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## Paratubal endosalpingiosis: a case report

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## ABSTRACT

**INTRODUCTION:** Endosalpingiosis is a rare condition and its pathogenesis and clinical significance are not fully understood. The unfamiliar presentation of this disease can misguide health care professionals. The reports available on this matter describe it as a disease of older age. In the majority of the cases it is an incidental finding in those with other concurrent gynecologic conditions.

**PRESENTATION OF CASE:** Here we report the case of a 14-year-old female who presented with abdominal pain and no specific past medical history. Imaging and physical examination were highly suggestive of appendicitis. With the initial diagnosis of acute abdomen, she underwent surgery. During the surgery gastrointestinal tract organs were found to be normal and in gynecological examination, the ovaries were normal, however the fallopian tube revealed a twisted paratubal cyst which was removed and sent to the laboratory for further investigation. On the basis of the cellular pathology, tubal like epithelium in the cyst was found and patient diagnosed with endosalpingiosis.

**DISCUSSION:** We suggest that in our case, the lesion possibly originated from the metaplasia of coelomic membrane into tubal cells. There are studies suggesting that co-occurring diseases should be considered when the diagnosis of endosalpingiosis is established and further studies are needed in regard to this matter.

**CONCLUSION:** Endosalpingiosis, although being a rare condition, should be included in our differential diagnosis since co-existing comorbidities are a possibility in patients with endosalpingiosis and can be of great importance.

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## 1. Introduction

Endosalpingiosis is the benign emergence of ectopic, cystic glands lined by tubal-like epithelium. It mostly involves the uterus, ovary and fallopian tube but can be found anywhere within the abdomen and pelvis [1,2]. This condition of uncertain origin was first found by Sampson in 1930 [3]. The appearance is seemingly due to metaplastic change of coelomic cells into tubal-like epithelium in which they have undergone tubal or serous differentiation [4].

It is fairly common upon checking several studies amongst patients undergoing gynecological procedures [5–7] with one of them suggesting that it was found in 7.6 % of 1107 women undergoing laparoscopic procedures [7]. However, since it is not widely recognized by gynecologists, it can be easily misdiagnosed with neoplastic lesions. Therefore, it is worthy of further attention. Alto-

gether the exact prevalence is not yet clear to epidemiologists. The diagnosis relies on biopsy of the lesion and inspection of cell morphology in the lab.

It occurs in women of all ages however a study had found that postmenopausal and older age women are at a much higher risk [1]. A study implied 52 percent (n = 15) of patients were between 30 and 49 years old [2].

Few cases of abdominal pain which ended up being known as endosalpingiosis have been reported but it appears to present more as pelvic pain. [1,8] At the same time, patients may suffer from one or more of these symptoms: fever, vaginal bleeding [9]; nausea and vomiting along with pain at iliac fossa [10]; dysmenorrhea, pelvic discomfort, dysuria and hematuria in the presence of bladder or ureter lesions or even asymptomatic [11]; chronic back pain for several years [12]. Additionally, they may have no remarkable history or have been administered to the hospital for several procedures. We should rely on symptomatic diagnosis cautiously because of the possible concurrent conditions, such as in a study which showed 34.5 % of patients had co-occurring endometriosis [1].

We report on a 14-year-old female who presented to the ER seeking treatment primarily for her abdominal pain which proved

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to be a case of endosalpingiosis on her fallopian tube. She was treated with surgery in which the excision of the tubal cyst was performed. Also the work has been reported in line with the SCARE criteria [13].

## 2. Case presentation

The patient, a 14-year-old female, presented to the emergency room with complaints of abdominal pain. The pain suddenly started about 8 h before admission, with higher intensity in the right lower quadrant of the abdomen that was slightly relieved by flexing the right hip. It was a non-colic constant pain with no relation to eating. She had no such pain before and the history of medication intake and previous diseases was unremarkable. An accurate investigation of the accompanying symptoms provided us with loss of appetite, nausea and vomiting containing food particles, without having or detecting fever. The defecation was normal. She had no complaints of urinary tract symptoms and the last menstruation was two weeks before (patient's irregular periods were also noticed but she didn't have heavy bleedings). On examination, the patient was alert and her vital signs were normal with no sign of icterus, paleness or cyanosis but a brief dehydration was revealed in the mucosa of the patient. Abdominal obesity was present. The abdomen was without distention, without any surgical scarring or muscle guarding. In palpation a rebound tenderness was observed in right lower quadrant of the abdomen.

Lab test revealed WBC of 15,600/mL, PMN of 82 %, Beta HCG Negative and CRP of 56. Urine analysis was reported normal as well. A blind loop which was aperistaltic, non-comprehensible, 7 mm in diameter and strongly in favor of appendicitis was reported in abdominal sonography.

After fluid therapy and starting antibiotics the patient was sent to the operating room with the initial diagnosis of acute abdomen. The abdomen was opened in the operating room with a McBurney incision. Upon entry into the abdomen a little bloody discharge was noted. There were no purulent and biliary discharges. Food and feces residues were not detected. The appendix, cecum and 100 cm distal to the terminal ileum were normal. The ovary was also normal. In examining of the fallopian tube, torsion of a paratubal cyst was evident and it was decided to release the bundle leading to torsion first. It was a single 2 cm cyst that became deteriorated (Fig. 1). As the gangrene was complete, resection was performed and the section was handed over to the pathology laboratory for histological investigation of the specimen. The patient was kept under observation for 1 day. Finally, by the time our interventions seemed to resolve the situation, with the patient having stable vital signs. After good tolerance of a normal diet and in a good general condition the patient was discharged, with prescriptions for analgesics and antibiotics and proper information about warning signs of possible further complications. Follow up showed no complications. Endosalpingiosis was reported in the pathological examination (Fig. 2). Written informed consent was obtained from the parent of the patient for publication of this case report and accompanying images

## 3. Discussion

In an article, Sampson described that in surgical procedures, parts of tubal or uterine epithelium may be introduced to another location be it in the same organ or to another location and seeding a tissue growth leading to endosalpingiosis; differentiating it from the endosalpingiosis with non-operative origin is of no clinical consequence but the ones with operative origin form most of the cases [3]. To the best of our knowledge, endosalpingiosis is commonly found in women of reproductive age and older, suggested



Fig. 1. A single 2 cm paratubal cyst shown during the surgery.

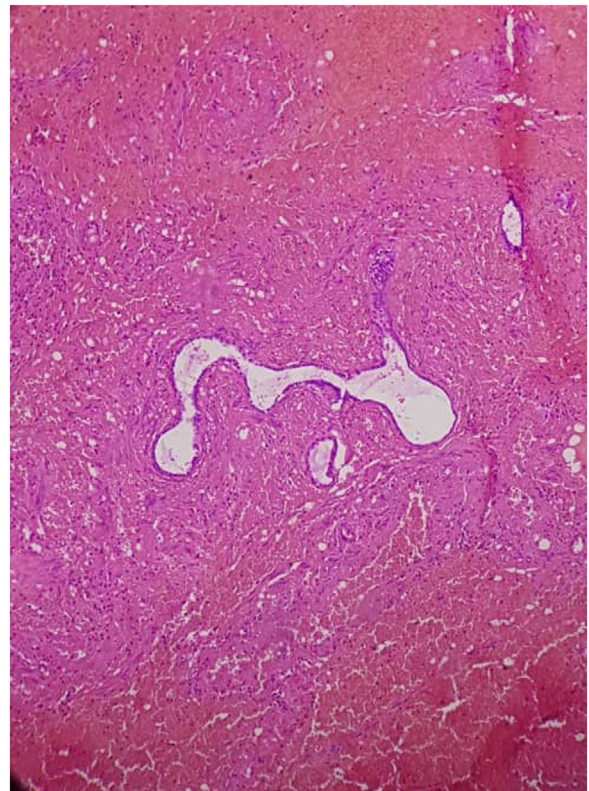


Fig. 2. Pathological examination showing endosalpingiosis.

mean age is 51.3 and 43 [1,14] and it is associated with a range of different symptoms including abdominal or lower abdominal pain, pelvic pain, hypermenorrhea or dysmenorrhea, abnormal uterine bleeding, bowel symptoms and plus asymptomatic cases, the list of symptoms goes on [1,14].

Presented in this report, is a 14-year-old female with the chief complaint being lower abdominal pain, who underwent surgery with the diagnosis of acute abdomen; however, since the gastrointestinal organs were clear of abnormalities, during exploratory procedures a tubal cyst was found and laboratory confirmed the diagnosis of endosalpingiosis.

Our patient didn't have any previous surgeries or specific history to guide us through the diagnosis; in addition, regarding the study with the theory of planted seedlings during surgery or trauma [3], that wasn't the case for us and it didn't induce the proliferation of ectopic tubal cells in our patient; thus other possible origins should be responsible and appears to support the theory of coelomic metaplasia [4] as Wang provided data in support of stem cell differentiation and metaplasia [15].

The aim of our report was to show how easily it can masquerade as appendicitis; in other cases, it can mimic the pain that is very typical of common diseases. We may even leave it untreated if asymptomatic or with no severe symptoms. There are studies which strongly suggest that endosalpingiosis may be a sign of more serious co-occurring diseases even though those abnormalities might not present simultaneously [16]. Several studies suggest that endosalpingiosis may have correlations to neoplasms [6] and Inflammatory tubal diseases. 42.1 % (n = 16 of 38) of tubal disease cases also had endosalpingiosis but this number is less significant in ovarian carcinoma which is 6.4 % (only 3 of 47) (17). In addition, there was another mention of coexisting conditions such as endometriosis in one third of patients with endosalpingiosis as well as uterine and ovarian cancers [18]. Gynecologic malignancies occur at a significantly higher rate in premenopausal individuals with endosalpingiosis, as in our presented case, than in those without [1]. Thus, patients should be informed about their risks.

It is made clear that the disease can happen in women of younger age with no specific history and we understand that the impact of this phenomenon is unclear for them.

It can be quite challenging to diagnose such abnormality due to its rarity and non-specific symptoms, specially at such an early age. This condition may be, as the definition implies, benign but we better recognize these patients as individuals susceptible to other important diseases. Amongst the possible symptoms of endosalpingiosis, infertility is one on the top of the list, and at such an early age we don't have the possibility of investigating this symptom, so patients may remain unaware of their potential infertility when diagnosed. Adding complexity and rarity to the presentation of our case. Thus, further studies and research in identifying risks of being diagnosed with this condition, are recommended.

In conclusion, this hard-to-diagnose condition merits further investigation due to its unknown potential for guiding future diagnoses of other co-occurring conditions in patients with endosalpingiosis. It should be included in differential diagnosis of possible cases because despite being benign in nature, potential risk of other comorbidities are as of yet unknown.

## Conflicts of interest

All authors (Fateme Sadat Mahdavi, Mehdi Tavallaei, Arsh Haj Mohamad Ebrahim Ketabforoush, Mahsa Bahadorinia) declare that they have no competing interests, and also the authors verifying that the manuscript is original, and the manuscript is not currently under consideration for publication in another journal.

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## Ethical approval

In our institution, this case report study is exempt from ethical approval.

## Consent

Written and signed consent was obtained from the parent of the patient for publication of this case report and accompanying images.

## Author contribution

FSM contributed in design of the study, acquisition of data, and drafting the article and approved the final version version of the manuscript for submission.

MT contributed in study design, writing the paper and review and edited the final version of the paper.

AEK and MB contributed in data gathering and drafting the main article, and final approval of the submission.

## Guarantor

Dr. Mehdi Tavallaei is the Guarantor of this work.

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## References

- [1] L. Prentice, A. Stewart, S. Mohiuddin, N.P. Johnson, What is endosalpingiosis? *Fertil. Steril.* 98 (4) (2012) 942–947.
- [2] R.K. Khangura, V.R. Katukuri, S.M. Lieblein, Z. Sharif, T. Anwar, Z. Zhang, Understanding Endosalpingiosis [89], *Obstet. Gynecol.* 125 (2015) 35S.
- [3] J.A. Sampson, Postsalpingectomy Endometriosis (Endosalpingiosis), *Am. J. Obstet. Gynecol.* 20 (4) (1930) 443–480.
- [4] R. Bermejo, A. Gómez, N. Galiana, A. Campos, R. Puente, E. Bas, et al., Peritoneal mullerian tumor-like (endosalpingiosis-leiomyomatosis peritoneal): a hardly known entity, *Case Rep. Obstet. Gynecol.* 2012 (2012), 329416.
- [5] A LH, K PA, K A, C SI, Cystic endosalpingiosis of uterine parametrium- a scarcely encountered and sparsely documented entity, *J. Clin. Diagn. Res.* 8 (10) (2014), Fd06-7.
- [6] K.M. Esselen, S.K. Ng, Y. Hua, M. White, C.A. Jimenez, W.R. Welch, et al., Endosalpingiosis as it relates to tubal, ovarian and serous neoplastic tissues: an immunohistochemical study of tubal and Müllerian antigens, *Gynecol. Oncol.* 132 (2) (2014) 316–321.
- [7] M.H. Hesselting, R.L. De Wilde, Endosalpingiosis in laparoscopy, *J. Am. Assoc. Gynecol. Laparosc.* 7 (2) (2000) 215–219.
- [8] M.R. Laufer, A.E. Heerema, K.E. Parsons, R.L. Barbieri, Endosalpingiosis: clinical presentation and follow-up, *Gynecol. Obstet. Invest.* 46 (3) (1998) 195–198.
- [9] A. Cajigas, C.A. Axiotis, Endosalpingiosis of the vermiform appendix, *Int. J. Gynecol. Pathol.* 9 (3) (1990) 291–295.
- [10] G. Chakhtoura, H. Nassereddine, J. Gharios, A. Khaddage, Isolated endosalpingiosis of the appendix in an adolescent girl, *Gynecol. Obstet. Fertil.* 44 (11) (2016) 669–671.
- [11] M. Habiba, I. Brosens, G. Benagiano, Müllerianosis, endocervicosis, and endosalpingiosis of the urinary tract: a literature review, *Reprod. Sci.* 25 (12) (2018) 1607–1618.
- [12] A.H. Scheel, J. Frasnec, W. Meyer, P. Ströbel, Cystic endosalpingiosis presenting as chronic back pain, a case report, *Diagn. Pathol.* 8 (2013) 196.
- [13] R.A. Agha, M.R. Borrelli, R. Farwana, K. Koshy, A.J. Fowler, D.P. Orgill, et al., The SCARE 2018 statement: updating consensus Surgical CAse REport (SCARE) guidelines, *Int. J. Surg.* 60 (2018) 132–136.
- [14] J. Heinig, I. Gottschalk, U. Cirkel, R. Diallo, Endosalpingiosis—an underestimated cause of chronic pelvic pain or an accidental finding? A retrospective study of 16 cases, *Eur. J. Obstet. Gynecol. Reprod. Biol.* 103 (1) (2002) 75–78.

- [15] Y. Wang, M.S. Sessine, Y. Zhai, C. Tipton, K. McCool, R. Kuick, et al., Lineage tracing suggests that ovarian endosalpingiosis does not result from escape of oviductal epithelium, *J. Pathol.* 249 (2) (2019) 206–214.
- [16] M.H. Chui, I.M. Shih, Oncogenic BRAF and KRAS mutations in endosalpingiosis, *J. Pathol.* 250 (2) (2020) 148–158.
- [17] K.R. Zinsser, J.E. Wheeler, Endosalpingiosis in the omentum: a study of autopsy and surgical material, *Am. J. Surg. Pathol.* 6 (2) (1982) 109–117.
- [18] K.M. Esselen, K.L. Terry, A. Samuel, K.M. Elias, M. Davis, W.R. Welch, et al., Endosalpingiosis: More than just an incidental finding at the time of gynecologic surgery? *Gynecol. Oncol.* 142 (2) (2016) 255–260.

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