INTRODUCTION
Intra uterine contraceptive devices (IUD) have been in use since the 19th century [1]. In 1960, Jack Lippes made the first model of his ‘Double-S’ Intra uterine Contraceptive Device - Lippes Loop (LL), a flexible polyethylene plastic loop of appropriate size for the uterine cavity which subsequently went on to become the standard for other IUD’s to be compared with [1,2]. Dr Jack Lippes analyzed 40,000 women from 1962 to 1968 & reported in Contraception Technological Update that he found no trouble with the Loop. However, Lippes Loop is no longer in use after Ortho Pharmaceutical Corporation stopped marketing LL-IUD citing economic reasons [3].

Due to the implant nature of these early IUD’s like Lippes Loop, they were intended for long term use until menopause. They are hence often retained for years and many patient present well into menopause still bearing a Lippes Loop either deliberately or forgotten [2]. Time and time again literature has documented side effects & complications following long term use of IUD’s.

We report here a case of removal of a Lippes Loop inserted 45 years ago, in a lady without her knowledge and who was totally unaware of this insertion until informed about it by us when she reported to our hospital with the complaint of post menopausal bleeding. The inserted LL thread was incidentally discovered during clinical examination and Ultrasonography. We also reviewed literature to determine the evidence for and against removal of an inert IUD.

CASE REPORT
A 65-year-old married lady, Gravida 2, Para 2, postmenopausal for 18 years presented to the Gynaecology Outpatient Department of Kasturba Hospital with complaints of vaginal discharge which was occasionally blood stained, associated pain abdomen for the past three months. Patient never had any complaints prior to achieving menopause and though she never used any contraception, she never conceived but she was not unduly worried about her secondary infertility as she already had two sons.

For the past few weeks her vaginal discharge had become pus like and was blood mixed with foul smell. She consulted a local Gynaecologist who gave her some local treatment and advised Ultrasonography of pelvis [Table/Fig-1] which however reported nothing remarkable other than postmenopausal atrophic status of uterus & ovaries. The lady not relieved of her symptoms sought a second opinion at Kasturba Hospital. On Per Speculum Examination cervix was seen to be unhealthy and atrophic but what caught the attention of the Consultant was a fine blue coloured Nylon Thread protruding the external os, appearing like an Intra Uterine Device tail. There was confusion in diagnosis as neither the lady could not recall any IUD insertion in her reproductive years nor did the USG done recently show any evidence of a situ device. A PAP smear was taken and it was decided to repeat the sonography keeping in mind the possibility of an IUD. On careful observation the Ultrasonologist diagnosed an intrauterine Lippes Loop [Table/Fig-2].

A detailed history and several leading questions revealed that 44 years back, some two years after her second childbirth she had developed pain abdomen for which she had consulted a Gynaecologist in a Government Hospital. While she was waiting for the doctor she was beckoned inside a room on the pretext of some preliminary examination and examined vaginally and probably inserted an IUD but was not told about it. Thereafter, she was sent to the Consultant for further check up. She did have complaints of discharge and pain abdomen occasionally for which she had repeated gynaecological consultations but somehow her thread never got detected.

After all routine investigations, the patient was posted for removal of IUD under anaesthesia in Operation Theatre. A few attempts at removal by pulling at the thread were unsuccessful and finally the thread broke [Table/Fig-3]. While preparations for Hysteroscopy were being done, an attempt at removal was made by introducing a curved artery forceps blindly into the uterine cavity and fortunately a Lippes Loop was successfully pulled out [Table/Fig-4-6], Endometrial Curetting were taken and sent for Histopathology. No malignancy was detected either in PAP Smear nor in Endometrial Curettage.
Following a detailed search and review of literature the reported complications of an IUD-Lippes Loop retained in uterus for several years often beyond menopause, are discussed at length below. A detailed study of LL over 10 years & 27954 women showed highest pregnancy rate (annual average 0.5) and highest removal rate for bleeding & pain in the first two years with maximum expulsion within the first three years of application [5]. Although pregnancy rates declined with length of use the rate of ectopic pregnancy remain constant with a 1 in 10 chance if the device has been in situ for more than three years [6]. Infections though common within the first year of inserting the device could however occur anytime [7]. Controversy continues whether this is due to migration of organism from vagina, & cervical canal to endometrial cavity along the thread of IUD [8,9]. Attention has also been drawn to Actinomycosis as a rare cause of PID associated with IUD as a few such cases have been reported in literature from time to time [10].

Little has been written about the difficulties encountered in removal of long term IUD as they tend to accumulate small deposits of calcium causing corrosion in the plastic to thus compromise the strength of the device and tail rendering it liable to fracture & breakage. With passage of time the loop tends to bury in the endometrium resulting in difficult removal with accompanying pain & bleeding. Removal may become more difficult after menopause because of atrophy of uterus & cervical canal [11].

Several interesting studies & rare case reports of complications following insertion of Lippes Loop discussed subsequently, which tilts the balance heavily in favour of removal of a Lippes Loop- Intra Uterine Device irrespective of it causing no difficulty or symptoms to the patient and it goes without saying that it is mandatorily removed in the event of any discomfort or symptom to the patient.

**Outcome of pregnancies with Lippes Loop in situ** was studied by Suporn K et al., In a total of 210 cases, 4 tubal pregnancies, 1 ovarian pregnancy, 2 extra uterine IUDs and 7 cases of proven expulsion were observed prior to pregnancy. The outcome of remaining 196 pregnancies with Lippes loop in situ observed a high incidence of spontaneous abortion and preterm delivery (56.8%) in the group of 102 patients whose LL was left in situ due to inaccessible thread as compared to the remaining 94 whose loop was removed when thread was still visible. The occurrence of pregnancy following IUD insertion is a distressing problem to both physician and patient, especially in countries where induced abortion is illegal [12].

**Perforation and Asymptomatic Migration of a LL-IUD**

Spontaneous perforation is a rare but well known complication of IUD insertion where the IUD initially embeds in the uterine wall followed by complete perforation to thus migrate in the peritoneal cavity. Inert IUD like Lippes Loop tend to remain asymptomatic for years & are often discovered incidentally during the investigative work up for some other disease [1,13].

**Migrated Loop incidentally discovered in a patient of endometrial carcinoma**

Chanin et al., report an incidental finding of a wandering radio opaque structure in the abdominal cavity bringing forth the need for removal of LL and its concurrent pathology [14].

**Search for Migrated Loop reveals concurrent ceacal carcinoma**

Bharathi et al., report an extremely rare case of migration of LL consequent to fimbrial extrusion and its detection after four decades in due course of investigations for GI symptoms in a 75-year-old lady. Exploratory laparotomy following initial impression of Sub acute intestinal obstruction due to LL led to detection of co existent caecal malignancy as a concurrent pathology [15].

**Perforation by LL 39 years after insertion resulting in impressive calculus formation and creating a vesico-vaginal fistula causing urinary incontinence and recurrent urinary tract infections, contributing to terminal kidney failure** is an unusual complication.

**REVIEW OF LITERATURE**

Lippes Loop manufactured by Ortho Pharmaceutical Corporation came in pre packaged Polyethylene/Paper pouch in four available sizes duly identified by different coloured tails namely Loop A (blue tail), Loop B (black tail), Loop C (yellow tail), Loop D (white tail). They were inert IUD intended for long term use till menopause, but this form is no longer in use having been replaced by the now popular copper bearing devices introduced in the1970s [4].

In August 1985, US Food & Drug Administration (FDA) published a proposed rule making it mandatory for any company wanting to market Lippes Loop or any non drug IUD to apply for a premarketing approval by submission of a detailed discussion and supporting clinical studies addressing the concerns of pelvic Actinomycosis, tubal infertility, duration of leaving IUD in situ, safety of leaving IUD in situ when contraception was no longer required. Ortho’s sale of LL dropped considerably as most physicians according to Ortho were prescribing copper-bearing IUD’s and the company discontinued sale of Lippes Loop in the year citing economic reasons [3].
reported by Karmmasters et al., where a 74-year-old presented with dribbling of urine, dysurea and a palpable mass in bladder. X-Ray revealed a large vesical calculus with Lippes Loop –IUD. On further Cystoscopic evaluation small inflamed bladder with calculus & fistula connecting the bladder with uterus was observed. Stone removal with fistula closure was done [16].

Perforation of the rectum by a Lippes Loop is another unusual complication reported by Abdalla O where a 26-year-old patient illustrated an unusual form of bowel complication encountered with a Lippes Loop. The young woman had the IUD fitted three years previously in the family planning clinic. Unfortunately, she became pregnant 1 year after insertion. As the device was not found at delivery, an ultrasound scan was performed which revealed that the coil was extraterine but intra-pelvic. It was left in situ as the woman was asymptomatic and it was causing no problems. The patient presented to the hospital with the thread protruding from the anus. Pulling on the thread caused severe pain as the tip of the IUD was deeply embedded in the rectal wall [17].

Uterine perforation with Lippes loop intrauterine device & associated Actinomycosis has been described by Phupong V et al.. A 67-year-old postmenopausal woman, with an uterine perforation from actinomycotic infection with Lippes loop IUD is reported. She had the Lippes loop IUD inserted for 35 years, and never had any pelvic examination or Papanicolaou smear. She presented with acute abdominal pain. The clinical picture mimicked peptic ulcer perforation. The woman underwent laparotomy and exudative fluid was discovered in the abdominal cavity with the tip of the Lippes loop IUD at one of the two small holes of the uterine fundus. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was performed. The postoperative microscopic pathological report demonstrated characteristics of actinomycosis [18].

Forgotten Lippes Loop and Pelvic Inflammatory disease has been reported by Pukale R et al.., who published a case report of a 50-year-old woman suffering from abdominal pain, fever & chills associated with foul smelling vaginal discharge and right sided adnexal mass suggestive of Pelvic Inflammatory Disease. Investigations/X-Ray revealed a Lippes Loop in uterine cavity inserted 30 years ago & forgotten by the lady. Cut section of uterus following Hysterectomy showed a deeply embedded loop [19].

Forgotten Lippes Loop and Post Menopausal Bleeding has been reported by Sujatha VV of two cases with retained intrauterine contraceptive devices (IUCDs). The first presented with perimenopausal bleeding and dysmenorrhoea and the symptoms were resolved following the removal of an IUCD. The second presented with postmenopausal bleeding and had a Lippes loop in the uterine cavity. The symptoms resolved after the removal of the device. There are few more case reports of PMB with forgotten LL [20].

Forgotten Lippes Loop with Secondary infertility and dysfunctional uterine bleeding: A case report has been presented by Agarwal N et al., where a 56-year-old postmenopausal woman suffered from secondary infertility and dysfunctional uterine bleeding due to a Lippes loop that was detected after 32 years of insertion. The patient presented with symptoms of lower abdominal pain and dysuria. Ultrasound/X-Ray revealed a foreign body in the pelvis leading to the suspicion of an intrauterine contraceptive device (IUCD) [21].

Breakage of Lippes loop in situ: Eight cases are reported by R Solal et al., in which breakage of a Lippes loop occurred in situ. The incidents were found to be caused by defective material. While this fragmentation did not cause any major complication, it was followed by expulsion and, in some cases, bleeding and pain. Fragmented Lippes loops should not be left in situ but removed [22].

Difficulty in removal of LL-IUD retained for long: Dwyer NA et al., describe the case of a 38-year-old women with lost LL-IUD, inserted 7 years before but confirmed to be in utero ultrasonographically, where all outpatient attempts to remove it were unsuccessful and patient had to be referred to specialist for removal. On Hysteroscopy it was found to be partially embedded at the fundus and an operating Room was used to finally remove the loop along with a strip of myometrium [23].

Stage Ia Endometrial carcinoma around Lippes Loop: Tracey et al., report an unusual case of a 49-year-old post menopausal woman presenting with offensive vaginal discharge 3 years later. On clinical examination IUD thread was noted at external os & apparently Lippes Loop inserted 20 years previously was forgotten by the patient. Removal was easy but suspicious abnormal tissue adherent to the loop turned out to be moderately/poorly differentiated Grade II/III – Endometrial Adenocarcinoma. Subsequent Total Hysterectomy & Bi-lateral Salpingo-oophorectomy showed no residual carcinoma of the endometrium [24].

Adenocarcinoma of uterus associated with Lippes Loop: Robert F.Harrison has presented the case report of a young lady with history of painful, heavy vaginal bleeding following 14 months of amenorrhea post insertion of Lippes Loop 20 months earlier. Curettage taken at the time of removal of loop turned out to be Adenocarcinoma of endometrium and patient was subjected to vaginal hysterectomy with Bi-lateral Salpingo-oophorectomy [1].

Pyometra & squamous cell carcinoma cervix with Lippes Loop in situ for 25 years: A post menopausal women presenting with pyometra & carcinoma cervix with a neglected Lippes Loop inserted 25 yrs back which had never been followed up with regular cervical smears and check up. Patient presented with atypical upper respiratory tract symptoms followed by brown vaginal discharge. CT imaging revealed a huge pyometra with a Lippes Loop in situ. At the time of pyometra drainage cervix was noted to be suspicious and biopsy confirmed Stage Ib poorly differentiated Squamous Cell Carcinoma of the cervix. Interestingly literature suggests a decreased incidence of Carcinoma cervix on the rationale frequent follow up advised after IUD insertions [25].

Primary carcinoma of fallopian tube after prolonged retention of Lippes Loop: Sanyal C et al., in this article present a case of primary carcinoma of the fallopian tubes with a co-existing Lippes loop inserted 20 years back, in a 50-year-old patient with complaints of vaginal bleeding 5 years after menopause. Histopathology following hysterectomy and bilateral salpingo-oophorectomy, showed features of hydrosalpinx follicularis along with papillary adenoacarcinoma in distal parts of both fallopian tubes. Although a small number of uterine malignancies have been reported in long-term IUD users, a cause-effect relationship maybe difficult to establish however the presence of atrophic endometritis suggests the possibility of carcinoma being preceded by salpingitis, initiated by prolonged IUD retention [26].

DISCUSSION

The removal of a dislocated IUD should be a foregone conclusion but surprisingly remains a point of controversy amongst several clinicians who are of the view that surgery for removal of a silent migrated Intra Uterine Device may cause further harm by causing increased adhesion formation [14]. Removal of a non migrated in situ IUD like LL also is not free from controversy where opinion is divided whether it merits removal or should it be left in situ & removed one to two years after menopause. However, consensus is clear on elective retrieval of asymptomatic migrated IUDs when incidentally discovered during abdominal exploration [15].

A forgotten IUD can lead to a range of problems starting form infertility to postmenopausal bleeding, pain abdomen due to PID, fever and malaise due to actinomycosis [19]. From time to time literature mentions side effects (Goldzieher, 1968) & complications
(Tietz, 1965) from using various types of Intra Uterine Devices. These complications include ectopic pregnancy (Tietz 1966; Weil1968) bowel obstruction following perforation (Hapsels 1969;Thambu 1965), uterine perforation (Liao 1969; Tietz 1966), infections & death (Sctt 1968) [1] which probably justifies the above arguments and recommendations in favour of removal of IUD like LL, once they have outlived their utility & function.

The incidence of difficulties associated with IUD removals is rarely defined, but may occur in up to 9% of follow-up visits of women who have been fitted with an IUD. IUDs which are ‘lost’ within the uterus require operative removal in 40% of cases even in the hands of skilled operators. These ‘lost’ IUDs should be removed as they are less likely to be correctly sited at the fundus than those with a thread visible at the external os, thus increasing the risk of pregnancy [23].

Coexistence of concomitant pathology is rarely encountered but it is very much a probability which cannot be ignored, especially in the event of a mismatch between clinical picture & preliminary investigations. Such cases warrant further investigations and are splendidly picked up in this current era of high resolution & advanced imaging techniques thus helping in deciding management protocols [15].

Although there is no established causal evidence linking IUD’s retained after menopause and cancer or other significant problems such retentions does confuse the diagnosis in instances of post menopausal bleeding and makes difficult such procedures as ultrasonic endometrial evaluation. When contraception is no longer an issue, it is prudent to remove IUD’s since they may cloud further necessary evaluation [26].

A search of literature identified a limited number of case reports where IUD left in situ for extended period. A non systematic review of 40 years of Lippes Loop conclude that despite a lack of evidence linking IUD’s in postmenopausal women with diseases such as endometrial cancer, the presence of these devices may be a hindrance to investigations if the patient presents later especially endometrial biopsy or ultrasound. The study recommends that if an IUD is no longer required for contraception it should be removed [27].

The Faculty of Sexual & Reproductive Health Care (FSRH) guidance on contraception for women aged over 40 years states that after the menopause intrauterine methods of contraception should be removed rather than left in situ as cases of Pyometra & Actinomycosis have been reported in postmenopausal women with Intra Uterine Devices [4,28,29].

WHO recommends removal of a misplaced IUD immediately after diagnosis is made [30]. Every consultation is an opportunity for health education & promotion [25]. In developing countries like India, due to lack of access to healthcare facilities, it is not uncommon to see patients with a forgotten IUD presenting years later with abnormal vaginal bleeding or discharge. An event like this may be the first contact for the person with health facility & one should ensure that such opportunities should not be missed [31].

CONCLUSION

Case reports are a useful source of evidence where evidence where evidence exists. Reporting of rare case particularly rare complications, serious or otherwise associated with prolonged contraceptive use should be widely encouraged.

The review of literature highlights the need to question whether there is justification in retaining an in situ inert Intra Uterine Device like Lippes Loop, which the woman has been happily wearing for years without any difficulty or signs & symptoms, especially if she has been coming for regular clinical & cytological follow up. It is also relevant to consider whether a comfortably worn inert IUD should be removed for further replacement with the current Copper containing IUCD’s which have much higher complication rates and cannot be worn indefinitely without frequent replacements.

After the detailed search of literature, I am of the opinion that we are not justified in leaving an IUD in situ even if inert and asymptomatic, as complications cannot be predicted and can develop at any stage. Complications are often of a severe and unusual nature which can result in high level of morbidity to the patient and many a times may also have life threatening implications. Loss of even a single life allegedly due to a complication which could well have been avoided by timely intervention seems unjustifiable, not to mention the medico- legal implications in this day & age of litigation, where the patient may litigate the responsible doctor and force him to face charges of medical negligence.

REFERENCES


PARTICULARS OF CONTRIBUTORS:
1. CMO-Consultant, Department of Obstetrics and Gynaecology, Kasturba Hospital, Delhi, India.
2. CMO, Department of Obstetrics and Gynaecology, Kasturba Hospital, Delhi, India.
3. Resident Physician, Department of Obstetrics and Gynaecology, Charleston Area Medical Center, West Virginia University, Charleston Division, Charleston, West Virginia.
4. Senior Specialist and Head of Department, Department of Anaesthesia, Kasturba Hospital, Delhi, India.
5. Senior Specialist and Head of Department, Department of Obstetrics and Gynaecology, Kasturba Hospital, Delhi, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Maruti Sinha,
C-2/176, First Floor, Janakpuri, New Delhi -110058, India.
E-mail: maruti_2203@yahoo.com

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