2277-2279.
13. **Gönenç I, Vural EZ, Aka N.** Cervical perforation by the strings of a levonorgestrel releasing-intrauterine system: a case report. *Eur J Contracept Reprod Health Care* 2013;18:415-418.
14. **Norman WV, Stothart D.** IUD String Perforation Through Anterior Cervix: A Case Report. *J Obstet Gynaecol Can* 2015;37:345-348. doi: 10.1016/S1701-2163(15)30285-1.
15. **Boog K, Kumar V.** Perforation of the anterior cervix by the threads of an intrauterine device. *BMJ Sex Reprod Health* 2018;44:314-315.
16. **Prema K, Malhotra U, Choudhary SD, et al.** Uterine perforations with Copper T IUDs. *Contracept Deliv Syst* 1981;2:327-329.
17. **Barnett C, Moehner S, Do Minh T, Heinemann K.** Perforation risk and intra-uterine devices: results of the EURAS-IUD 5-year extension study. *Eur J Contracept Reprod Health Care* 2017;22:424-428.