

## Case Report

# Pediatric pyosalpinx without sexually transmitted infection: A report of 3 cases

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#### ABSTRACT

Pelvic inflammatory disease commonly occurs in adults and is most frequently caused by sexually-transmitted organisms. When left untreated, it can progress to abscess formation and subsequent infertility due to tubal scarring. This condition rarely occurs in the pediatric population and even less frequently in the absence of sexual activity. The cases presented here depict 3 cases of pyosalpinx due to noncommunicable infectious agents. Since children are typically not subjected to transvaginal ultrasound, they are particularly at risk for delays in diagnosis and appropriate treatment. Cases described here also demonstrate the value of the pediatric interventional radiology service in treating this gynecological source of infection. Both transabdominal and transrectal approached to ultrasound-guided drainage are described.

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## Introduction

Pelvic inflammatory disease (PID) is an infection of the female genital tract and adjacent peritoneal structures which is classically associated with sexually transmitted infections. These infections typically ascend from the lower genitourinary tract in sexually active females. The most common infectious agents are *Neisseria gonorrhoeae* and *Chlamydia trachomatis*, but anaerobes and other enteric organisms have been implicated when abscesses have formed [1]. Diagnosing and treating PID is important in preventing the complications and long-term sequelae of infection. These include pyosalpinx, hydrosalpinx, tubo-ovarian abscess, infertility, chronic pelvic pain, and increased risk for ectopic pregnancy [2]. The incidence of long-term complications such as infertility has been reported to be 25% [1].

PID occurs primarily in sexually active females, but rare cases of PID in virginal women have been described [3-6]. Many of these cases occur in the setting of pelvic adhesions following surgery or anatomic abnormalities. This population may be especially at risk for the complications of PID since diagnosis can be delayed. To exacerbate the diagnostic difficulty, virginal females are typically not subjected to

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Fig. 1 – Case 1. Axial CT of the pelvis in the first patient reveals a ring-enhancing fluid-filled tubular structure (white arrow) in the right adnexa. An organized abscess (asterisk) is present in the left lower quadrant.

transvaginal ultrasonography which could provide better anatomic detail and increase the sensitivity for detecting early infectious collections in young patients. Transabdominal ultrasound is less sensitive, particularly in patients with increased abdominal wall thickness. Computed tomography (CT) may uncover the diagnosis, but children are also less likely to undergo CT due to radiation-related risks.

Presented here are 3 cases of pediatric pyosalpinx in the absence of sexually-transmitted infections. These cases raise awareness of this pathology which aid in prompt diagnosis and treatment. Likewise, educating pediatric sonographers of this pathology may aid in catching pyosalpinx early, as sonographers in pediatric hospitals are likely to encounter this diagnosis only rarely.

## Case report 1

A 12-year-old virgin female presented with a 5-day history of fever, chills, nausea, and left lower quadrant pain. She had undergone radical vaginectomy at the age of 5 years to resect an embryonal rhabdomyosarcoma. Complex vaginoplasty was also performed using a buccal graft, and she had been undergoing vaginal dilation up until the time of presentation.

CT of the abdomen and pelvis showed a ring-enhancing fluid collection in the left lower quadrant with dilated tubular structures surrounding the collection (Fig. 1). The pediatric interventional radiology service was consulted to drain the fluid. The radiologist identified the collection sonographically and advanced an 18-gauge needle into it through the anterior abdominal wall using ultrasound guidance. A 0.038-inch J-tip guidewire was placed through the needle into the collection, the needle was removed over the wire, and a 10-French pigtail catheter was placed over the wire. The wire was removed, and a sample was obtained for culture. The catheter was connected to a drainage bag that was left to drain via gravity. Culture of the purulent material revealed mixed anaerobic flora including *Bacteroides fragilis* group. No growth of aerobic organisms was uncovered.

## Case report 2

A 12-year-old virgin female was transferred from an outside hospital with a 4-day history of increasing suprapubic abdominal pain. On the first day of the abdominal pain, a clear/white vaginal discharge was present. The patient had not yet had a menstrual period, denied sexual activity, and did not have a boyfriend. Testing for *C. trachomatis* and *N. gonorrhoeae* was negative.

CT of the abdomen and pelvis revealed dilated tubular structures in the bilateral adnexa with an organized abscess in the pelvis (Fig. 2). The pediatric interventional radiology service was consulted to drain the fluid collection. The patient was placed in a dorsal lithotomy position, and an ultrasound probe was placed transrectally. An 18-gauge needle was advanced into the collection, and 30 ml fluid was aspirated. The material was partially complex and partially simple in character. The fluid collection collapsed entirely, and the needle was removed. The culture did not recover bacteria, but it is noted that the patient had been on intravenous metronidazole and ampicillin-sulbactam before the procedure.

#### Case report 3

A 16-year-old female was transferred from an outside hospital with a 7-day history of progressive lower abdominal pain accompanied by subjective fever. Her past medical history included type 2 diabetes mellitus, polycystic ovarian syndrome, and obesity with a body-mass index of 58.96 kg/m<sup>2</sup>. The patient endorsed a single sexual encounter with a male 3 months prior who was also a virgin at the time. Testing for *C. trachomatis*, *N. gonorrhoeae*, reactive plasmin reagin, and human immunodeficiency virus was negative.

CT of the abdomen and pelvis and follow-up pelvic ultrasound demonstrated dilated tubular structures in both adnexa (Fig. 3A). Upon consultation with the pediatric gynecology service, it was determined that her pyosalpinx was due to an ascending infection from normal proliferation of gynecologic flora as a result of poor personal hygiene. She was placed on oral metronidazole, cefoxitin, and doxycycline and discharged.

She presented 2 months following discharge from the hospital with recurrent pain. A subsequent CT resulted in a similar appearance of the pelvis with persistence of bilateral pyosalpinx. Repeated testing for *C. trachomatis* and *N. gonorrhoeae* was again negative. She had been referred to the pediatric interventional radiology service twice, but in both cases, a favorable response to antibiotics precluded sonographically-guided drainage. The patient was eventually managed surgically with bilateral salpingectomies and later underwent a gastric sleeve procedure to manage her obesity.

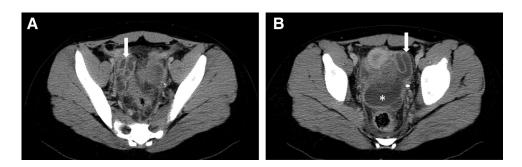


Fig. 2 – Case 2. Axial CT of the pelvis in the second patient demonstrates a ring-enhancing fluid-filled tubular structure (white arrow) in the right adnexa (A) as well as a ring enhancing tubular structure (white arrow) in the left adnexa (B) which is continuous with an organized abscess (asterisk) anterior to the rectum.

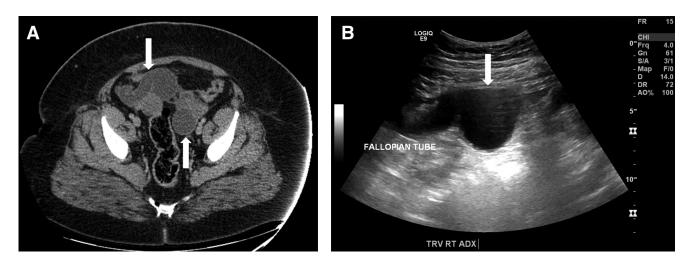


Fig. 3 – Case 3. Axial CT of the pelvis (A) and transabdominal ultrasound (B) in the third patient show dilated fluid-filled structures in both adnexa (white arrows).

## Discussion

The likelihood of encountering pyosalpinx from nonsexually-transmitted infections in female children is relatively low, but the consequences of missing the diagnosis are severe. The cases presented here are rare, but they bring attention to the possibility of pyosalpinx in children from noncommunicable infections.

Postsurgical scarring has been described as a source of virginal hydrosalpinx and pyosalpinx. In a case series of 6 nonsexually active afflicted children described by Cabral and Siqueira [3], 5 of them had undergone pelvic surgery previously. Both cases presented by Moralioğlu et al [4] were secondary to prior pelvic surgery. The history of surgery in the first patient presented here was likely a predisposing element to her gynecologic infection.

Diagnosis of this condition is frequently delayed in children due to a lack of sexual history. Cho et al [5] previously described 5 cases of pelvic inflammatory disease in virginal females, and 3 had been diagnosed as adnexal masses on preoperative imaging. In the case of the second patient presented here, the surgeon specifically noted that PID was unlikely given the patient's lack of sexual activity.

As pediatric obesity increases, pediatric radiologists should consider pyosalpinx as differential considerations for pelvic fluid collections. With pediatric obesity and diabetes on the rise, the third case described here may serve as a harbinger for future cases.

Pyosalpinx is often treated surgically. Surgical treatment was utilized in 4 of the 6 cases reported by Cabral and Siqueira [3], both the cases detailed by Moralioğlu et al [4], all 5 of the cases discussed by Cho et al [5], and both cases described by Kielly and Jamieson [6]. In the cases depicted here, the pediatric interventional service was contacted to drain the fluid. Pediatric interventional radiologists can use cases such as these to demonstrate their value and possibly avoid surgical intervention. Given the fact that surgery is a predisposing factor to pyosalpinx from noncommunicable disease, interventional radiology services may be particularly beneficial to these patients.

The major risks of undergoing pyosalpinx and tuboovarian abscess using image guidance include infection at the site of access, injury to the regional pelvic structures, bleeding, and pain.

## Conclusion

Pyosalpinx unrelated to sexually-transmitted infection is rare but should be considered in nonsexually-active children. Pediatric interventional radiologists may provide value in draining pelvic fluid collections of gynecologic origin.

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