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AN UNUSUAL CULPRIT: A CASE REPORT OF XANTHOGRANULOMATOUS INFLAMMATION OF URACHUS IN AN ELDERLY WOMAN

General Surgery

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ABSTRACT

Background: The majority of adult patients (less than 2/300,000) have urachal abnormalities. A common challenge in diagnosis is that infected urachal cysts may look like either bladder cancer or abscesses. **Case Presentation:** We are reporting the case of a 55 year old diabetic woman who was experiencing a prolonged history of fever and urinary tract symptoms for 15 days. In addition she had experienced suprapubic pain, midline swelling, and obstructive urinary symptoms. On physical exam she had a tender 4x3 cm mass in her midline suprapubic area. When she was catheterized, the catheter drained purulent material and resolved the mass. She underwent ultrasonography that showed a 7.7x4x4 cm, irregularly shaped, heterogeneous hypoechoic mass in the midline infraumbilical peritoneal cavity. It was attached to the bladder fundus. She also underwent CT scan after intravenous contrast that found an ill-defined 4x4.8x7.3 cm soft tissue density between the umbilicus and pubis, adjacent to the dome of the urinary bladder, with loss of fat planes to the anterior abdominal wall. Cystoscopy showed an opening at the fundus of the bladder that was draining purulent material with contrast leaking out of the opening into the abdominal cavity. Following treatment with antibiotics to treat her infection, the definitive treatment of the urachal cyst was performed via an open approach with a partial cystectomy. Her histological findings were xanthogranulomatous inflammation without evidence of malignancy or atypia. The patient made an uneventful recovery and was able to have her catheter removed on postop day 7. **Conclusion:** Infected urachal cysts should be considered in adults who are having difficulty urinating and/or have a mass in their abdomen. The best way to prevent further complications or malignant transformation from occurring is to perform staged treatment consisting of treating the infection first and then removing the infected urachal cyst.

KEYWORDS

Urachus; Urachal Cyst; Xanthogranulomatous Inflammation; Lower Urinary Tract Symptoms; Partial Cystectomy

INTRODUCTION

Embryologically, the urachus is derived from remnants of the allantois that usually obliterate completely as the foetus develops. In this process the urachus forms the median umbilical ligament that extends from the dome of the urinary bladder to the umbilicus[1]. If the urachus fails to be obliterated completely, then a variety of urachal anomalies may result. These include: patent urachus, urachal cyst, urachal sinus, and vesicourachal diverticulum[2]. Although most urachal anomalies will manifest early in life; some may go undiagnosed until later in life and have complications of either infection, rupture, or malignant degeneration[3].

Adult urachal anomalies occur infrequently, with an estimated occurrence of < 2/300,000[4]. Approximately one-third of urachal anomalies in adults are infected urachal cysts[5] and therefore are difficult to diagnose due to their ability to mimic bladder cancer, bladder abscesses or other intra-abdominal conditions[5]. Presenting complaints may include: abdominal pain, a palpable mass, lower urinary tract symptoms or fever [6].

Diabetes mellitus is well recognized as a risk factor for developing an infected urachal cyst since diabetes increases the patient's susceptibility to infections[7]. A high level of clinical suspicion in combination with the use of imaging studies (ultrasound and/or CT with contrast) should aid in making the diagnosis[8]. Treatment generally consists of a two-stage procedure: antibiotics to manage the acute infection initially, followed by surgery to remove the cyst and eliminate the risk of future recurrence and malignant degeneration[9-10].

This paper presents a case of an infected urachal cyst in a 55 year old diabetic female who was presenting with a suprapubic swelling and lower urinary tract symptoms, and will outline our approach to diagnosing and managing her condition as well as the outcome of treatment.

Case Presentation

The patient is a 55-year-old woman diagnosed with type 2 diabetes mellitus who came into the Urology Department of our hospital due to complaints of right supra-pubic pain and a palpable mass in the midline supra-pubic area that developed over the past 15 days. The patient experienced both obstructive and irritative urinary symptoms, such as difficulty initiating urination, poor urine flow, an increase in the number of times she urinated daily, the urge to urinate frequently, and night-time urination. The patient also reported having intermittent fevers over the past 15 days.

The patient's physical examination included a soft abdomen with a suprapubic midline, tender and palpable mass measuring about 4x3cm. The mass was firm and non-movable and was very tender on palpation. The patient had no indication of peritonitis present. All other parts of the patient's system were normal.

Laboratory results indicated an elevation in white blood cells with high levels of inflammation markers. Due to the patient's LUTS and the large amount of residual urine after urinating, the decision was made to place a Foley catheter, which produced a quick release of pus and significant decrease in the size of the palpable mass.

Ultrasound of the abdomen and pelvic region showed an irregularly-shaped, 7.7 x 4 x 4 cm heterogenous hypoechoic mass in the middle infraumbilical peritoneal cavity that was attached to the superior aspect of the urinary bladder and a large residual urine volume. The ultrasound images of the mass contained internal echoes that suggested complex fluid content.

Contrast Enhanced Computed Tomography (CECT) of the abdomen and pelvic regions revealed an ill-defined soft tissue density area of 4 x 4.8 x 7.3 cm located between the umbilicus and pubis, directly adjacent to the dome of the urinary bladder. The CT scans show loss of the fat planes between the mass and the anterior abdominal wall indicating inflammatory changes and adhesions. The CT scan images are indicative of an infected urachal cyst with possible bladder fistula.

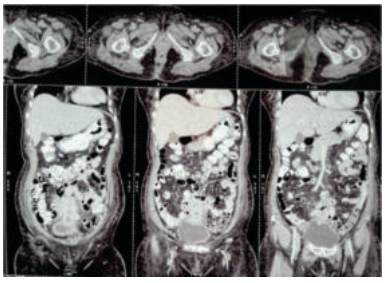

Image 1

A cystoscopy was performed in order to evaluate the bladder's potential involvement and identified a tract with purulent discharge from its fundus that communicated with the abdominal cavity as confirmed by contrast injection through the opening. The remainder of the bladder mucosa had no abnormalities indicative of malignancy and thus seemed normal.

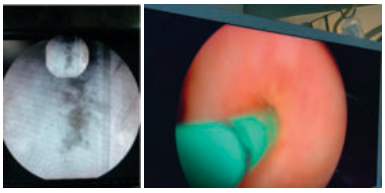

Image 2

Management

The initial treatment involved giving the patient empirically administered intravenous broad spectrum antibiotics that were based on the local antibiotic resistance pattern, along with continuous urinary drainage by means of a foley catheter.

This conservative approach was meant to manage the acute infection and decrease inflammation prior to further surgical interventions to definitively treat the condition.

Within one week of this treatment the patient began to show clinical improvement as evidenced by a resolution of her fever and decreased levels of inflammatory marker levels.

Following adequate control of the patient's infection, the patient underwent definitive surgical management. The patient underwent a partial cystectomy and an open excision of a urachal cyst while under general anesthesia. During the surgery, an infected urachal remnant that extended from the bladder dome to the umbilicus, which was densely adherent to the anterior abdominal wall, was found intraoperatively. An en bloc excision of the urachal cyst and bladder cuff was done. The bladder was then closed in two layers and a drain was placed in the prevesical space. A foley catheter was left in place for continued urinary drainage post operatively.

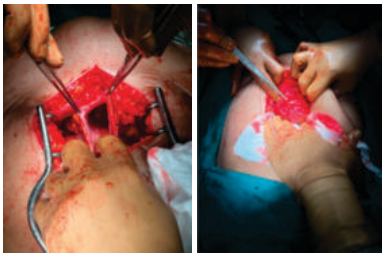

Image 3

Specimen gross measurements were; (9 × 5 × 2) cm, which represented the Urachal Cyst, Urachus and bladder cuff. Purulent fluid filled the cyst. Microscopic evaluation of this specimen demonstrated histiocytic cells that were foamy along with a lymphocytic infiltrate and a giant cell response. This microscopic appearance is consistent with xanthogranulomatous inflammation of the urachus. Cellular atypia or malignant cells were not found in multiple sections examined.

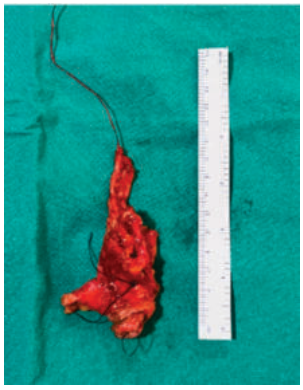

Image 4

The post-operative period progressed as expected. There were no post-operative complications to speak of. A drain was removed from the patient by the third post-operative day and a Foley catheter was removed on the seventh post-operative day after a cystogram confirmed there was no leakage. The patient returned home with recommendations regarding follow-up and experienced full recovery with normal voiding function.

DISCUSSION

Incomplete obliteration of the allantois during embryogenesis results in abnormal formation of the urachus. Normally, involution of the urachus occurs between the 4th to 5th months of gestation to create a fibrous band called the median umbilical ligament[1,2]. Failure to obliterate completely may lead to 4 forms of urachal anomaly; a patent urachus (50%), urachal cyst (30%), urachal sinus (15%), and vesicourachal diverticulum (5%)[11]. These anomalies typically manifest in children, however urachal cysts may be asymptomatic until adult years when they may become complicated by infection, rupture, or malignant changes[3,12].

The incidence of urachal anomalies in adults is extremely low, occurring in < 2/300,000 people[4]. Approximately one-third of symptomatic urachal pathologies in adults are due to infected urachal cysts[5]. Diabetes mellitus, immunosuppression, trauma, and urinary tract infections are several risk factors that increase the likelihood of developing a urachal cyst infection[7,13]. It is likely that the patient's diabetic condition contributed to the development of an infection.

The clinical manifestations of infected urachal cysts are often non-specific and can mimic other conditions, such as bladder cancer, bladder abscesses, infected remnants of the urachus, or intra-abdominal abscesses[5,6]. Symptoms commonly reported by patients with infected urachal cysts include: lower abdominal pain, lower urinary tract symptoms (obstructive and irritative), fever, and possibly umbilical discharge[6,14]. The patient in our case had symptoms consistent with a classic triad of lower abdominal pain, palpable mass, lower urinary tract symptoms, and fever that led to the suspicion of an infected urachal cyst.

Imaging studies are critical for diagnosing urachal pathology. Ultrasonography is the most commonly used initial imaging study to identify a midline cystic or complex mass in the space of Retzius located between the bladder dome and the anterior abdominal wall[8]. Contrast-enhanced CT is superior in identifying the spatial anatomy of the mass relative to the surrounding structures, specifically the bladder dome and the umbilicus[8,15]. Imaging studies in our case provided clear visualization of the mass characteristics and the close relationship of the mass with the bladder fundus, which were key elements in determining the surgical plan.

Cystoscopy is a valuable tool in the evaluation of infected urachal cysts. Cystoscopy allows the examiner to confirm communication with the bladder and evaluate for intravesical extension of the mass or associated bladder pathology[9]. The findings of cystoscopy in our patient supported the diagnosis of an infected urachal cyst with bladder communication. Furthermore, cystoscopy helps differentiate bladder malignancies, which are an important differential diagnosis in similar presentations[10].

Management of infected urachal cysts involves a multi-stage process. The first stage of management focuses on controlling the infection with broad spectrum antibiotic therapy and adequate drainage. Drainage can occur through spontaneous drainage through the bladder, as seen with catheterization in our patient, or through percutaneous drainage if necessary[9,13]. The conservative phase of management will allow for resolution of acute inflammation and make subsequent surgical intervention safer and easier to perform. Percutaneous drainage has been proposed as an initial temporizing measure in severely infected cases[13].

Surgical intervention is required to remove the entire urachal remnant, along with a portion of the bladder, to ensure complete eradication of the disease and prevent recurrence[10,14]. Partial excision of the urachal remnant is associated with a higher rate of recurrence and persistent symptoms[14]. Surgical interventions have been performed using both open and laparoscopic approaches, with equivalent outcomes[15]. An open approach was used in our case, due to the significant amount of inflammation and adhesions to the anterior abdominal wall. The en bloc excision of the urachal remnant with partial cystectomy ensured complete removal of the disease.

Histopathologic evaluation of the removed tissue is mandatory to assess for malignancy, as urachal adenocarcinoma can occur in approximately 0.01% of bladder cancers and is more common in adults with urachal anomalies[12]. Histopathologic analysis in our case revealed xanthogranulomatous inflammation, a rare type of chronic inflammation consisting of clusters of foamy histiocytes, lymphocytes, and giant cells. Xanthogranulomatous inflammation can occur in many organs, including the kidney and bladder, and is typically associated with chronic infection. The lack of atypia or malignancy in the histopathologic sample was reassuring and confirmed the benign nature of the lesion.

Complete excision of the urachal remnant with a bladder cuff is the gold standard of treatment for infected urachal cysts, as it eliminates the risk of recurrence and malignant transformation, and provides tissue for definitive histopathologic diagnosis[10,14]. Long-term follow-up is recommended to detect recurrence, but the risk is low after complete excision[15].

CONCLUSION

Although rarely seen in adult populations, infected urachal cysts should always be considered in differential diagnoses for patients that present with suprapubic mass formations or other manifestations associated with lower urinary tract disease, especially in diabetic individuals who have an increased risk. In order to obtain a precise diagnosis, it will be important to combine an elevated index of clinical suspicion with a combination of diagnostic imaging modalities, i.e., ultrasonography and/or enhanced CT scans of the abdomen/pelvis. While cystoscopy is also useful for establishing a bladder connection (urachus) to exclude malignant disease processes, this procedure should only be performed after histologic confirmation has been obtained.

The best method to manage infected urachal cysts involves a two-staged process beginning with initial conservative treatment of the infection, typically consisting of antibiotics and percutaneous drainage to treat acute infection and reduce pressure within the cyst. Following resolution of the infection, a definitive surgical intervention must occur and must involve total excision of all remaining urachal tissue with removal of a portion of the bladder (cuff). This allows the surgeon to remove all potential sources of future infections and malignancies. Additionally, histopathological examination of any resected tissues provides confirmation of the diagnosis and rules out malignant changes in the involved tissue(s). The results from this type of surgical intervention provide excellent patient outcomes as evidenced in this report where the patient underwent complete recovery without any post-operative complications.

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