IDCases 17 (2019) e00578

Contents lists available at ScienceDirect

IDCases

journal homepage: www.elsevier.com/locate/idcr

Case report

Ovarian abscess caused by *Helicobacter cinaedi* in a patient with endometriosis

Hiroshi Sato*, Atsuko Ogino, Sunao Matsuzaka, Yuna Asami, Shota Kanbayashi, Miho Masuda, Ayaka Nakashima, Miki Yasuda, Hajime Morishita, Yukiko Ando, Kenji Oida, Nao Taguchi, Masaya Hirose

Department of Obstetrics and Gynecology, Hyogo Prefectural Amagasaki General Medical Center, Amagasaki, Japan

ARTICLE INFO

Article history:
Received 17 May 2019
Received in revised form 13 June 2019
Accepted 13 June 2019

Keywords:
Helicobacter
Laparoscopy
Ovary
Abscess
Endometriosis

ABSTRACT

Helicobacter cinaedi is a rarely encountered pathogen that easily induces bacteremia. Various foci of *H. cinaedi* infection have been reported; however, no case of adnexal abscess caused by *H. cinaedi* has been reported in the English literature. We herein report a case of ovarian abscess caused by *H. cinaedi*.

A 38-year-old nulligravid Japanese woman was admitted to our hospital with an adnexal abscess. Clinical findings included fever, leukocytosis, and elevated C-reactive protein. Laparoscopic right partial oophorectomy with abdominal lavage was performed. *H. cinaedi* was isolated from cultures of blood and ovarian abscess fluid after surgery. Intravenous ampicillin/sulbactam was administered for 2 weeks, followed by oral amoxicillin for an additional 2 weeks. The postoperative course was uneventful and clinical findings improved. There was no evidence of relapse. *H. cinaedi* can cause ovarian abscess and is likely an under-recognized pathogen.

Introduction

Helicobacter cinaedi is a gram-negative spiral bacterium. *H. cinaedi* infection was first reported in 1984 in a man with proctitis. Since then, various foci of *H. cinaedi* infection have been reported [1]. In Japan, the first reported isolation of *H. cinaedi* was in 2003. A few cases of intrauterine infection during pregnancy are the only reports of *H. cinaedi* infection of the female reproductive system [2]. There are no reports of ovarian abscess caused by *H. cinaedi* in the English-language literature to date. Herein we report a case of ovarian abscess caused by *H. cinaedi* in a patient with endometriosis.

Case report

A 38-year-old nulligravid Japanese woman visited our hospital for further evaluation of intermittent fever. She was experiencing her usual menstrual cramps, without additional abdominal pain. The patient was otherwise healthy and had no pertinent medical history. Pelvic examination revealed a small amount of white, non-odorous vaginal discharge. A fist size mass was palpable. The patient had no tenderness on palpation. Initial laboratory tests included the following findings: white blood cell count, 14,000/mm^3 (differential: 88.0% neutrophils, 2.0% monocytes, 10.0% lymphocytes); hemoglobin, 12.0 g/dL; platelet count, 526,000/mm^3; and C-reactive protein, 10.55 mg/dL. Human immunodeficiency virus antibody was negative. A blood sample was taken for culture at that time.

A right ovarian cyst measuring 9 cm in diameter in its smallest dimension was found on transvaginal sonography. Computed tomography showed a well-enhanced, thick-walled, double-lobed ovarian cyst (Fig. 1). Uterine leiomyoma measuring 8 cm in diameter was also shown. The patient was admitted with a diagnosis of ovarian abscess. Soon after admission, percutaneous aspiration of the abscess was performed for bacterial culture. Dark brownish fluid was aspirated. After aspiration, intravenous ampicillin/sulbactam (12 g daily) was initiated. On hospital day 2, percutaneous drainage was attempted. However, the drainage tube could not be inserted into the abscess because of the thick cyst wall. The patient's clinical symptoms, including fever and abdominal pain, worsened. Therefore, laparoscopic right partial oophorectomy and abdominal lavage with 3000 mL normal saline was performed on hospital day 3. Mild adhesions were present between the right ovarian cyst and the retroperitoneum. The

* Corresponding author at: Department of Obstetrics and Gynecology, Hyogo Prefectural Amagasaki General Medical Center, 2-17-77 Higashinaniwa-cho, Amagasaki, Hyogo Prefecture 660-8550, Japan.
E-mail address: shiro801219@yahoo.co.jp (H. Sato).

https://doi.org/10.1016/j.idcr.2019.e00578

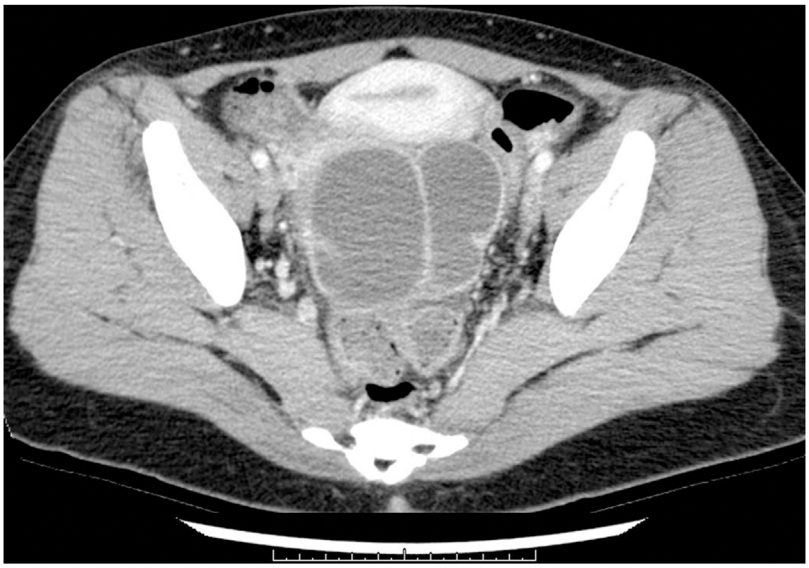

Fig. 1. Enhanced abdominal computed tomography reveals a well-enhanced, double-lobed ovarian cyst with a thick wall.

subserosal uterine myoma was shown at the fundus of the uterus. The left ovary and the both fallopian tubes were normal (Fig. 2A, B). During the operation, dark brownish fluid leaked from the right ovarian cyst. The postoperative course was uneventful.

The blood culture obtained at admission yielded gram-negative spiral bacteria on hospital day 5 (Fig. 3). The time to blood-culture positivity was 114 h. The blood agar plate showed characteristic thin, spreading colonies. We requested extension of the incubation period for the abscess culture; the culture tubes were monitored for a total of 14 days. Microaerobic culture of the abscess fluid was performed at 35 °C for 7 days. Intravenous antimicrobials were continued for 2 weeks. The patient's clinical symptoms and laboratory markers of inflammation improved. She was discharged on hospital day 12. The abscess culture yielded the same bacteria as the blood culture after 14 days of incubation. Because of the microbiological characteristics, we concluded that the bacteria was *H. cinaedi*. Bacterial identification of *H. cinaedi* was confirmed at an outside laboratory with polymerase chain reaction (PCR) targeting a 16S ribosomal RNA sequence. The patient's treatment was changed to oral amoxicillin (1000 mg/day) and was continued for 2 additional weeks. Histology indicated a right ovarian endometrial cyst with abscess (Fig. 4A, B). There was no evidence of infection relapse.

Discussion

To our knowledge, this is the first published case of an ovarian abscess caused by *H. cinaedi*. A few cases of intrauterine infection with *H. cinaedi* during pregnancy have been reported, but there are no reports of *H. cinaedi* infection in other female reproductive organs [2]. Various foci of *H. cinaedi* infection have been reported. The prognosis is generally good, but 30%–60% of patients have recurrent symptoms. The Centers for Disease Control and Prevention recommends long-term therapy lasting 2–6 weeks for *H. cinaedi*, rather than short-term therapy of up to 10 days [1]. Therefore, identification of *H. cinaedi* is very important when deciding the treatment duration.

The reported culture period to detect *H. cinaedi* in blood is at least 4–10 days [3]. If blood samples are incubated for 5 days or fewer, half of *H. cinaedi* bacteremia cases will be overlooked. In many cases, *H. cinaedi* is first detected on blood culture with an automatic blood culture system. Several automatic blood culture systems are available, including the BACTEC, BacT/ALERT, and VersaTREK systems. Our hospital laboratory uses the BACTEC system. Whereas blood culture samples are incubated for 3 or 4 days in many hospitals, the incubation time in our hospital is 7 days, which improves the sensitivity of detecting slow-growing organisms. Some studies have reported that the VersaTREK system can detect *H. cinaedi* more quickly and with higher sensitivity than other systems [1].

Gram-negative spiral bacteria detectable with blood culture include *H. cinaedi*, *Campylobacter* species, *Arcobacter butzleri*, *Desulfovibrio desulfuricans*, *Brachyspira pilosicoli*, and *Anaerobiospirillum succiniciproducens* [1,4–8]. These six kinds of bacteria are distinguishable according to microbiological characteristics. The bacteria that are likely to be positive in aerobic culture are *H. cinaedi*, *Campylobacter* species, and *Arcobacter butzleri* [1,4,5]. The other bacteria are likely to be positive in anaerobic culture [6,5–8]. A thin, film-like colony is characteristic of *H. cinaedi* and *Brachyspira pilosicol* [1,7]. There have been reports of adnexitis caused by *Campylobacter fetus* [9]. It is important to distinguish *H. cinaedi* from *Campylobacter* species. Yamamoto et al. [4] concluded that a cut-off of 75 h for time to blood-culture positivity was useful in distinguishing *Helicobacter* species bacteremia versus *Campylobacter* species bacteremia when using the BACTEC systems. Therefore, we distinguished between *H. cinaedi* and *Campylobacter* species on the basis of colony shape and the time to blood-culture positivity. PCR targeting the 16S ribosomal RNA cannot be performed at our institution. Therefore, we distinguish bacteria according to their microbiological characteristics. These characteristics are important because few hospitals have laboratories that can perform PCR targeting the 16S ribosomal RNA.

The route of bacterial infection is mainly transvaginal in tuboovarian abscess, with ovarian involvement occurring secondary to salpingitis. The infectious process in ovarian abscess is thought to differ from that in tuboovarian abscess. The following possible explanations of ovarian infection have been proposed [10]. Bacterial invasion of the parenchyma in the postoperative period after pelvic surgery is thought to be the most frequent route of ovarian infection. Contamination via the bloodstream or the lymphatic system are other possible sources of bacterial entry. The route of *H. cinaedi* infection is thought to be mainly fecal–oral. *H. cinaedi* colonizes the gastrointestinal tract and has a strong ability to invade blood vessels, which results in bacterial translocation from the intestinal tract to the vascular system.

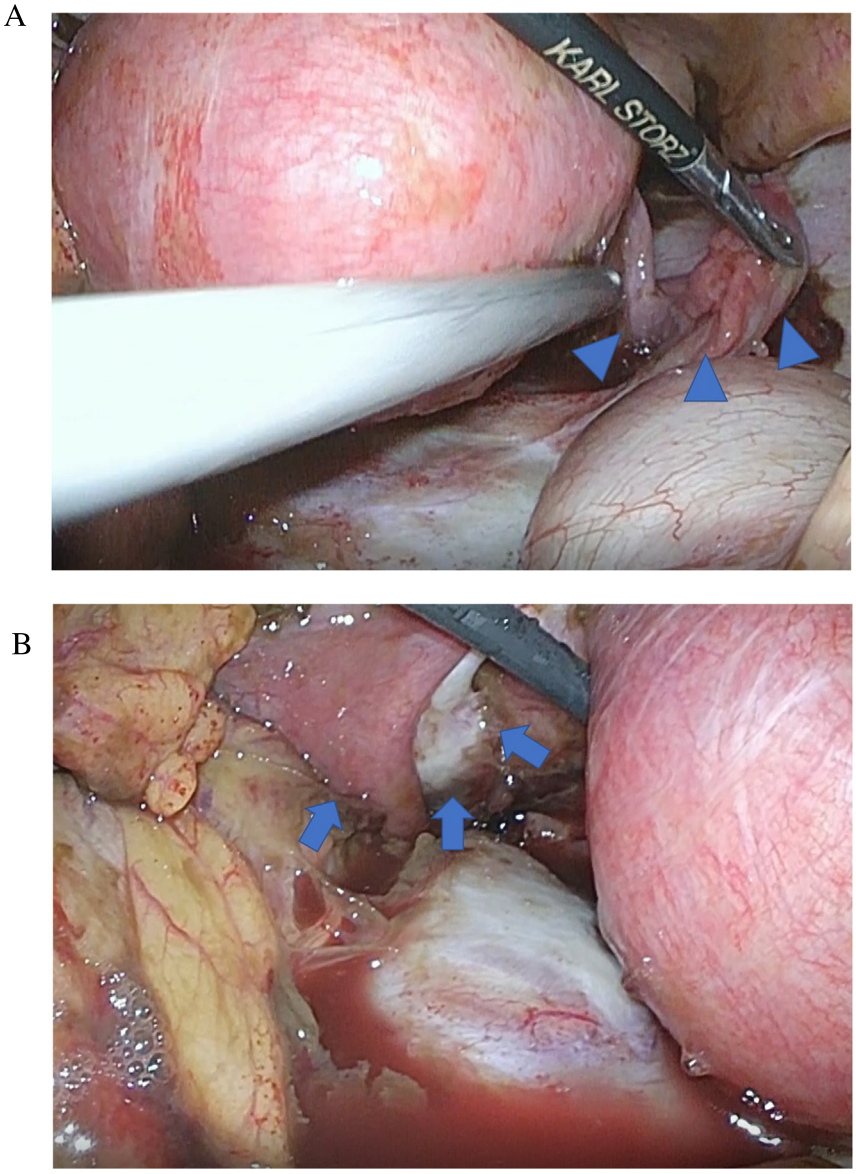

Fig. 2. Laproscopy revealed mild adhesion between the right ovarian cyst and the retroperitoneum. findings. The right fallopian tube(2A, Arrow head) and the left adnexa(2B, Arrow) were grossly normal. Uterine leiomyoma was shown at uterine fundus.

H. cinaedi causes bacteremia more frequently other *Helicobacter* species. In this case, the patient had not undergone pelvic surgery and bacteremia may have preceded endometrial cyst infection. The lack of infection in the right fallopian tube supports this theory. Ovarian endometriosis was detected in the histopathology of the resected ovary. Kubota et al. [11] concluded that the presence of endometrial cyst was a risk factor for the development of a tuboovarian abscess or an ovarian abscess. In this review, all ovarian abscesses were isolated in patients with endometrial cyst. The following factors were considered as the reasons for the increase of adnexal infection in patients with endometrial cysts: (1)Blood that is retained in the endometrial cyst might act as a medium for bacterial growth [12]. (2)The cyst wall of the endometrial cyst is weak and more susceptible to bacterial invasion compared with healthy ovarian epithelium [13]. Moreover, the recent studies investigating the link between endometriosis and the immune system revealed several immunological abnormalities [14]. Immunologicaldisorders may have some relevance to bacterial infection. Most women with endometriosis

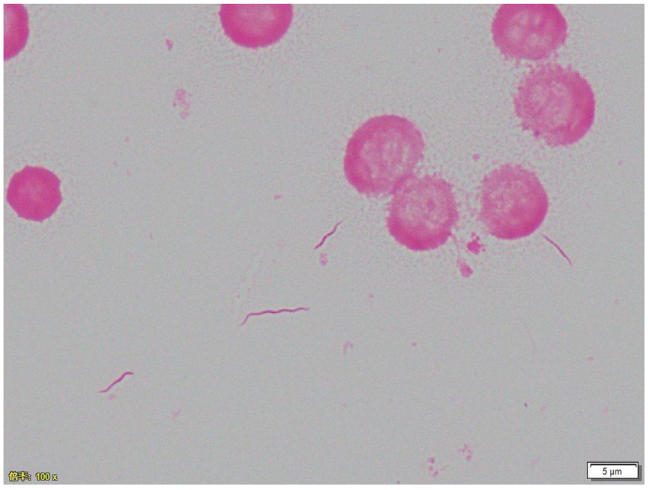

Fig. 3. Gram staining of spiral bacteria isolated in blood culture.(×1000).

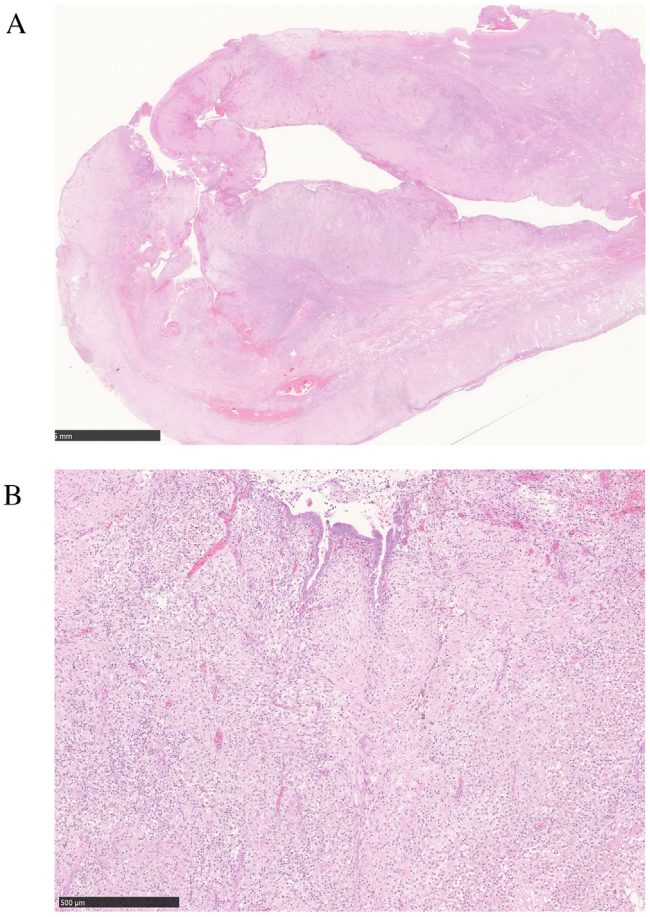

Fig. 4. On histopathology of right ovary, many neutrophils are seen in the ovarian epithelium and stroma(4A, Hematoxylin-Eosin stain, ×20). The low cuboidal epithelium with hemosiderosis is consistent with an endometrial cyst(4B, Hematoxylin-Eosin stain ×200).

including this patient may be apparently healthy. However, they may have some immunological disorders which can cause infection and/or cancer although further research is necessary. In this patient, ovarian endometrial cyst was thought to increase susceptibility to adnexal abscess for aforementioned causes.

There are few reports on the treatment of ovarian abscess. Most reports of adnexal abscess have described tuboovarian abscesses. Rosen et al. [15] concluded that laparoscopy should be considered in all patients with tuboovarian abscess who desire future conception. In their report, they proposed the following advantages of immediate laparoscopy: ability to obtain an accurate diagnosis, effective treatment under magnification with minimal complications, possibly faster response rates with shorter hospitalization times, and decreased infertility. However, image-guided percutaneous drainage is less invasive than laparoscopy. A retrospective study of image-guided drainage procedures in 49 women reported successful treatment without subsequent need for surgery in 74% of patients [16]. Although we successfully performed percutaneous aspiration, we could not insert a pig-tail tube for drainage and iatrogenic rupture of the abscess occurred. The findings at surgery indicated that the causes of the failure of percutaneous drainage were the thick wall and mobility of the abscess cyst, which did not have severe adhesions.

Laparoscopy and laparotomy are alternative treatment options for tuboovarian abscess. Garbin et al. [17] concluded that the expanded vision field of laparoscopy enabled conservative surgery, with shorter hospitalization, fewer complications, and faster resolution of fever than laparotomy. In both laparoscopy and laparotomy, either organ-preserving surgery (abscess incision and lavage only) or radical surgery (salpingectomy or salpingo-oophorectomy) may be performed. Buchweitz et al. [18] reported that operative laparoscopy with incision of the abscess cavity and abdominal lavage alone improved intraoperative and postoperative safety and long-term fertility compared with laparoscopic salpingectomy or salpingo-oophorectomy. We performed laparoscopic right partial oophorectomy and abdominal lavage. Because the ovarian cyst could have been a neoplastic lesion, partial resection of the ovary was needed. As a result, the ovarian endometrial cyst which bacteria could infect easily was resected and the recurrence of *H. cinaedi* infection might be prevented. When surgical treatment is needed for hemodynamically stable patients with adnexal abscess, a laparoscopic organ-preserving operation should be performed when possible. However, a resection of ovarian cysts which are likely to be endometrial cysts may be taken into consideration in order to prevent a recurrence of infection.

Adnexal abscess is a common complication of ovarian endometrial cysts [11,19]. Empirical treatment with antimicrobials may be initiated without taking blood culture samples and/or abscess samples. Various bacteria have been identified in reported cases of adnexal abscess. Some bacteria cannot be identified easily and require specific culture conditions; these are often overlooked as pathogenic organisms. Some cases of ovarian abscess caused by *H. cinaedi* may have been overlooked. Sampling of the abscess and blood culture is very important for identification of disease-causing bacteria. Gynecologists managing adnexal abscesses should cooperate with the bacterial laboratory at their hospital and remember that *H. cinaedi* is an under-recognized pathogen.

Authorship statement

Hiroshi Sato designed the study, and wrote the initial draft of the manuscript. Masaya Hirose contributed to analysis and interpretation of data, and assisted in the preparation of the manuscript. All other authors have contributed to data collection and interpretation, and critically reviewed the manuscript. All authors approved the final version of the manuscript, and agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All authors meet the ICMJE authorship criteria.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Acknowledgments

We thank Rebecca Tollefson, DVM, from Edanz Group (www.edanzediting.com/ac) for editing a draft of this manuscript.

References

[1] Kawamura Y, Tomida J, Morita Y, Fujii S, Okamoto T, Akaike T. Clinical and bacteriological characteristics of Helicobacter cinaedi infection. J Infect Chemother 2014;20:517–26, doi:http://dx.doi.org/10.1016/j.jiac.2014.06.007.
[2] Maki Y, Furukawa S, Kodama Y, Sumiyoshi K, Kino E, Sameshima H. Preterm labor and neonatal sepsis caused by intrauterine Helicobacter cinaedi infection. J Infect Chemother 2016;22:414–6, doi:http://dx.doi.org/10.1016/j.jiac.2015.12.008.
[3] Araoka H, Baba M, Kimura M, Abe M, Inagawa H, Yoneyama A. Clinical characteristics of bacteremia caused by Helicobacter cinaedi and time required for blood cultures to become positive. J Clin Microbiol 2014;52:1519–22.
[4] Yamamoto K, Hayakawa K, Nagashima M, Shimada K, Kutsuna S, Takeshita N, et al. Comparison of the clinical and microbiological characteristics of Campylobacter and Helicobacter bacteremia: the importance of time to blood

culture positivity using the BACTEC blood culture systems. BMC Res Notes 2017;10:634, doi:http://dx.doi.org/10.1186/s13104-017-2981-2.
[5] Arguello E, Otto CC, Mead P, Babady NE. Bacteremia caused by Arcobacter butzleri in an immunocompromised host. J Clin Microbiol 2015;53:1448–51, doi:http://dx.doi.org/10.1128/JCM.03450-14.
[6] Hagiwara S, Yoshida A, Omata Y, Tsukada Y, Takahashi H, Kamewada H, et al. Desulfovibrio desulfuricans bacteremia in a patient hospitalized with acute cerebral infarction: case report and review. J Infect Chemother 2014;20:274–7, doi:http://dx.doi.org/10.1016/j.jiac.2013.10.009.
[7] Hampson DJ. The spirochete Brachyspira pilosicoli, enteric pathogen of animals and humans. Clin Microbiol Rev 2017;31:, doi:http://dx.doi.org/10.1128/CMR.00087-17 pii: e00087-17.
[8] Kuir DG, Maloney S. Anaerobiospirillum succiniciproducens bacteraemia in the era of MALDI-TOF mass spectrometry. Pathology 2017;49:654–6, doi:http://dx.doi.org/10.1016/j.pathol.2017.05.011.
[9] Kitamura S, Matsumura N, Ohtake N, Kita M, Konishi I. Tubo-ovarian abscess with endometrial cyst probably infected by Campylobacter fetus: two cases. J Obstet Gynaecol Res 2016;42:1052–7, doi:http://dx.doi.org/10.1111/jog.13004.
[10] de Souza GA. Ovarian abscess with spontaneous vaginal drainage. Sao Paulo Med J 1997;115:1596–8.
[11] Kubota T, Ishi K, Takeuchi H. A study of tubo-ovarian and ovarian abscesses, with a focus on cases with endometrioma. J Obstet Gynaecol Res 1997;23:421–6.
[12] Padilla SL. Ovarian abscess following puncture of an endometrioma during ultrasound-guided oocyte retrieval. Hum Reprod 1993;8:1282–3.
[13] Daly JW, Monif GRG. Tuboovarian and ovarian abscesses. In: Monif GRG, editor. Infectious diseases in obstetrics and gynecology. Maryland: Harper & Row Publishers; 1974. p. 396–403.
[14] Izumi G, Koga K, Takamura M, Makabe T, Satake E, Takeuchi A, et al. Involvement of immune cells in the pathogenesis of endometriosis. J Obstet Gynaecol Res 2018;44:191–8, doi:http://dx.doi.org/10.1111/jog.13559.
[15] Rosen M, Breitkopf D, Waud K. Tubo-ovarian abscess management options for women who desire fertility. Obstet Gynecol Surv 2009;64:681–9, doi:http://dx.doi.org/10.1097/OGX.0b013e3181b8b0d6.
[16] Levenson RB, Pearson KM, Saokar A, Lee SI, Mueller PR, Hahn PF. Image-guided drainage of tuboovarian abscesses of gastrointestinal or genitourinary origin: a retrospective analysis. J Vasc Interv Radiol 2011;22:678–86, doi:http://dx.doi.org/10.1016/j.jvir.2010.10.032.
[17] Garbin O, Verdon R, Fauconnier A. Treatment of the tubo-ovarian abscesses. J Gynecol Obstet Biol Reprod (Paris) 2012;41:875–85, doi:http://dx.doi.org/10.1016/j.jgyn.2012.09.012.
[18] Buchweitz O, Malik E, Kressin P, Meyhoefer-Malik A, Diedrich K. Laparoscopic management of tubo-ovarian abscesses: retrospective analysis of 60 cases. Surg Endosc 2000;14:948–50.
[19] Chen MJ, Yang JH, Yang YS, Ho HN. Increased occurrence of tubo-ovarian abscesses in women with stage III and IV endometriosis. Fertil Steril 2004;82:498–9.