Case Report

Para-urethral cyst in a female neonate: what pediatricians must know?

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ABSTRACT

Para-urethral cyst, arising from cystic dilatation of a paraurethral gland in a female, is rarely reported in neonatal age group. The low frequency and little awareness about its clinical course may lead to inappropriate management. The choice of treatment for this lesion still remains controversial. Surgical excision has been advocated, but non-operative treatment is also gaining popularity nowadays. We are hereby reporting a symptomatic female neonate, whose paraurethral cyst was managed surgically with good recovery. The purpose of this report is to clarify the natural course of paraurethral cysts in female neonates and to generate awareness among physicians and surgeons for correct diagnosis and proper management. The embryology, clinical features and various treatment options are discussed along with the management of our case.

Keywords: Neonate, Paraurethral cyst, Skene's duct cyst

INTRODUCTION

Para urethral cyst, arising from cystic dilatation of a paraurethral gland in a female, is a rare entity particularly in the neonatal age group. The exact etiology of paraurethral cysts is unknown in neonates unlike adults where these cysts result mostly due to inflammation.¹ Only 49 cases have been reported previously in the English literature. These congenital cysts may be diagnosed by vigilant inspection in the initial physical examination of the new-borne. Clinically, these cysts are typically round bulging structures of orange or yellow colour and causing distortion of urethra. Radiological evaluation is helpful for differential diagnosis and in determining the proximal extension of the cystic pathology. The choice of management of this lesion is controversial and there is no consensus on treatment as yet. Surgical management has been advocated off late but spontaneous regression with conservative management has also been supported recently. However, in some

instances spontaneous resolution has not occurred despite long observation period. The lesion although rare can present to any pediatrician in neonatal age group. It is therefore imperative to understand the embryology and clinical features in order to select correct modality of treatment. Pertinent literature regarding embryology and management are discussed herewith along with a case report.

CASE REPORT

A 2-day old female neonate following an uneventful spontaneous delivery at term was referred to us for not passing urine since birth. There were no associated anomalies. Clinical examination was not remarkable with normal anal, vaginal and urethral openings present separately along with mildly palpable bladder. The patient had 1.5 x 1.5 cm, interlabial spherical, whitish cystic swelling obscuring both urethral and vaginal orifice (Figure 1).

Figure 1: Para-urethral cyst in female neonate.

Patient was catheterized with 6 Fr feeding tube and urine drained freely (Figure 2). Vaginal patency was also verified. Laboratory investigations included a complete blood count, renal function tests and urinalysis. All laboratory results were normal.

Figure 2: Catheterisation of urethra and adjacent compressing cyst.

Figure 3: Lesion after de-roofing.

Renal and pelvic ultrasounds revealed an echo dense avascular nodule located between anteriorly displaced urethra and anterior part of distal vagina. There was no evidence of hydrocolpos, outflow obstruction or urinary tract infection. Kidneys and bladder were normal. There was no evidence of pelvic abnormalities, renal pelvic

dilatation, or urethral duplication. Considering the symptomatic nature of the lesion and difficult follow up (the patient belonged to remote tribal region) the incision and de-roofing of cyst was performed (Figure 3). Histology revealed that cyst was lined with urothelium. The subsequent course was uneventful. Follow-up examination was normal with complete resolution. The patient has had no recurrence to date.

DISCUSSION

Paraurethral glands and ducts form, as an outpouching of the urethra, during the third trimester. They are rudimentary homologues of the male prostate gland and empty into the female urethra with their own ducts.¹ Huffman reported the presence of 6 to 30 paraurethral ducts, the largest two of which were referred to as Skene's ducts.^{2,3} Skene's ducts secrete a small amount of mucoid material with sexual stimulation and lubricate the urethral meatus during coitus.² Skene, described significance of these ducts and discussed the possibility of cyst formation due to ductal obstruction.^{4,5}

Paraurethral cysts are uncommon in any age group, particularly amongst the newborns.¹ The reported incidence of paraurethral cysts is between 1 in 2074 and 1 in 7246 live female births still only 49 cases have been reported in the English literature.^{1,5,6} Most of the paraurethral cysts are unilateral and are more common on the right side.⁵ The characteristic appearance is a unilateral, bulging mass with a whitish color which is either oval or round in shape, 1-2 cm in diameter and has small vessels on their surface.¹

The paucity of data suggests that the lesion is under recognized. The low frequency of the Skene's duct cyst and the little awareness of it and its benignity may lead to wrong diagnosis that underestimates its incidence. Hence, detailed examination of female newborn is of paramount importance and should be a routine practice.

The etiology of paraurethral cyst in female neonates is not fully understood, however, the histological appearance of the cyst epithelium which is usually urothelium suggests its embryonic origin. One of the accepted theories is that dislocation of the urothelium from sinus urogenitalis into an adjacent area of inflammation leads to blockage of Skene's duct resulting in paraurethral cyst formation. It has also been postulated that maternally acquired female hormones may provoke glandular secretion and cause cyst formation in newborns.^{6,7} The presence of paraurethral cysts and the exposure to maternal estrogens in the early neonatal period has also been proposed.⁶

Paraurethral cysts are frequently asymptomatic and are discovered during routine neonatal genital examination. Identification of urethral meatus separate from the mass and laterally deviated urine outflow are highly suggestive. Rarely, they present with pain, vaginal

discharge, or visible displacement of the urethral meatus.^{1,2} Recently, two cases have been reported with vaginal bleeding and breast enlargement owing to maternal hormone stimulation.

Paraurethral cyst must be differentiated from other interlabial masses with similar presentation like urethrocele, imperforate hymen, Gardner's duct cyst, vaginal inclusion cyst, urethral diverticulum, ectopic ureter, vaginal agenesis, or neoplasia (rhabdomyosarcoma). Previously, complete urologic evaluation was recommended for all cases. But current experience recommends ultrasonography (high resolution) of abdomen and perineum for all cases as the investigation of choice.

The management of this lesion is controversial with lack of proper guidelines. Since, paraurethral cysts are generally asymptomatic and have a high rate of spontaneous resolution hence an initial conservative approach is advised. However, the conservative management warrants strict and regular follow up. The duration of conservative management is also not precisely mentioned. While conservative treatment is also appropriate in many circumstance, urinary obstruction requires treatment for resolution.^{8,9} Current experience suggests that spontaneous rupture of the lesion in neonates is unlikely after 2 months of age and hence surgical intervention in the form of needle aspiration, marsupialisation or excision should be considered after this time.⁵ Needle aspiration can be attempted in symptomatic infant but has higher rate of recurrence. Surgical excision and marsupialization should be upfront treatment for infants and children with urinary obstruction or other manifestations.⁹ Patients in whom conservative management is not successful or feasible should also be treated surgically. For older children and adolescents also surgical excision and marsupialisation is the preferred mode of treatment.⁹ The advantages of surgical excision and marsupialisation include quick resolution of symptoms, early healing and no need for long follow up, in addition to obtaining tissue for histopathological analysis.

CONCLUSION

In summary, paraurethral or Skene's duct cysts are rare introital masses in neonates. Maternal estrogens should

be considered a potential factor in the development of these cysts. Awareness among physicians and surgeons is imperative for proper identification and management. Conservative treatment is an appropriate option in the asymptomatic neonate but surgery is preferred in infants and neonates who are symptomatic and less likely to come for follow up as also for older children and adolescents.

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