Case Report

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Torsion of an epididymal cyst in a patient of androgen insensitivity: a rare case report

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ABSTRACT

Torsion of an epididymal cyst in a patient of androgen insensitivity is an extremely rare presentation. No single case is reported till date which adds to its rarity. We present here such a bizarre and certainly an interesting case report of a 45 years old female patient came with complaints of pain and swelling in left inguinal region since five days. Pain increased in severity in last 3 days without any relief. A single globular tender swelling 6x6 cm size was noted with no cough impulse. With a diagnosis of obstructed hernia the patient was shifted for USG of abdomen which reveals a surprise to all attending surgeons. On USG bilateral cystic swelling were noted with no uterus and cervix. The cystic swelling on left side showed hypoechogenecity. A diagnosis of Mullerian duct agenesis was made with torsion in the left sided cystic swelling. Decision for immediate exploration was taken. On exploration a gangrenous torsion of left epididymal cyst was seen with normal testes. The diagnosis was immediately changed with androgen insensitivity after noting the presence of fully developed testes in this female. Left inguinal orchidectomy was performed in patient with reconstruction of inguinal floor. The post-operative period was uneventful. Patient was discharged on day 7. In the highly advanced robotic age where we exclaim that we know extreme and have explored science to its limits this simple and strange presentation reveals the bizarre and vivid reality of nature. It only cumulates the unseen probabilities of human biology.

Keywords: Tortion, Epididymal cyst, Androgen insensitivity

INTRODUCTION

Cysts of the epididymis are very common and almost without exception are benign. Swelling is usually located in the upper rear part of the testicle because the epididymis lies behind the testis. The cysts are almost always painless and, because most of these cysts are asymptomatic, treatment is usually not necessary. Torsion of epididymal cyst in patient of androgen insensitivity is extremely rare condition.

CASE REPORT

A 45 year old female patient came to hospital with complaint of pain and swelling in left inguinal region since five days. Patient had fever since three days. Pain aggravated in last three days. On clinical examination there was single globular swelling in left inguinal region around 6x6 cm with indurated overlying skin. Swelling was tender, irreducible with absent cough impulse, clinically appeared as obstructed inguinal hernia. On investigation there was leucocytosis as the only positive

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finding from blood indices. X-ray abdomen was within normal limits. On USG abdomen there were cystic swellings on both inguinal region with signs of inflammation on left side with pericystic collection with absent uterus and cervix. Patient was diagnosed as a case of Mullerian duct agenesis with absent uterus and cervix with normal vaginal canal. Considering the condition of patient and proper fitness from anaesthetic point of view patient was posted for explorative laparotomy.



Figure 1: Torsion of epidydimal cyst with testis in left inguinal canal.



Figure 2: Gangrenous epidydimal cyst with tortion with normal testis.

On exploration a normal left testes (later confirmed to be normal on histopathology) was seen with a gangrenous torsion of epididymal cystic appendage was noted (Figure 1 and 2). A decision for performing a left sided inguinal orchidectomy with excision of cyst was taken and also an inguinal canal repair with prolene no 1 was performed. Postoperative period was uneventful. The presence of testes in a female with full pubertal characteristics of female once again changed our diagnosis into androgen insensitivity syndrome. The patient was explained about the genetic disease she possessed and was warned for further consequences of the condition. We explained her that she possessed testes gonad (confirmed on histology) and is suffering from androgen insensitivity. We also explained she need to further undergo karyotyping for her

disease and also undergo other side orchidectomy for preventing future risk of malignancy and its consequences. It was difficult to explain that she is a genetic male with clinical phenotype female. Finally with all possible valiant efforts, patient was not at all convinced and she did not comply for further studies and operative interventions. Hence patient was finally discharged on post-operative day 7.

DISCUSSION

Acute scrotal swelling is a potential urologic emergency and requires urgent evaluation and emergency surgical management. Many conditions might present with acute scrotal swelling. These include spermatic cord torsion, torsion of a testis and epididymal appendage, epididymitis, orchitis, hydrocele, hernia, varicocele, trauma, tumor, idiopathic scrotal edema and Henoch-Schönlein purpura.¹ Cysts of the epididymis are very common and almost without exception are benign. These are fluid-filled swellings that develop in one or more of the many tubes leading from the testis to the epididymis. Epididymal cysts are usually acquired rather than congenital, which is why they typically occur in older men. Torsion of a mass in the epididymal head is described by Hedelin.2 To our knowledge, torsion of epididymal cyst is a very rare condition and only one other case has been reported previously.3 Androgen insensitivity is a genetic disorder with the gonads as testes which produce normal androgens. The lack of androgen receptors results in sparse hair or no hair in pubic and axillary region. Patients usually have normal breasts because of peripheral conversion of androgens to estrogens. They may have a short vagina or normal full length vagina and no cervix and uterus due to production of Mullerian inhibiting substance by testes. In pubertal females a diagnosis between androgen insensitivity and Mullerian agenesis can be made by karyotyping and measuring serum testosterone levels. 4-8 The acute pain and torsion of the epididymal cyst in patient of androgen insensitivity, reported in the present study is the only case of its kind to be reported in the literature.

CONCLUSION

Rare diagnoses are seen rarely and are correct rarely is known from decades. We not only know that but can say we actually have seen that. In this highly advanced robotic age where we exclaim that we know extreme and have explored science to its limits this simple and strange presentation reveals the bizarre and vivid reality of nature. It only cumulates the unseen probabilities of human biology.

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