

Post-ablation tubal sterilization syndrome: A systematic literature review

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Abstract: The objective of the study was to summarize the findings regarding Post-ablation Tubal Sterilization Syndrome (PATSS) and to verify whether there are other conditions besides tubal ligation that may delineate a patient profile with higher propensity to developing PATSS. Methods: During the second semester of 2013, two databases (LILACS – Latin American and Caribbean Health Sciences Literature, and MEDLINE®, the U.S. National Library of Medicine® main bibliographic database) were consulted in order to identify studies regarding PATSS, considering also ‘cornual hematometra’ as an alternative search term. Studies developed using original data on pain attributed to PATSS were considered for inclusion in the present review. Publications with certain formats such as letters and commentaries were excluded. Results: ten studies were selected, most of which developed in the United States (8/10). Brazil and China were home to one study each. Study designs were case report (4/10), case series (3/10), retrospective cohort (2/10), and cross-sectional (1/10). The factor reported in all studies was tubal ligation. Uterine leiomyoma, endometriosis, adenomyosis, and endosalpingiosis were also considered as facilitating factors. Conclusion: scientific evidence for PATSS is scarce and not very consistent, suggesting that more comparative studies may be necessary to clarify the causality of this syndrome.

Keywords: Treatment Outcome, Sterilization, Tubal, Endometrial Ablation Techniques, Risk Factors

1. Introduction

Post-Ablation Tubal Sterilization Syndrome (PATSS) corresponds to a set of signs and symptoms manifested belatedly in some women following endometrial ablation. It is characterized clinically by uni- or bilateral cyclical pelvic pain, which may or may not be associated with vaginal bleeding. It was originally described by Townsend et al. [1] following reevaluation, by laparoscopy and hysteroscopy, of six women who developed a clinical condition of sudden abdominal pain resistant to medicinal treatment, which appeared following a rollerball endometrial ablation.

The physiopathology of syndrome is related to endometrial regeneration in the cornual region and to the accumulation of menstrual blood in the uterine cavity [2]. On one hand, the tenuous anatomic structure of the cornual region requires that the ablation procedure be delicately performed, avoiding thus possible accidents, such as perforation, for instance. On the

other hand, the cicatrization process and fibrosis of the lower segment of the uterus following ablation impedes the flow of menstrual blood that emanates from the remaining endometrial glands. The blood ends up being regurgitated toward the Fallopian tube. This may manifest itself as a hematometra within the body of the uterus (central hematometra) or in the cornual region. Pain is due to the presence of elements that include retrograde menstruation, tubal dilation complicated by obstruction due to the sterilization or previous infection, inflammatory process, and formation of hydro- or hematosalpinx [3].

PATSS frequency varies from 6% to 8%, with clinical manifestations appearing two to three years following endometrial ablation [3]. In Brazil, the reported incidence of cases in São Paulo was of 1.7% [4], but in general it is estimated that the syndrome may affect 8% to 33% of patients subjected to endometrial ablation [5]. The lack of serial anatomopathological examination of the cornual area

of the uterus and of the proximal fallopian tubes in all cases which requires major surgery were the justifications presented to explain the low frequency found in the Brazilian setting [4].

In parallel with the widespread acceptability of endometrial ablation procedures by clinicians and patients alike, the emergence of late complications such as PATSS, which in most cases require definitive treatment by major surgery, may induce the erroneous interpretation of method failure. Additionally, it is important to mention that the literature is scarce regarding PATSS, which justifies the development of this review, with the objective of summarizing the data regarding post-ablation tubal sterilization syndrome and to verify whether there are other conditions besides tubal ligation that may delineate a patient profile with higher propensity to developing PATSS.

2. Methods

Throughout the second semester of 2013, a review of the literature was conducted without language or period restrictions using the following online databases: LILACS (Latin American and Caribbean Health Sciences Literature), and MEDLINE®, the U.S. National Library of Medicine® main bibliographic database.

The descriptors/keywords employed were “*postablation tubal sterilization syndrome*” OR “*post-ablation-tubal sterilization syndrome*” OR “*postablation-tubal sterilization syndrome*” OR “*post-ablation tubal sterilization syndrome*” OR “*cornual hematometra*” OR “*cornual hematometrium*”.

Study selection was conducted independently by two evaluators. The reading of abstracts guided the selection of studies to be evaluated in their entirety, and any disparity was resolved by consensus. Independently of the design (case report, case series, or comparative studies), studies developed from original data regarding pain attributed to PATSS were considered for the present review. Publications formatted as letters or commentaries were excluded.

Additionally, reference lists in the selected articles were consulted in order to identify other studies of importance on the theme which could by chance have been missed on the review.

Data extraction considered characteristics of the studies (year of publication, authors, language, country, and design) and of the cases (technique used to perform the procedure, conditions postulated as related to PATSS development).

Compiled data were entered and analyzed using the *MS-EXCEL* program.

This study consisted of literature review conducted by consultation of databases available on the internet. No interviews or direct contact with humans occurred, therefore no violation of any individual's privacy or identity took place.

3. Results

Figure 1 shows the word combinations used on the respective database searches, and the number of studies found. As abstracts were read, it was ascertained that all studies found on LILACS were also present in the MEDLINE database.

Words used	Sources researched	Studies found
“ <i>postablation tubal sterilization syndrome</i> ” OR “ <i>post-ablation-tubal sterilization syndrome</i> ” OR “ <i>postablation-tubal sterilization syndrome</i> ” OR “ <i>postablation tubal sterilization syndrome</i> ” OR “ <i>postablation tubal sterilization syndrome</i> ” OR “ <i>cornual hematometra</i> ”	Medline	16
(<i>postablation tubal sterilization syndrome</i> OR <i>postablation tubal sterilization syndrome</i> OR <i>Post-Ablation-Tubal Sterilization Syndrome</i>)	LILACS	5
Articles found in reference lists	–	1

Figure 1. Words used, sources researched and studies found.

Following the application of inclusion and exclusion criteria and consideration of the bibliographical references of selected articles, ten studies were selected for analysis in the review. Most studies were developed in the United States (8/10). Brazil and China were each the site for one study. Only three studies were comparative, one was a cross-sectional work conducted in Brazil [4], and two were retrospective cohorts conducted in the United States [6, 7]. All studies were published in English, except for the Brazilian study [4]. Studies published in specific sections such as ‘Image of the Month’ [8] and Letters [9] were discarded.

Table 1. Studies and suspected factors as related to post ablation tubal sterilization syndrome (PATSS).

Authors	Year	Country	Design	N/PATSS	Technique	Factors
Townsend et al [1]	1993	USA	Case series	6/6	Rollerball	Tubal Ligation (TL)
Webb et al [12]	1996	USA	Case report	1/1	Rollerball	TL, endometriosis
Bae et al [13]	1996	USA	Case series	305/6	Rollerball	TL
Mall et al [7]	2002	USA	Cohort	174/3	Rollerball	TL
McCausland & McCausland [11]	2002	USA	Case series	50/5	Rollerball	TL,adenomyosis,endosalpingoblastosis
Nichols et al [10]	2009	USA	Case report	1/1	ThermaChoice®	TL, endometriosis
Leung & Yuen [14]	2011	China	Case report	1/1	ThermaChoice®	TL
AlHilli et al [6]	2012	USA	Cohort	63/6	Radiofrequency	TL, leiomyoma
Hellier & Berkhoudt [5]	2012	USA	Case report	1/1	NovaSure®	TL
Takahashi et al [4]	2012	Brazil	Sectional	114/1	Rollerball	TL

Besides tubal ligation performed previously or concurrently with endometrial ablation, other conditions found to have a similar clinical evolution as PATSS were (i) uterine leiomyoma [6], (ii) endometriosis [10-13], (iii) adenomyosis [11, 13], and (iv) endosalpingoblastosis [11]. The studies considered in the present review are shown in Table 1.

4. Discussion

This review confirmed how PATSS evidence is scarce and not very consistent. The studies found were, in their majority, case reports or case series. Besides tubal ligation, the few conditions suspected as being facilitators for the development of PATSS were presence of uterine leiomyoma [6], adenomyosis [11,13], endometriosis [10-13], and endosalpingoblastosis [11].

Although two cohorts [6,7] have been analyzed, PATSS was reported only as an intermediary condition, and the risk for syndrome development was not estimated. While AlHilli et al. [6] focused on uterine findings identified by ultrasound in women subjected to radiofrequency endometrial ablation, Mall et al, [7] investigated whether a previous tubal ligation was a risk factor for hysterectomy following rollerball endometrial ablation. Tubal ligation was found in five of the six Mayo Clinic cases reported as cornual hematometra or endosalpingiosis [6]. Uterine leiomyoma was suspected to be an important contributing factor for PATSS by the Mayo Clinic. In the other cohort analyzed [7], the anatomopathological findings on the women who required additional post-endometrial ablation surgical procedures identified five PATSS cases, all in women with previous tubal ligation [7] and with no other conditions reported.

In Brazil, results of a cross-sectional study [4] that included 114 patients who underwent endometrial ablation using loop resection combined with rollerball electrocoagulation at the *Hospital do Servidor Público de São Paulo* showed 21 cases classified as therapeutic failure, and that required hysterectomy. The authors identified in one of the two cases that evolved with cyclical postablation pelvic pain the presence of hydrohematosalpinx in the anatomopathological exam, characterizing the post-ablation tubal sterilization syndrome. Occurrence of tubal ligation was highlighted as a factor that inversely influenced the success of the ablative procedure, with no report of other conditions. In that study, the low frequency of PATSS was attributed to the absence of detailed serial examinations of the tubal segment in hysterectomy cases.

Besides relating PATSS to tubal ligation [5, 10, 12, 14], case report studies demonstrated, primarily, that the complication may also follow ablations performed with second-generation equipment, such as NovaSure® [5] and ThermoChoice® balloon system [10, 14]. It was also found that, due to the low accuracy for detection of low-volume collections in the cornual region, ultrasonography and computed tomography have not contributed much to the

diagnosis, and a magnetic resonance imaging exam during the cyclical pain symptomatic period was recommended in a few cases [5].

Besides the suspicion regarding the syndrome initially described based on observation of case series [1], this type of design also contributed to evidence that focal adenomyosis and tubal endometriosis [13] were conditions that, along with tubal ligation, could facilitate the development of PATSS. A summary of six cases carefully analyzed by Bae et al [13] demonstrated that the syndrome is characterized anatomopathologically by the presence of hematosalpinx and several microscopic aspects, including endometriosis, acute and chronic inflammation of the tubes, and chronic myometritis. The series evaluated by McCausland & McCausland [11] also emphasized the contribution of the magnetic resonance imaging exam in PATSS diagnosis.

While describing PATSS, Townsend et al. [1] recommended that it be suspected if patients with a history of tubal ligation who were subsequently subjected to endometrial ablation presented condition of uni- or bilateral cyclical pelvic pain associated or not with vaginal bleeding. Although they have attempted to explain these symptoms, questions persisted. One of these questions was whether findings truly constituted a new syndrome, or whether they were simply unusual clinical manifestations associated with tubal sterilization and/or endometrial ablation. In this review, the only study [7] designed to test the hypothesis of causality of tubal ligation in women with endometrial ablation in different outcomes showed a high risk for a new episode of pain (HR 3.2; 95% CI: 1.0-10.6), although with a bordering confidence interval.

The present study was greatly limited by the impossibility of summarizing measures via meta-analysis, due to the fact that the majority of studies were cases series or case reports. On the other hand, it was possible to verify that the frequency of PATSS was less than 10%, with manifestations appearing late and acutely independently of the technology employed, demanding invasive treatments, generally by means of a major surgery such as hysterectomy. According to McCausland and McCausland [11], it is important to consider alternative such as partial ablation, which can prevent intrauterine scarring and contracture, as well as avoid painful complications.

5. Conclusion

In the absence of consistent scientific evidence, this study showed that the main factor related to PATSS was tubal ligation. Uterine leiomyoma, endometriosis, adenomyosis, and endosalpingiosis were also found in some women with PATSS. Even though the risk for developing PATSS seems low, long-term follow up subsequent to endometrial ablation - with monitoring of painful symptoms and alterations in cyclic menstrual bleeding such as amenorrhea - can contribute to determining the actual frequency of PATSS and to clarify its causal relationships.

It is important that the gynecologists alert their patients with tubal sterilization of the possibility of painful complications following ablation, and that alternative techniques such as partial endometrial ablation are offered. In this procedure, only the anterior or posterior endometrial surface is ablated, while the other wall is carefully preserved. That helps avoid the formation of adhesions or synechiae which follow intrauterine contracture and scarring, in an attempt to prevent PATSS from developing.

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