

Salpingitis isthmica nodosa: review and hysterosalpingography diagnosis of 4 cases in a private radiology clinic

Salpingite ístmica nodosa: revisão e diagnóstico histerossalpingográfico de 4 casos de uma clínica radiológica privada

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DOI: 10.5935/2238-3182.20150073

ABSTRACT

Objective: to identify the radiological findings of salpingitis isthmica nodosa (SIN) through hysterosalpingography and evaluate the site of involvement and its impact on infertility. **Methods:** 2,800 protocols were reviewed in a retrospective study between January of 1987 and January of 1997, in patients with a clinical history of female infertility; four of them with a radiological diagnosis of SIN. Ethnicity, age, marital status, current clinical complaints, medical history, location, and side of involvement were analyzed. **Results:** among the four patients with SIN, three were dark-skin, and one was Caucasian, with an average age of 35 years at diagnosis, two single, one married, and one divorcee. Three patients presented complaints of secondary infertility with previous tubal pregnancy history, and another with primary infertility without pregnancy history. In the four patients with SIN, the tubal portion affected was the cornual isthmian, bilateral tubal in three, and unilateral in one. Regarding peritonization, one showed positive bilateral Cotte proof, two with complete bilateral obstruction, and one with positive Cotte in one of the tubes. **Conclusion:** SIN is more common in dark-skin patients, in the mean age of 35 years, with a clinical history of infertility, with initial and recurrent tubal pregnancy, and subsequent tubal obstruction characterized by lesion in the Isthmian cornual portion of the fallopian tube, usually with bilateral involvement.

Key words: Salpingitis; Salpingitis/radiography; Hysterosalpingography; Infertility; Pregnancy, Tubal.

RESUMO

Objetivo: identificar os achados radiológicos da salpingite ístmica nodosa (SIN) a partir da histerossalpingografia e avaliar o local de comprometimento e sua repercussão na infertilidade. **Métodos:** em estudo retrospectivo foram revistos 2.800 protocolos, realizadas entre janeiro de 1987 e janeiro de 1997, em pacientes com história clínica de infertilidade feminina, tendo quatro com diagnóstico radiológico de SIN, sendo analisados etnia, idade, estado civil, queixa clínica atual, antecedentes clínicos, local e o lado do comprometimento. **Resultados:** entre as quatro pacientes com SIN, três eram melanodérmicas e uma caucasiana, com idade média de 35 anos na época do diagnóstico, duas solteiras, uma casada e uma divorciada. As queixas de infertilidade secundária estavam presentes em três pacientes com antecedentes de gravidez tubária prévia e outro de infertilidade primária, sem antecedentes de gravidez. Nas quatro pacientes com SIN, a porção tubária acometida foi a ístmica cornual, tubário bilateral em três e unilateral em uma. Em relação à peritonização, uma apresentava prova de Cotté positiva bilateral, duas com obstrução completa bilateral e uma com Cotté positiva em uma das tubas. **Conclusão:** SIN é mais frequente em pacientes melanodérmicas, na faixa etária média de 35 anos, com história clínica de infertilidade, de gravidez tubária inicial e recorrente e consequente obstrução

Submitted: 2014/07/20
Approved: 2015/06/25

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tubária caracterizada por lesão na porção ístmica cornual da tuba uterina, geralmente de comprometimento bilateral.

Palavras-chave: Salpingite; Salpingite/radiografia; Histerossalpingografia; Infertilidade; Gravidez Tubária.

INTRODUCTION

Salpingitis isthmica nodosa (SIN),¹⁻⁵ also called^{6,7} adenosalpingitis, productive glandular salpingitis, epitheliomatosis, adenoid-hyperplasia, tubal diverticulosis, endosalpingiosis, and tubal adenomyosis is a rare condition involving the fallopian tubes characterized by small diverticula or nodular thickening of the tunica muscularis, extending into the lumen wall, commonly involving the isthmus portion of one or two uterine tubes. It was first described by Chiari in 1887 and has since been the subject of discussion as to its etiology and pathology.¹

It can be reliably diagnosed by hysterosalpingography,^{2,3} being associated with an increased incidence of infertility and ectopic pregnancy.³⁻⁵

The objective of this study was to present the review results of four cases of SIN in a private radiology clinic within 10 years and demonstrate the main findings through hysterosalpingography, site of involvement, and its impact on female infertility.

MATERIAL AND METHODS

A retrospective study reviewed 2,800 hysterosalpingography protocols performed between January of 1987 and January of 1997, in patients with a history of infertility assisted in a private clinic in the city of Belo Horizonte, Minas Gerais, Brazil.

Radiographic aspects found by two radiologists with specialization in Radiology and Diagnostic Imaging were analyzed in these four patients including the site of involvement, peritonization or not in the

contrast medium, ethnicity, age, marital status, current clinical complaints, and clinical background.

The hysterosalpingography was carried out between the eighth and twelfth days of the menstrual period, using iodinated water soluble (+ polyvidone acetate meglumine) as the contrast medium.

The instruments and materials used for the exam were: vaginal speculum, Cheron forceps, Pozzi clamp, hysterometer, cannulae, and specific cones for the hysterosalpingography examination.

All examinations were performed in a radiological device with the Toshiba model 850 DG seriograph with imaging intensifier.

The radiographic criteria used as SIN diagnostic from the hysterosalpingography (HSG) were: contrast medium globules in periluminal tissue in continuity with the lumen of the fallopian tubes, with the aspect of multiple small diverticula such as a “honeycomb”. The clinical data of these patients were registered along with the record of when the HSG was performed, marital status, ethnicity, current clinical complaints, history, and the radiological findings, such as the site of involvement, unilateral or bilateral, contrast medium peritonization – positive Cotte proof.

RESULTS

The accumulation of contrast medium in the cornual tubal isthmus wall, unilaterally or bilaterally, was found radiologically in all four patients with the appearance of multiple small diverticula, such as a “honeycomb,” which characterized the diagnosis of SIN.

Patients were evaluated for all studied characteristics such as ethnicity, age, marital status, current clinical complaints, and medical history (Table 1). Table 2 reports the site and side of involvement and presence or absence of contrast medium peritonization (positive or negative Cotte proof).⁸

Table 1 - Clinical characteristics observed in four patients with salpingitis isthmica nodosa among 2,800 examinations conducted in a retrospective study of hysterosalpingography from January of 1987 to January of 1997, in patients with history of infertility assisted in a private clinic in the city of Belo Horizonte, Minas Gerais – Brazil

Cases	Ethnicity	Age	Marital status	Current complain	Clinical history
1	Melanodermic	36	Single	Primary infertility	None
2	Melanodermic	33	Single	Secondary infertility	Right tubal pregnancy
3	Caucasian	27	Married	Secondary infertility	Left tubal pregnancy
4	Melanodermic	45	Divorcee	Secondary infertility	Left tubal pregnancy

Table 2 - Description of the site and side of involvement, and the presence or absence of peritonization of the contrasting medium (positive or negative Cotté proof) in four patients with salpingitis isthmica nodosa selected among 2,800 examinations conducted in a retrospective study of hysterosalpingography from January of 1987 to January of 1997, in patients with history of infertility assisted in a private clinic in the city of Belo Horizonte, Minas Gerais – Brazil

Cases	Site	Side	Cotté proof	Illustrations
1	Cornual isthmic	Bilateral	Positive	Figure 1
2	Cornual isthmic	Bilateral	Negative	-
3	Cornual isthmic	Bilateral	Positive to the right	Figure 2
4	Cornual isthmic	Left	Negative	-

Among the four patients with SIN, three of them were melanodermic, and one was Caucasian, with age ranging from 27 to 45 years, and the average age at diagnosis of 35, two were single, one was married, and one was a divorcee. The complaint of secondary infertility was present in three patients with a history of prior tubal pregnancy; one patient complained of primary infertility and had no history of pregnancy.

In the four patients with SIN identified through the investigation of infertility using hysterosalpingography, the cornual isthmus region was the affected tubal portion, with bilateral tubal involvement in three and one-sided in one of the patients. The peritonization of the contrast medium showed evidence of bilateral positive Cotté proof in one, complete bilateral obstruction in two and positive Cotte proof in one of these two tubes, with the right one permeable and the left one obstructed (Figures 1 and 2).

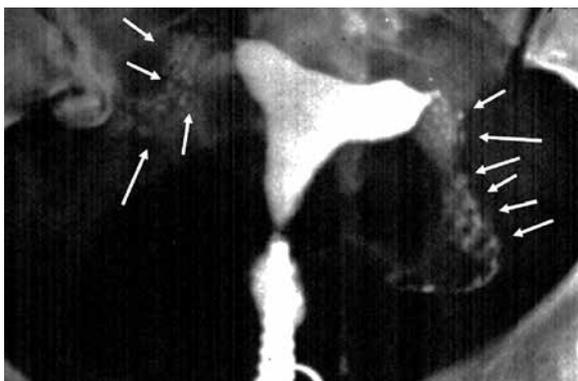


Figure 1 - Both tubes are completely opaque, with positive Cotté proof, with multiple parietal diverticular formations in the cornual isthmic portion (white arrows).



Figure 2 - Patient with a history of left tubal pregnancy. The x-ray shows multiple saccular formations in the cornual isthmic portion of both tubes (contained inside the white circle). The right uterine tube is fully opaque with peritonization of the contrasting medium (positive Cotté proof) while the left tube is fully obstructed (negative Cotté proof).

DISCUSSION

Even with recent technological advances such as the use of ultrasound and magnetic resonance imaging in the study of diseases of the female pelvis that can assess infertility, HSG remains as a relatively quick, safe, and noninvasive method and the best method for the evaluation of tube obstruction.⁹

In 1896, Von Recklinghausen⁷ proposed that the SIN findings were due to Wolffian remains in the region where the Mullerian and Wolffian ducts cross. However, the histopathological evaluation of tubes tissue from female embryos and adolescents did not show Wolffian remains. Among the various hypotheses to explain the possible etiology of SIN, one is based on previous inflammatory processes occurring in the fallopian tube with abnormal proliferation of the epithelium in the isthmic portion and slow penetration into the tube wall forming a maze of trails and cysts leading to secondary muscle hypertrophy.⁷ Benjamin and Beaver consider that SIN would be an acquired anomaly, similar in all respects to adenomyosis. The subsequent metaplastic alteration would be responsible for the transformation of the tubal epithelium into endometrial in the pseudo glands present in the mesosalpinx. This, in turn, undergoes hypertrophic and hyperplastic alterations in analogy to what is observed in the myometrium.¹ Its etiopathogenesis can have a post-infectious nature, a congenital or developmental defect, or be the result of metaplasia endosalpingiosis. It can also be a morphological ex-

pression of a chronic tube spasm.⁷ The analysis of the exposed theories suggests an etiologic multifactorial concept responsible for the pathogenesis of SIN.⁶

SIN is commonly discovered during the investigation of primary and secondary infertility, based on the HSG, with no typical pattern of gynecological symptoms. In the cases presented here, it was revealed that out of the four patients with SIN, three were melanodermic, and one was Caucasian, with age ranging from 27 to 45 years, and an average age at diagnosis of 35, two were single, one was married, and one was a divorcee. Three patients had complains of secondary infertility with the previous history of tubal pregnancy and one patient had complains of primary clinical infertility and no history of pregnancy, consistent with data in literatura.^{1,10-12}

Although its exact incidence is unknown, some literature reports estimate it between 0.6 and 3.9% of cases.^{1,6,13} In the four infertile patients with SIN, the prevalence, was 0.14% HSG in 2800, with three clinical history of ectopic pregnancy detected, similar to that reported in the literature.^{4-6,9,14} The low incidence found is due to the fact that HSG was performed in a private radiology clinic while the incidence found in the literature^{1,15,16} is from radiological examinations in hospital services, as demonstrated by Creasy et al.¹, who found 45 cases of SIN in 1,194 HSG performed in a six-year interval.

SIN was first demonstrated by HSG in 1951¹¹ and radiologically characterized by the accumulation of contrast medium into the tubal wall distributed in the cornual isthmic portion, unilaterally or bilaterally, with the appearance of multiple small diverticula, which may have the aspect of “honeycombs”, as observed in these four cases. According to Tulandi et al.¹¹ in 1983, these diverticular formations in the tubes would be the cause of infertility or ectopic pregnancy.

In all four patients with SIN presented here, the tube portion affected was the cornual isthmic region showing bilateral tubal involvement in three patients (75%) and unilateral in one (25%). The peritonization of the contrasting medium was observed through the bilateral positive Cotté proof, two with complete bilateral obstruction and one positive Cotté proof in one of the tubes, with the right one permeable and the left one obstructed. Creasy et al. found SIN with isthmus cornual bilateral involvement in 55.5% and Karasick et al.¹² in 80% of cases.

SIN seems to be associated with the cause of female infertility^{6,9,17} resulting from anatomical and functional alterations in the cornual isthmian tubal

region. The tubal obstruction is not pathognomonic in this disease because most cases of SIN present permeable tubes with the imaging aspect of multiple diverticula in the cornual isthmic portion of the tube, with or without associated hydrosalpinx. However, the patient with SIN is more likely to: develop tubal pregnancy in the Isthmian and ampullar portions¹⁻¹⁴ and show infertility and miscarriage. The diagnosis is established by HSG, considering endometriosis and fallopian tube tuberculosis as differential diagnoses.

Rough and irregular diverticular formations are observed in tubal endometriosis, not confined to the cornual and/or ampullar isthmian tubal region associated with tubal obstruction and typical symptoms;¹⁸ while in tubal tuberculosis the tube is fixed, rigid, and obstructed with irregular formations on its wall, sometimes alternating with stenosis and dilations in “rosary”, usually involving adnexal calcifications and/or lymph nodes in the pelvic excavation.^{18,19}

CONCLUSION

SIN is more common in melanodermic patients in the average age of 35 years, with a history of infertility, initial and recurrent tubal pregnancy, and subsequent tubal obstruction characterized by a lesion in the cornual isthmic portion of the fallopian tube, usually with bilateral involvement. Radiologically, it is characterized by the accumulation of contrast medium into the tubal wall, with the appearance of multiple small diverticula, with the possible appearance of “honeycombs”, making up the differential diagnosis of endometriosis and tuberculosis in the tubes.

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