

Case Report

Management of Isolated Tubal Torsion (ALADIN) during Emergency Laparotomy in Infertile Women

Tata Laksana dari Torsio Tuba Terisolasi (ALADIN) selama Laparotomi Emergensi pada Perempuan Infertil

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Abstract

Objective: To report ITT and primary infertility patient which came with acute abdominal pain.

Methods: Case report.

Case: A 36 yo female came to Koja Hospital with acute abdominal pain without sign of infections. Ultrasound findings show cystic mass on bilateral adnexal sized 76x28x39 mm and 31x51x43 mm with minimal ascites. During laparotomy, proximal right fallopian tube was torsioned four times clockwise. There was hydrosalping founded on contralateral tube, while right and left ovaries and uterus were normal. We performed salpingectomy.

Discussion: The clinical presentation of ITT is non-specific and it has become a challenge to physician to develop preoperative diagnosis. The spectrum of imaging findings may be wide range depending on adnexal pathology, degree of severity, and the duration of adnexal torsion. The recommendation of primary approach to tubal torsion is conservative management considering **ALADIN** (mALignAncy-Death tissue, INFertility).

Conclusion: The diagnosis is rarely be made before operation, due to non-specific clinical symptoms and imaging findings. The considerations to perform conservative management are malignancy, death tissue/necrotic, and infertility.

Keywords: hydrosalping, isolated tubal torsion malignancy-death tissue infertility.

Abstrak

Tujuan: Untuk membahas tentang pasien torsio tuba terisolasi dan infertilitas primer yang datang dengan keluhan nyeri abdomen akut.

Metode: Laporan kasus.

Kasus: Seorang perempuan 36 tahun datang ke RS Koja dengan nyeri abdomen akut tanpa tanda infeksi. Pemeriksaan USG menunjukkan massa kistik pada adneksa bilateral dengan ukuran 76x28x39 mm dan 31x51x43 mm dengan asites minimal. Selama laparotomi, tuba falopi proksimal kanan mengalami torsio empat kali searah jarum jam. Selain itu, ditemukan hidrosalping pada tuba kontralateral dan sementara ovarium kanan, kiri, dan uterus dalam batas normal. Kami melakukan salpingektomi.

Diskusi: Presentasi klinis dari torsio tuba terisolasi tidak spesifik dan menjadi tantangan dalam diagnosis preoperatif. Gambaran penunjang dapat bervariasi bergantung pada patologi adneksa, derajat keparahan, dan lama torsio adneksa. Rekomendasi pendekatan torsio tuba ialah konservatif melalui ALADIN (mALignAncy-Death tissue, INFertility).

Kesimpulan: Diagnosis dapat dilakukan sebelum operasi karena pemeriksaan klinis dan gambaran penunjang yang tidak spesifik. Pertimbangan melakukan tatalaksana konservatif ialah keganasan, jaringan yang nekrotik, dan infertilitas.

Kata kunci: hidrosalping, keganasan – infertilitas, torsio tuba terisolasi.

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INTRODUCTION

Isolated tubal torsion (ITT) is rare cases with incidence 1 in 1.5 million women. The challenge of diagnosing ITT preoperatively is its non-specific symptoms such as lower abdominal pain, while in acute condition it is accompanied with nausea, vomiting¹ Moreover, imaging findings for ITT are non-specific commonly diagnosed as ovarian cysts. One factor that may cause ITT is hydrosalping and it is often correlated with infertility condition. Therefore, management regarding ITT should be carefully decided based on each patient. However due to delayed diagnosis, surgical salpingectomy is commonly performed as decision during intraoperative findings. Nevertheless, there are very few cases managed with detorsion of fallopian tube.² Here we reported case of ITT patients and primary infertility came to hospital with acute abdominal pain in order to discuss its diagnostic features and surgical management.

CASE

A 36 yo female came to Koja Hospital with acute abdominal pain without sign of infections. Ultrasound findings show cystic mass on bilateral adnexal sized 76x28x39 mm and 31x51x43 mm with minimal ascites (Figure 1), both ovaries were

difficult to identified due to the abdominal pain. Laboratory results show normal hemoglobin and leucocytosis. During laparotomy, there was minimal ascites found on the pelvic floor with proximal right fallopian tube was torsioned four times clockwise. There was hydrosalping founded on contralateral tube, while right and left ovaries and uterus were normal. (Figure 2A-C). The right fallopian tube was gangrenous seen with hemorrhagic fluids enlarged to sized 7x4x3cm. Salpingectomy was performed due to the necrotic condition. Histopathology result shows hydrosalping and paratubal cyst (Figure 3A-B).



Figure 1. Ultrasound findings of bilateral adnexal masses with minimal ascites



Figure 2-A. Right fallopian tube was torsioned four times clockwise (arrow); 2-B. Further exploration: right and left ovaries and uterus were normal, with left hydrosalping; 2-C). Right salpingectomy was performed

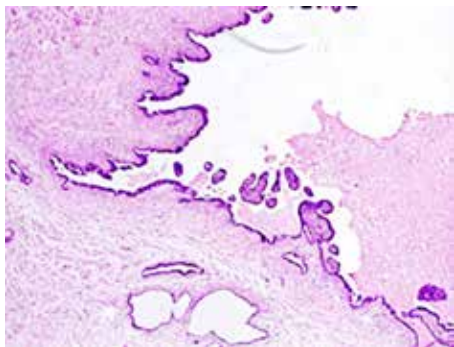


Figure 3-A. Histopathology finding of the fallopian tube showed distortic plicae lined by ciliated simple columnar epithelium. Acute and chronic inflammatory cells infiltrated the stroma. (Magnification 40x)

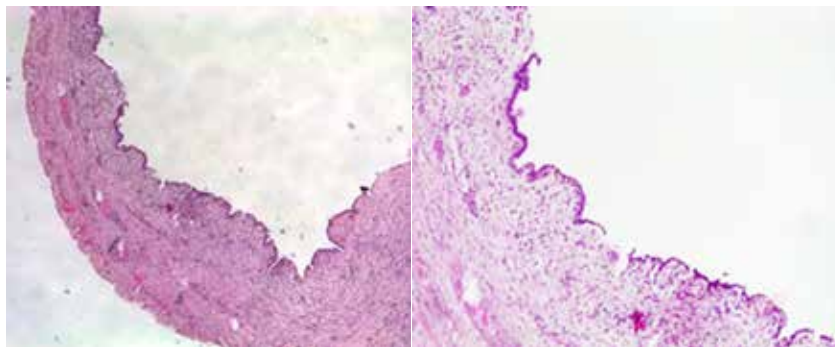


Figure 3-B. Histopathologic findings of the fallopian tube showed a cystic space lined by simple columnar epithelium. Chronic inflammatory cells infiltrated the stroma (Magnification Left: 40x, Right : 100x)

DISCUSSION

Isolated tuba torsion (ITT) is a very rare entity and comprises of minor group among etiologies of abdominal pain in adolescents. It is torsion of fallopian tube without ovarian torsion involvement. This condition can be precipitated by intrinsic and extrinsic factors. Intrinsic factors are pelvic inflammatory disease (PID), hydrosalping, prior tubal surgery, primary fallopian tube malignancy, and abnormal length of mesosalping and spiral course of the salping. The extrinsic pathologies are scarring from endometriosis, prior pelvic surgery, gravid uterus, malignancy or tumor of the adjacent structures, and paraovarian or paratubal cysts.^{3,4} ITT may be considered as one causes of unexplained infertility. Two hundred and thirteen of four hundred and nine (52.1%) unexplained infertility patients were found to have hydatid of Morgagni cysts⁵ Through impeding ovum pick-up, hydatid of Morgagni may be considering as new factors in infertility. This cyst can trigger the isolated torsion of morgagni hydatids with or without fimbria.⁶ One theory that may include in this pathology findings is stimulation of reproductive axis in high follicle stimulating hormone (FSH) levels long before the onset of menses in a case with asymptomatic distal occlusion of the tube and asymptomatic pelvic inflammation in adnexal area.⁷ Hydrosalping might be a secondary finding in ITT cases and might not be responsible as the main causes of ITT.^{3,4} But in this case, hydrosalping might be consider as one underlying cause due to the left hydrosalping findings.

The clinical presentation of ITT is non-specific and it has become a challenge to physician to

develop preoperative diagnosis. Acute severe lower abdominal pain is commonly present, with or without accompanied by nausea, vomiting, and fever. Physical and laboratory exams are also not specific to diagnosed ITT. Some findings show leukocytosis caused by necrosis condition.^{8,9} ITT is predominantly appears on the right side due to mild immobilization of left tube by the proximity to sigmoid mesentery. Thus, patients with right lower abdominal pain will be undergoing operation faster than left side because suspicion of appendicitis.¹⁰ However, 6 cases in which ITT occurred on the left side.¹¹

The spectrum of imaging findings may be wide range depending on adnexal pathology, degree of severity, and the duration of adnexal torsion. Normal fallopian tube is rarely visible on ultrasonography because of its narrow diameter and lack of clear echogenic features. Thus, dilated tube might show thickened, echogenic walls, with internal debris or echogenic mass. It is recommended to use color Doppler ultrasound, to differ normal ovary with other pelvic cyst. The whirlpool sign represents tissue mass twisted around central axis, by moving transvaginal ultrasound probe back and forth along axis of suspected torsion.^{1,9} CT-scan might not be superior compare to transvaginal ultrasound, therefore ultrasound might considered as first line of diagnostic imaging.¹² The progression of torsion causes hematosalping, tubal rupture, and peritubal hematomas, which become more complicated and harder to diagnose. This heterogenous mass may confused with ruptured ectopic pregnancy. ¹Magnetic Resonance Imaging (MRI) is valuable in recognizing adnexal torsion with most common findings thickening

tube, ascites and uterine deviation to twisted side. Whirlpool sign might be observed in T2-weighted imaging.¹³

The recommendation of primary approach to tubal torsion is conservative management considering **ALADIN** (mALignAncy-Death tissue, **IN**fertility). MALIGNAncy, if imaging/intraoperative findings shows possibility of malignancy then it should be performed radical surgery (referred to oncologist if possible).⁹Death tissue/necrotic, Due to delayed diagnosis, most cases were presented with necrotic tube. If detorsion was performed then reperfusion (color appearance), vascularization, and viability. This salvation has risk of thromboembolic events. Thus, salpingectomy was preferred in most necrotic cases.² Infertility, fertility status is important to consider the conservative management in this patient, Fertility-sparing surgery may be performed in infertility patients, using laparoscopic detorsion of fallopian tube to preserve fertility.⁹ After detorsion, it should be watched for reperfusion and associating pathologies. The salvage of the tube should be evaluate its viable because of future fertility.²

Early laparoscopy is reference standard in diagnosis and treatment. Recovery after laparoscopy is faster and causes fewer post-operative adhesions. Timing of diagnosis is very important in ITT which delay may cause irreversible consequences; therefore best treatment can be performed for the patients.⁹ After laparoscopic detorsion, follow up using ultrasound must be performed to document possibility of recurrent torsion or hydrosalping.³

CONCLUSION

Gynecologist should be aware of isolated tubal torsion as differential diagnosis in lower abdominal pain. Possible complications possess a risk for future fertility. The diagnosis is rarely be made before operation, due to non-specific clinical symptoms and imaging findings. The considerations to perform conservative management are malignancy, death tissue/necrotic, and infertility (**ALADIN**). Thus,

laparoscopic may be consider as diagnostic tools and laparoscopic detorsion may be consider as primary conservative management to preserve fertility.

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