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Case Report

A Pyocolpos, Pyometra, Acute Renal Impairment and Sever Sepsis Caused by Klebsiella Strain in a 14 years old Adolescent Girl with an Imperforate Hymen, A Rare Case Report and a Literature Review

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ABSTRACT

Background: Imperforate Hymen (IH) is considered the most common obstructive anomaly of the female reproductive tract. Infections, endometriosis, subfertility, or obstructive urinary symptoms could be complicated if went undetected. Treatment of uncomplicated IH is simple through hymenotomy (cruciate incision or excision of the hymen). Sepsis is not common to occur secondary to IH, but this case highlights it as a possible and evitable cause of sepsis in pediatrics and adolescents. Pyometra is rarely seen in children and clinical experience in managing this condition is limited. This review reported a rare case scenario with uncommon severe presentations seen in adolescent gynecology, it is a serious case, and fortunately, the pediatricians, emergency room physicians, and gynecologists are rarely facing such an issue. We provided our valuable experiences in the management of imperforate hymen that are complicated with pyocolpos and pyometra and extremely rare and severe sepsis caused by virulent Klebsiella strains in children which is extremely rare to infect them.

Conclusion: Besides the easiness of management of IH, it represents an evitable cause of more complications such as acute urine retention, sepsis, and subfertility. Suspicion should be raised for IH in adolescent girls presented with primary amenorrhea, acute abdomen, urinary manifestations, and emergencies.

Keywords: Imperforate Hymen; Pyocolpos; Pyometra; Sepsis; Hymenotomy; Adolescents; Klebsiella

INTRODUCTION

Imperforate Hymen (IH) occurs approximately in 1/1000 newborn girls and is considered the most common obstructive anomaly of the female reproductive tract. IH is at the extreme of a spectrum of variations in hymeneal configuration. Variations in the embryologic development of the hymen are common and result in fenestrations, septa, bands, micro perforations, anterior displacement, and differences in rigidity and/or elasticity of the hymeneal tissue. This causes accumulation of cervical secretions with subsequent complications such as infections, endometriosis, subfertility, or obstructive urinary symptoms. Sepsis is not documented as a common side effect of IH, but it represents a serious condition and remains a public health issue, with high prevalence, morbidity and mortality rates [1-4].

This review reported a rare case scenario with uncommon severe presentations seen in adolescent gynecology, it is a serious case, and fortunately, the pediatricians, emergency room physicians, and gynecologists are rarely facing such an issue. We provided our valuable experiences in the approaches of diagnosis and treatment for IH that complicated with pyocolpos and pyometra and extremely rare and sever sepsis caused by virulent Klebsiella strains in children which is extremely rare to infect the children. We described the diagnosis and management of such a case in a 14-year-old girl.

CASE REPORT

A 14-year-old virgin girl presented to the emergency room with a bad general condition, abdominal distention, vomiting, fever, and abdominal pain that refer to the back and inability to void urine for >24 hours. Her parents gave a history of gradual onset and progressive course of abdominal distention and pain and they confirmed that she had primary amenorrhea, the menarche did not happen till that day. As regards the rest of the complaints (vomiting >38.8°C, fever, and constipation) were with a hectic onset and progressive course. The parents seek medical help in many public hospitals and the physicians ordered many courses of antibiotics without any noticeable improvement in their child's condition.

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On general examination

She showed sever pallor, disturbed conciseness, weakness, general malaise, and bad general condition, and on abdominal examination showed general abdominal distention with tenderness and muscle guard mainly infra-umbilical, abdominal central dullness, and fair resonant flanks. Sever supra umbilical tenderness with a full bladder that reaches just to or even above the umbilicus, on catheterization pus was obtained as turbid dull yellow colored urine.

Her vitals were temperature 38.8°C, heart rate 110 beats/min, and blood pressure 90/50 mmHg. CBC showed anemia and leucocytosis (Hb 8.5 gm, TLC 20,000), CRP 180 mg/l, urea 123 mg/dl, creatinine 4.8 mg/dl, and her Arterial Blood Gases (ABG) showed metabolic acidosis. Urine analysis showed pus cells over 100/ml, epithelial cells +3, negative virology (HAV, HBV, and HIV), and normal liver functions, these investigations and unfortunately she was in septic shock.

On vaginal examination

Showed imperforate hymen, bulged and whitish fluid behind it (not bluish colored as the usual in such cases of IH which is complicated with hematometra and hematocolpos) [Figures 1 and Figure 2]. She stated 1ry amenorrhea, with cyclic abdominal pain over the last 4 months.

Pelvic abdominal ultrasound

Examination showed distended bladder, hydro ureter and hydronephrosis, enlarged size uterus and intra-cavitary turbid fluid, distended vagina (piccolos), minimal to mild intra-abdominal turbid free fluid as well as in Douglas pouch. The diagnosis of septic shock was made, and the patient started an urgent triple antibiotic regimen to cover (Gram-positive and negative bacteria as well as anaerobes) (Figure 3).

The decision was made by the consultant to do an urgent hymenotomy through a cruciate incision and unfortunately, a gush of the huge amount of pus from the vagina was obtained and foley's catheter was inserted to evacuate the intrauterine fluid (about 1.5 liters of whitish very offensive odor fluid) and pus cultures were obtained and revealed Klebsiella, antibiotics were adjusted to the results.

The patient was followed up in the ICU unit, she continued the antibiotics, and intravenous infusion fluids, and received two whole blood units and one unit of fresh frozen plasma, and inotropic agents were administrated and underwent dialysis for two sessions.

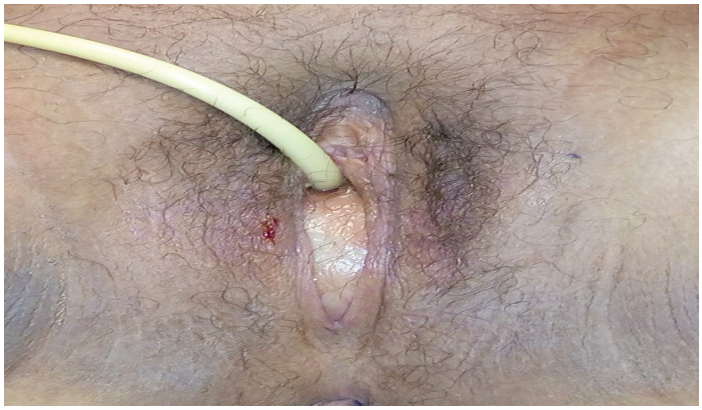

Figure 1: Showing imperforate hymen with unusual whitish bulge due to accumulated pus behind the IH instead of the common bluish bulge which is caused by accumulated retained blood behind the IH.

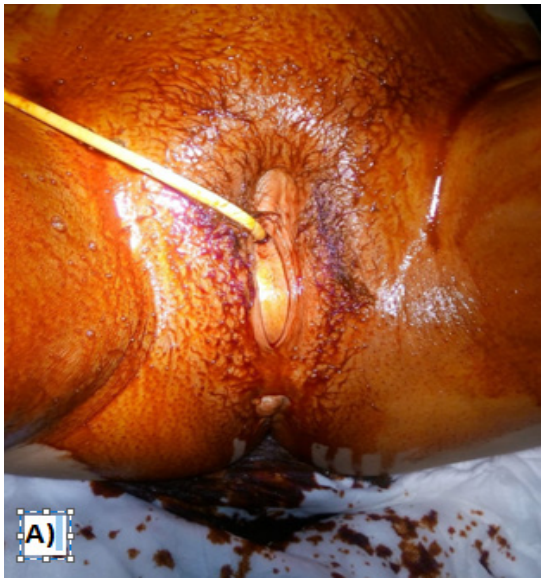

Figure 2: The abdominal distention and infra-umbilical bulge denoting the pyometra.

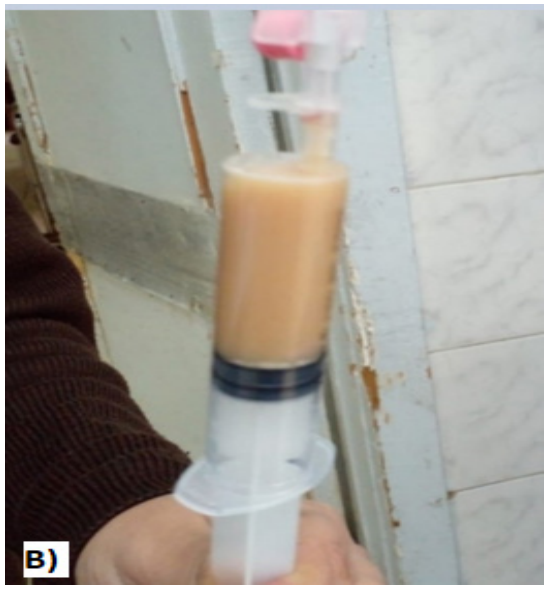

Figure 3: a) Showing the imperforate hymen before the hymenotomy b) The pus which aspirated from pyocolpos.

After 5 days of management and strict observation and follow-up, the renal functions started to normalize with normal urine output.

Pelvi-abdominal u/s were repeated and showed hydroureter and hydronephrosis, and the patient is on medical management in the ward. On follow-up of the case, the urinary manifestations were ceased, and normal temperature and general condition became better. As regards her menstrual cycle, it was hypomenrrhea in the first 3 cycles then became within a normal amount of menstrual bleeding. And she is now under the supervision of pediatric nephrologists and urologists till now.

RESULT

Pediatric and adolescent gynecology is a complex and highly specialized area requiring an in-depth understanding of embryology and adolescent development and requires a multi-disciplinary team of professionals including adolescent gynecologists, pediatricians, specialist nurses, endocrinologists, pediatric surgeons, geneticists, infertility specialists, and psychologists, to both treat the condition

and address the psychological and reproductive sequelae associated with the diagnosis [1].

This can cause a delay in care, especially in urgent or emergent situations. The incidence of congenital abnormalities of the female genital tract is 2%-4%. There are 3 main types:

- Isolated abnormalities, e.g. imperforate hymen or vaginal septum.
- Complex abnormalities of the genital outflow tract involving the cervix, uterus, and fallopian tubes, e.g. unicornuate uterus or uterus didelphys.
- Multi-organ abnormalities. These abnormalities may be related to syndromes or adjacent organs may be involved, e.g. OHVIRA syndrome (Obstructed Hemi-Vagina and the ipsilateral renal anomaly is a rare condition characterized by the triad of uterine didelphys, obstructed Hemi-vagina and ipsilateral renal anomaly.

Consider anatomic anomalies that can be confused with IH in the differential diagnosis as the following

- Acquired labial adhesions.
- Obstructing or partially obstructing vaginal septa (longitudinal or transverse).
- Vaginal cyst or hymenal tag/cyst.
- Hymenal variants as a hymenal band/septum.
- Vaginal agenesis (Mayer-Rokitansky-Kuster-Hauser syndrome) with or without the presence of a uterus or functional endometrium.
- Complete androgen insensitivity syndrome (testicular feminization) [1,2].

Embroidery and pathophysiology

IH is likely the most frequent obstructive anomaly of the female genital tract, but estimates of its frequency vary from 1/1000 population to 1/10,000 population. The frequency incidence of IH at 0.5/1000 women [2, 3].

The hymen is a small mass of mesodermal embryologic remnant. During the embryological period, a hymen is formed from cells urogenital sinus and the sinovaginal bulbs. By the 12th week, the paired müllerian ducts have fused into a single tube (i.e., primitive uterovaginal canal). Two solid invaginations from the distal aspects of the müllerian tubercle form the sinovaginal bulbs (of urogenital sinus origin) or vaginal plate. The initial or cephalad portion of the müllerian ducts forms the fimbria and fallopian tubes; the more distal segment forms the uterus and upper vagina [1,2].

The canalization of the paramesonephric ducts and/or upper vagina joins with the vaginal plate, which canalizes beginning caudally and creates the lower vagina. By the 5th month of gestation, the canalization of the vagina is complete. An imperforate hymen results when this "sheet" of tissue fails to completely canalize. Varying degrees of perforation results in findings such as cribriform or septate hymen [3].

When canalization fails to take place persistence of the septum is clinically represented as IH. This septum obstructs vaginal outflow and results in the accumulation of cervical, vaginal secretions, and menstrual blood as well as leads to hematocolpos and hematometra. IH is at the extreme of variations in hymeneal configuration. Variations in the embryologic development of the hymen are common and result in fenestrations, septa, bands, micro perforations, anterior displacement, and differences in rigidity and/or elasticity of the hymeneal tissue. IH leads to the accumulation of cervical secretions with subsequent complications such as infections, endometriosis, subfertility, or obstructive urinary symptoms. As the mesodermal layer contributes to the development of the kidneys, gonads, and ductal structures, defects or insults in embryologic development may lead to congenital defects of the kidneys or ureters that accompany abnormalities of the vagina and uterus. The lining of the urethra and urinary bladder derives from the endoderm, and the urogenital sinus forms the urethra and vestibule in females. The ectoderm fuses with the endoderm to contribute to the patency and canalization of the genital tract [2,3].

Clinical presentation and investigations

Diagnosis of IH can be made easily by inspection of the external genitalia if an uncertain abdominal ultrasound or by using MRI. IH has been diagnosed with prenatal ultrasound documentation of bladder outlet obstruction due to hydrocolloids or mucoceles. However, despite the recommendations for inspection of the external genitalia during the neonatal and early childhood period, variations in hymeneal anatomy commonly escape diagnosis until the time of menarche. Although IH is a benign congenital condition, late detection and/or improper management may result in severe morbidities such as infections, endometriosis, subfertility or hydronephrosis, obstructive urinary symptoms, and constipation, dysuria, and renal failure in rare cases [1,4,5].

Careful examination with pressure applied to the fourchette may reveal micro perforations, sometimes with an anteriorly displaced opening just beneath the urethra. Some authors described a surgical technique similar to a perineotomy to correct such a defect; however, in asymptomatic patients, waiting until puberty is generally recommended before deciding whether such a technique is necessary [4].

Radiographic imaging must demonstrate that the true diagnosis is not due to an obstructing transverse vaginal septum or another anomaly. Pelvic U/S *via* the transabdominal, trans-perineal, or trans-rectal route is indicated as the initial diagnostic test, followed by MRI if any questions remain about the anatomy. Trans-perineal U/S can help measure the thickness of the septum. As renal and urologic abnormalities are associated with müllerian abnormalities, imaging of the upper urinary tract can help diagnose ipsilateral renal agenesis, duplex collecting systems, and other complex renal anomalies if there are uterovaginal anomalies other than IH. The incidence of renal agenesis is 1/600-1200 persons in patients with müllerian anomalies. As many as 25%-90% of women with renal anomalies are suggested to have concurrent genital anomalies; thus, abdominal and pelvic imaging of these patients is also warranted for these patients [3-6].

Pelvic examination under anesthesia before deciding on appropriate surgical repair of an anomaly that may be more complex than a simple IH, a pelvic examination under anesthesia may aid surgical planning [5].

Typically, the condition is not diagnosed until post-menarchal, when the young woman presents with cyclic abdominal pain, pressure symptoms, and often with an abdominal and/or pelvic

Table 1: Description of Patients.

Author	Year	Country	Patient	Age	Presentation	Preceding Illness	Congenital Abnormality of Uterus, Vagina, or Cervix	Culture
Hospital [6,11]	1948	Zanzibar	A	6 mo	Mass+urine retention	VGastroenteritis	No	*Escherichia coli*
Lwahori [12]	1956	Japan	B	5 mo	Fever+mass	Gastroenteritis	No	NA
Endo [13]	1884	Japan	c	12 y	Pain+anorexia+mass	Anorexia	No	NA
Chandra [14]	1985	India	D	3 y	Fever+mass	Urinary infection	No	NA
Lmamoglu[15]	2005	Turkey	E	12mo	Mass	Urinary infection	Distal vaginal atresia	*E coli*
			F	11mo	Urinary	Urinary infection	Distal vaginal atresia	NA
				9 y (2)	retention	NA	NA	NA
Greggie[16]	2006	U.K.	G	4 mo	Mass	Gastroenteritis	No. Bladder diverticulum	NA
A lgin[17]	2011	Turkey	H	4 mo	Mass	NA	Vaginal atresia	NA
				6 mo	Mass	NA	Proximal vaginal atresia	*E coli*
				4 mo	Fever+mass	Surgery	Cloaca	*Pseudomonas infection*

mass representing a large hematometra and hematocolpos. This morbidity can potentially be avoided if clinicians (e.g., pediatricians, obstetricians, gynecologists, and family physicians) are trained to examine the genitalia of newborns and young children. Timing of the surgical correction could then be planned more appropriately; while this is not a controversial recommendation, implementation is far from universal in clinical practice [3,4].

The hymenal changes that result from nitrogenization (increased elasticity and fimbriation) may reveal the hymen to be open and obviate the need for surgery. Also, surgical procedures to the vagina and hymen during childhood, when endogenous hypo estrogen levels, may lead to scarring and the need for subsequent surgical revision. Surgery during this time should generally be avoided if possible. If the IH is diagnosed during childhood, re-examination should be performed after the onset of estrogen production, which is signaled by breast development. If required, surgery can be performed at this time when healing is optimal and before the accumulation of a hematocolpos [5-10].

An IH presenting after the onset of menstrual shedding is visible upon examination as a translucent thin membrane just inferior to the urethral meatus that bulges with the Valsalva maneuver. This bluish discoloration is due to the presence of a hematocolpos visible behind the translucent hymeneal membrane. Vaginal septa do not typically appear translucent. Depending on the size and volume of the hematometra, hematocolpos, or hematosalpinges, a pelvic or abdominal mass may be palpable during an abdominal or rectal examination. Urinary pressure and even retention, with hydroureter and/or hydronephrosis, may occur due to the mass effect and resultant obstruction. Vaginal and rectal pressure is typically present. Severe constipation and low-back pain are described as presenting symptoms. The labor like menstrual cramps may be severe and cyclic, although the cyclic nature of the symptoms may not be easily or immediately appreciated by the young woman or her family [3, 5, 8].

Unfortunately, the typical findings at diagnosis may include a large collection of blood within the uterus (hematometra) and distensible vagina (hematocolpos), also blood-filled fallopian tubes (hematosalpinges), and signs of retrograde menses, occasionally to the point of intra-abdominal endometriosis and severe pelvic adhesions development [6,7].

Surgical repair after the onset of puberty but before menarche is optimal. This situation is preventable, as routine examinations of the genitalia can detect this obstruction and allow correction before menarche. While these young adolescents typically present to ER with relatively acute pain, this condition should generally not be managed emergently until a definitive diagnosis is made. Defining the anatomy with appropriate imaging techniques and arranging for the most skilled and experienced gynecologist to perform surgery on a scheduled rather than emergent basis is essential. If necessary, menstrual suppression with gonadotropin-releasing hormone (GnRH) analogs can minimize pain pending appropriate imaging and clarification of anatomy. This is more likely to be necessary with complex genital anomalies than with a straightforward IH [3, 6,8].

DISCUSSION

Treatment of uncomplicated IH is simple through hymeneotomy (cruciate incision or excision of the hymen). In the case of patients desiring virginity hymen-preserving surgeries is a choice, such as simple vertical incision and annular hymenotomy. Alternatively, a cruciate incision along the diagonal diameters of the hymen, rather than anterior to posterior, avoids extension injury to the urethra and can be enlarged by the removal of excess hymeneal tissue. In either approach, hemostasis is required using interrupted stitches with fine absorbable sutures [3,5,10]. The presence of endometriosis in young women with obstructing anomalies has been discussed in support of the theory of retrograde menstruation. The endometriosis and pelvic adhesions associated with obstructive anomalies spontaneously resolve once the obstruction is treated [11-19].

The potential risks and benefits of this component of the surgical procedure should be explained to the young woman and her parents to facilitate informed decision-making and consent. A careful surgical technique with the adequate opening of the vaginal orifice prevents stenosis and re-accumulation of the hematocolpos or mucocele, which carries a risk for pelvic inflammatory disease with pyocolpos, pyometra, endometritis, salpingitis, or turbo-ovarian abscess. These have implications and risks for subsequent

infertility, pelvic pain, and ectopic pregnancy [18,20].

Pyometra is the accumulation of purulent material in the uterine cavity and occurs when the cervix is occluded. It is found most commonly in postmenopausal women and its incidence is 0.2% and it is associated with malignancy cervical stenosis. It is less commonly seen in premenopausal women, as a complication of hematomata 2nd to cervical trauma. In children, pyometra is exceedingly rare, and clinical experience in managing this condition is limited. An extensive literature search using PubMed, MEDLINE, and OVID was performed using the keywords pyometra, childhood, and infant; we identified 7 relevant articles, 6 of which are in English and 1 in Japanese (image of the table below), it outlines the pertinent features of each case including the index case. The patients are labeled A-K to facilitate discussion. Two articles reporting patients E, F, H, I, and J describe cases of pyometrocolpos, which is the accumulation of pus in the uterus and upper vagina where the obstruction is not at the level of the cervix but in the vagina. These cases are included because although there is an obstruction to uterine drainage albeit at a slightly lower level, they share some similar features with the other cases. The average age at presentation was 9 months (range from 4 months to 3 years) excluding patient C who was a 12-year-old with anorexia nervosa.

Patient F had 2 presentations, the 1st occurring at 11 months of age and the 2nd at 9 years. We have no other information about the first presentation except that vaginal atresia was diagnosed and an angioplasty was performed [8].

Severe sepsis or septic shock in children is associated with high mortality, especially in developing countries, and accounts for about 8% of pediatric intensive care units with estimated mortalities of about 25%>17% of survivors may show moderate disabilities. Globally, the most frequent sources of infection are respiratory (40%) and blood (19%). Common treatments include mechanical ventilation, vasoactive infusions, and corticosteroids [7,8].

Pyometra occurs rarely in childhood and a review of cases hypothesizes that causation may relate to a hypo-estrogens endometrium, cervical stenosis, and exposure to transient bacteremia. In adult women, the role of ovarian steroids in endometrial protection against ascending infection is established but this hormonal protection is not available to an infant. Functional or anatomical obstruction to uterine drainage may facilitate the development of a pyometra in these circumstances [10,18].

As we mentioned in our extremely rare case report, the bacteriological analysis of pus drained from the female child's vagina and uterus revealed Klebsiella. After long research about such a case, it is clear that in the first case report as regards IH and pyometra in the young child, furthermore the causative organism was extremely rare and this case is considered to be a new clinical presentation of the Klebsiella group of bacteria's. We provided our valuable experiences in the approaches of diagnosis and treatment for IH that complicated with pyocolpos and pyometra and extremely rare and sever sepsis caused by virulent Klebsiella strains in children. We described the diagnosis and management of such a rare complicated case in a 14-year-old girl.

Klebsiella species cause a wide range of diseases including pneumonia, Urinary Tract Infections (UTIs), bloodstream infections, and sepsis. It is a bacterium that normally lives inside the human intestines, where it doesn't cause disease. These infections are particularly a problem among neonates, the elderly, and immunocompromised individuals. It is also responsible for a significant number of community-acquired infections and their morbidity and mortality [19].

K. pneumoniae has become of increasing concern in the clinical environment over the last 20 years. The isolation of multiple drug-resistant strains to cause nosocomial infections and the relatively rapid spread of this resistance phenotype among strains have made the treatment and management of these types of infections difficult and the possibility of systemic dissemination. Infections of medical devices such as urinary catheters are a major site of K. pneumoniae infections and have been suggested to involve the formation of biofilms on these surfaces [19-21].

Klebsiella may disseminate to other tissues causing life-threatening infections including pneumonia, UTIs, bloodstream infections, and sepsis. It associated with these infections is regarded as hypervirulent, and recent studies indicate that these strains share specific genetic characteristics. It is gaining attention due to the rise in the number of infections and the increasing number of strains resistant to antibiotics [19-24].

More than a third of the K. pneumoniae isolates reported to the CDC were resistant to at least one antimicrobial group, the combined resistance to fluoroquinolones, 3rd -generation cephalosporins, and aminoglycosides being the most common resistance phenotype. The symptoms differ depending on the site of the infection. Bloodstream infections (bacteremia and sepsis) from klebsiella can cause fever, chills, fatigue, light-headedness, and altered mental states. Risk factors associated with Klebsiella-UTI are; Diabetes mellitus- urinary-tract obstruction -chronic renal insufficiency, immunosuppression and catheterization [20, 22, 23].

Another factor regarding the growth of K. pneumoniae in the urinary tract is the ability of the bacteria to internalize within epithelial cells and avoid host defense mechanisms. The evolution of K. pneumoniae strains producing factors that mediate rapid dissemination from the urinary tract and subsequent tissue injury and destruction may be a possibility [18-24].

CONCLUSION

There is much to be learned about how Klebsiella disseminates from the 1st infection site, either the lung or the gut, to other sites. One troubling aspect of K. pneumoniae infections is the emergence of strains causing disseminated pyogenic infections. Challenges that face the future management of these infections include the development of non-antibiotic-based therapies since the ability of K. pneumonia to rapidly evolve to antibiotic-resistant strains is alarming. The prevention of infection and management of patients with infections will provide enormous challenges in the future.

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