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Zinner's Syndrome: A Confusing Diagnosis in the Face of Chronic Disabling Perineoscrotal Pain in the Young Subject, about a Case

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Abstract

Zinner syndrome is a rare congenital malformation related to an abnormality in the development of the Wolffian duct, the clinical picture of which consists of a triad of unilateral renal agenesis, ipsilateral seminal vesicle cyst, and obstruction of the ejaculatory duct. Chronic perineoscrotal pain may be an indication of the diagnosis of Zinner syndrome to some extent. We report the observation of a 23-year-old patient, married and father of one child, who presented with chronic perineo-scrotal pain revealing on morphological assessment (ultrasound, uroscanner, prostatic MRI) a cystic formation of the seminal vesicle, left renal agenesis with an empty left renal compartment. Given the patient's refusal of any surgical procedure, treatment consisted of echo-guided puncture of the seminal vesicle cyst bringing back 30 cc of a seroviscous liquid whose analysis revealed spermatozoa, without atypical cells, compatible with a cyst. The clinical evolution was marked by a progressive remission of the scrotal pain with a delay of several months.

Keywords

Agenesis, Congenital Malformation, Kidney, Perineoscrotal Pain, Seminal Vesicles, Zinner's Syndrome

1. Introduction

Zinner's syndrome is a rare congenital disorder related to an abnormality in the development of the Wolff duct and consists of a triad of unilateral renal agenesis, ipsilateral seminal vesicles, cyst, and obstruction of the ejaculatory duct.

After its first description by Dr. Zinner slightly more than one hundred years ago (in 1914), about 150 to 200 cases have been published in the literature. Zinner's syndrome is usually detected in the third to fourth decade of life but can be detected earlier with the increasing use of CT and MRI scans [1].

We compiled a report based on a new case of chronic disabling perineoscrotal pain in a 23-year-old man.

2. Patient and Observation

This is a 23-year-old patient, married and father of one child, who presents with a chronic perineo-scrotal pain evolving for 4 years, of a heavy type, disabling and preventing any prolonged standing. This pain was not accompanied by any associated urinary problems, haemospermia or pain during sexual intercourse. Initially diagnosed as a varicocele, the patient underwent surgical treatment before being referred to our department due to the persistence of the symptoms.

The clinical examination revealed a patient in good general condition, the abdomen and external genitalia were normal, with pain provoked by palpation of the left lobe of the prostate.

Abdominal and pelvic ultrasound revealed a 22 mm left retrovesical cystic formation with a thin wall, a prostate of normal volume and appearance, and an empty left renal space (**Figure 1**).

The uroscanner confirmed the cyst of the seminal vesicle measuring 25 × 35 mm lateralized to the left, associated with left renal agenesis and a compensatory right renal hypertrophy (**Figure 2**). The prostatic MRI supplement allowed an accurate analysis of the relationship with neighbouring structures (**Figure 3**).

After discussion with the patient and his refusal of any surgical procedure, and given the highly symptomatic nature of the cyst, the treatment consisted of an echo-guided evacuation of the cyst bringing back about 30cc of a sero-viscous fluid, the cytological analysis of which revealed the presence of spermatozoa, without atypical cells, compatible with a cyst of the seminal vesicle.

The evolution marked by a regression of the symptomatology with a delay of several months.

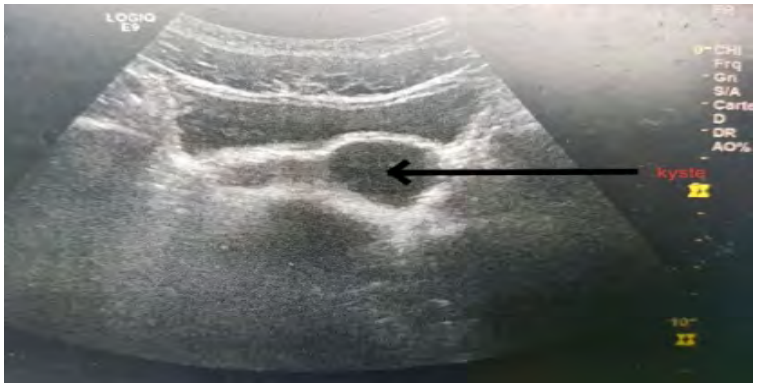

Figure 1. Abdominal-pelvic ultrasound showing a 22 mm left retrovesical cystic formation.

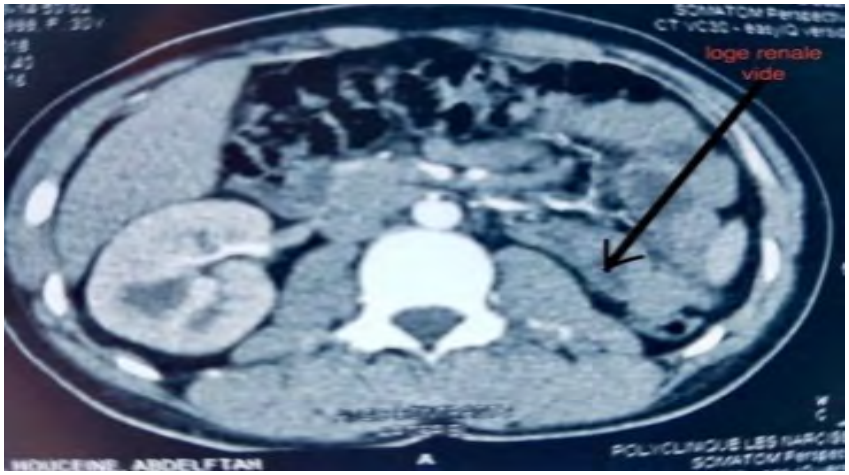

Figure 2. Uroscanner shows a hypodense retrovesical mass on the left.

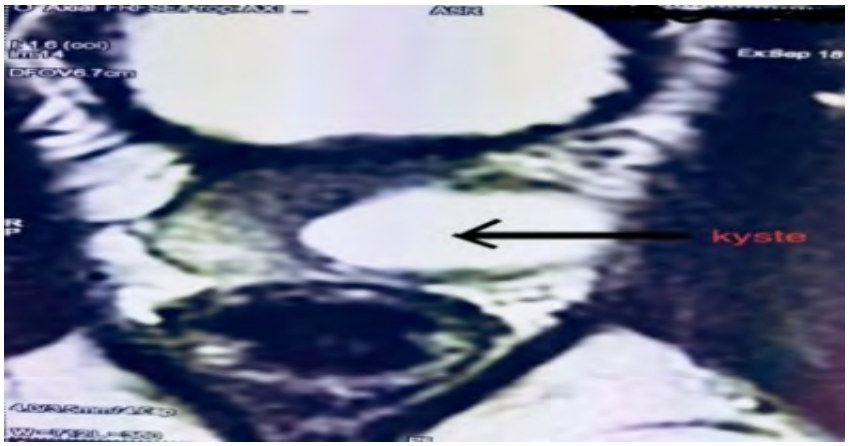

Figure 3. Prostate MRI allows a precise analysis of the relationship with neighbouring structures.

3. Discussion

Epidemiologically, Zinner's syndrome is a rare congenital condition that has a frequency of 0.00464% and characterized by a triad including a seminal vesicle cyst, renal agenesis and obstruction of the ipsilateral ejaculatory canal [2].

Embryologically, the mesonphric canal is responsible for the development of the upper urinary tract, epididymis, of the referential canal, seminal vesicles, ejaculating ducts, urethra, hemitrigon, and bladder neck. Aggression occurring before the 7th week of gestation of the distal part of the Wolff Canal, will cause atresia of the ejaculator canal, resulting in obstruction and dilation of the seminal vesicle, while an abnormality of the ureteral bud causes agenesis or renal dysplasia [3].

It is often during the 2nd and 3rd decade of life that the diagnosis is made because of the genital activity promoting the accumulation of seminal fluid in the vesicles.

The symptomatology is dominated by dysuria (37%), pollakiuria (33%), perineal pain (29%) and epididymite (27%) [1].

But in our patient there were only perineo-scrotal pains that had a particularly disabling character initially directing the etiological diagnosis towards a varicocele.

The diagnosis is essentially based on transrectal ultrasound which specifies the characteristics of the cyst of the seminal vesicle (anechogen, thin wall), and when the cyst is complicated from hemorrhage or infection, internal echoes appear. Exceptionally, the cyst may appear on ultrasound as a hypoesogenic tissue mass due to the hyperviscosity of its contents [4]. CT scans and MRI scans can be used to confirm or confirm the diagnosis in questionable cases. They also show associated anomalies [5] [6].

MRI appears to be superior to CT for the study of pelvic cystic formations [3]. MRI also allows an accurate study of cyst reports to other organs for surgical treatment and provides a differential diagnosis [5] [6] [7].

Nevertheless a cystoscopy in search of an absence of a hemi trigone is desirable.

Therapeutically, simple monitoring is recommended for non-symptomatic cysts; only symptomatic forms are indicated for essentially surgical treatment by a trans-rectal or trans-perineal guided echo aspiration of the cyst [8] [9]. In the event of recurrence after aspiration of the seminal vesicle cyst, some authors recommend a new suction and then supplement by injection of a sclerosir [9].

Another alternative is either by excision of the seminal bladder cyst by trans-vesical, extra-vesical or laparoscopic [7] [9].

Either by a transurethral detachment of the ejaculator canal and the cyst of the seminal vesicle, a complete surgical resection of the cyst by open surgery or laparoscopic, the latter pathway can even be assisted robot] [10] [11].

4. Conclusion

Zinner's syndrome is a rare condition that is often difficult to recognize, at first sight, our observation illustrating this by poor symptomatology made only of chronic perineo-scrotal pain. Nevertheless, the realization of a minimal workup including an adequate clinical examination and an initial radiological workup including an abdomino-pelvic ultrasound can orient the diagnosis and avoid a delay in diagnosis and management which can be deleterious on the functional level and on the patient's comfortable life.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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A Rare Interstitial Type of Post Appendectomy Incisional Hernia

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Abstract

Intraparietal Hernias are hernias occurring in the anterior abdominal wall at different anatomical planes. An interparietal hernia has a hernial sac that passes between the layers of the anterior abdominal wall. Appendectomy is a very common surgical procedure, and post appendectomy incisional hernia is a very rare complication. Here we present a case of a 24-year-old male with swelling in the right hypochondrium and lumbar region with an open appendectomy scar. He was diagnosed to have an interparietal hernia in the anterior abdominal wall. After obtaining consent patient was taken up for surgery. At surgery, the patient was found to have a defect in the transverse abdominis muscle with a medial leaf far from the incision site. Open repair of the defect along with double breasting of external oblique done. Interparietal hernias are rare in post appendectomy scar and this case is of significance since it Highlights a rare interstitial type incisional hernia, as a complication of post appendectomy scar, and not many cases reports are mentioned in literature.

Keywords

Interparietal Hernia, Post Appendectomy Incisional Hernia, Interstitial Type, Abdominal Wall Defect, Double Breasting

1. Introduction

One of the rare hernias occurring at the anterior abdominal wall is the Interparietal hernia which occurs at various anatomical (parietal) planes. An interparietal hernia has a hernial sac that passes between the layers of the anterior abdominal wall. This sac may be associated with, or communicate with, the sac of a concomitant ventral hernia. These types of hernias are difficult to suspect clinically or to diagnose radiologically hence it is usually missed or diagnosed lately or most-

ly as an intraoperative finding. In our case also, both USG could not diagnose it as an interparietal hernia, CT gave a picture of plane of the swelling, However, the extent of defect could not be accurately assessed which was finally seen intraoperatively and was treated accordingly by approximating transverse abdominis in upper 2/3rd and lower 1/3rd was sutured with internal oblique since the in lower part two leaf of transverse abdominis was far apart followed by double breasting of external oblique was done. This is a significant rare case interstitial type incisional hernia following post appendectomy as not many cases are found in the literature.

2. Case Report

A 24-year-old male came to Surgical Op with complaints of swelling in the Right hypochondrium, which was gradually increasing in size for the past 6 months. It increased with straining and reduced on lying down. Patient underwent open emergency appendectomy one year back. At physical examination of patient, a 5 × 5 cm swelling with an expansile cough impulse was noted in the region of appendectomy scar. His laboratory investigation showed hemoglobin 17.1 g/dl, hematocrit-51.6%, leucocytes-12,900, platelets-249,000, Na-139 mg/dl, K-4.0 mg/ dl, Cl-106 meq/dl, CT abdomen-A defect seen in anterior abdominal wall along the right transverse abdominis muscle fibers of size 5 × 4.2 cm (AP × TR) through which herniation of omentum and ileal loops is noted with resultant laxity and stretching of the right external oblique muscle fibers noted (**Figure 1** and **Figure 2**). The patient was posted for open repair with the diagnosis of Incisional interparietal hernia. Incision was made over the previous scar site and scar tissue was excised; Incision was deepened. Thick fibrous scar tissue was seen. External oblique fascial plane made. Defect of size 6 × 5 cm noted, hernial sac was adhered to external oblique muscle & aponeurosis. Sac opened and the contents were found to be small bowel and omentum (**Figure 3**). Adhesions between bowel and sac released and contents reduced. Defect was seen in transverse abdominis muscle with medial leaf 4 cm from incision site. After releasing bowel adhesions, to the inner surface of external oblique the Transverse abdominis muscle defect was approximated with 1.0 prolene only in upper 2/3rd of defect

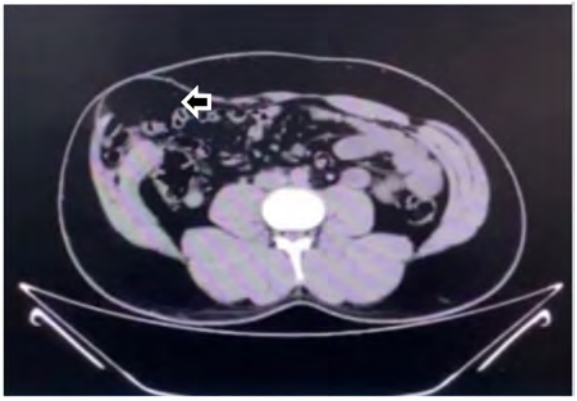

Figure 1. Pre-operative CT image arrow showing the interstitial incisional hernial sac.

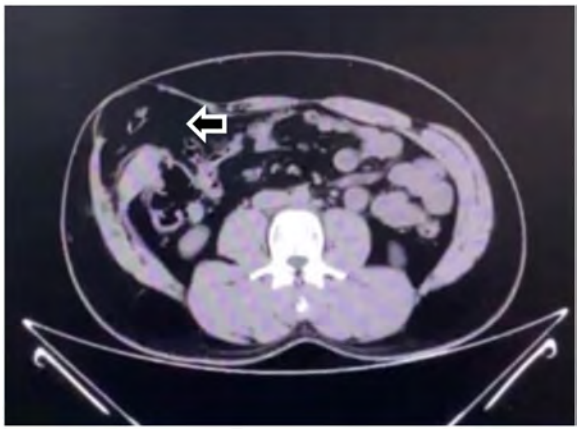

Figure 2. Defect of transverse abdominis Arrow showing bowel and omentum as content in hernial sac.

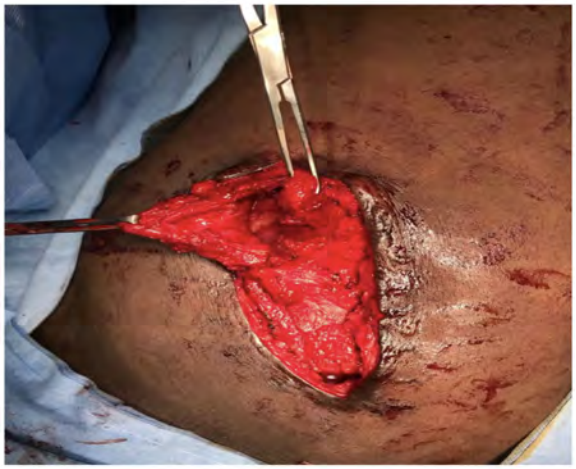

Figure 3. Defect with Internal oblique and transverse abdominis held.

(**Figure 4**). In lower 1/3rd Transverse abdominis muscle lateral leaf is seen near Anterior superior iliac spine and medial leaf seen near pubic symphysis hence medial leaf of muscle is sutured with internal oblique muscle (**Figure 5**). Double breasting of external oblique done (**Figure 6**). Suction drain placed skin sutured. Patient was comfortable post-operatively and was discharged on 4th post-operative day. He was free of any symptoms after his follow at 3 months.

3. Discussion

Bartolin was the pioneer to describe interparietal hernia in 1661 [1]. Interparietal hernia comprises nearly 0.01% - 1.6% of all hernias. Lower abdomen interparietal hernias are usually confused with the inguinal hernia mostly determined only at the time of operation. Sometimes lower abdomen intraparietal hernias frequently present with small bowel obstruction. Sub-types of Interparietal hernias include preperitoneal (between the peritoneum and transversalis fascia), Interstitial (between the muscle layers of external oblique and internal oblique), Superficial (between the skin and external oblique muscle) [2].

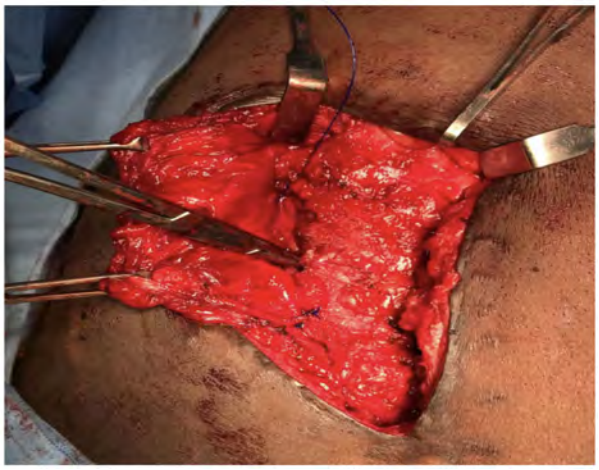

Figure 4. Upper half of transverse abdominis after suturing.

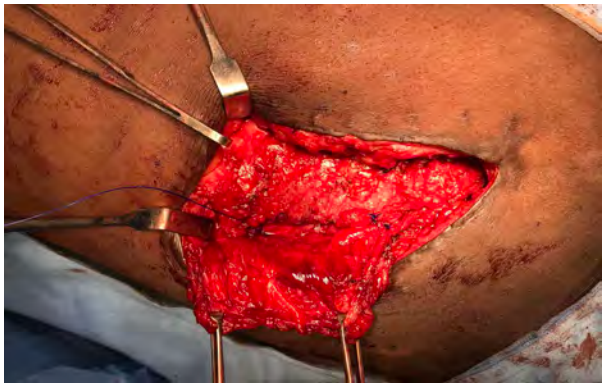

Figure 5. Lower half of transverse abdominis sutured with internal oblique.

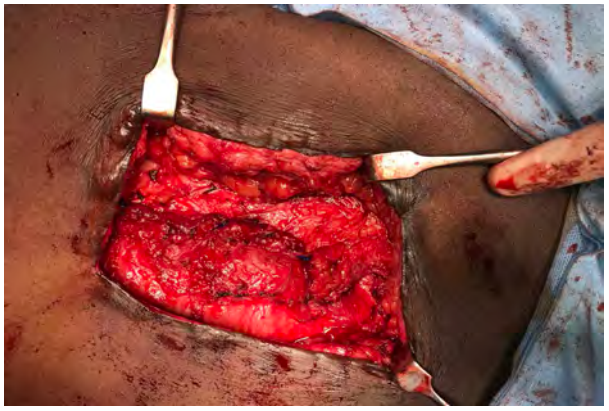

Figure 6. Double breasting of external oblique.

Prevalence of interstitial Sub-type is 60%. While other two combined accounts for 29%. Incidence of Interstitial hernia following abdominal surgeries is 10% [3].

These interparietal hernias predominantly occur in males and more common in children.

Interparietal hernia has a hernial sac which passes between layers of anterior abdominal wall. The sac may be associated with or communicating with sac of

concomitant inguinal or femoral hernia. These types of hernia present with obstruction or strangulation, which is often missed or diagnosed late because swelling is often absent in preperitoneal variety.

Interparietal hernia can sometimes present as Intestinal Obstruction appearing as an odd inguinal hernia. So, it is essential for surgeons to have correct preoperative diagnosis and plan for emergency surgery [4]. After appendectomy incidence of hernia is usually due to infection of wound in advanced peritonitis associated with local purulent peritonitis. Other reasons are drain when fixed tightly through incision may cause fleshly internal oblique and transverse abdominis muscle leading to necrosis [5].

Repair of the incisional Intrabdominal hernia is mostly done by anatomical repair. When tissue is atrophic, a thin overlap kind of repair is done and sometimes sutured with underlying structure along with strengthening of above layer by double breasting techniques [6]. Sometimes these defects are best treated with a sheet of polypropylene mesh, with a margin measuring at least 6-8 cm beyond the periphery of the defect, placed between the peritoneum and transverses abdominis muscle. Both techniques can be successfully implemented if the correct indications based on the extent of the defect and the clinical characteristics of the patient are respected [7]. Interparietal hernias are a significant complication resulting after any abdominal surgical incision. Significantly large defects of this variety can be successfully managed entirely with laparoscopic technique using barbed suture for primary repair of defect with subsequent mesh reinforcement [8]. In our case patient underwent open primary defect closure with double breasting of the external oblique muscle.

4. Conclusion

The combination of Incisional in post appendicectomy scar site is a very rare event. Even though the clinical diagnosis is challenging the treatment is surgical repair. Mostly diagnosed intraoperatively and not many cases find mention in a review of the literature. Hence great caution needs to be excised in the management of the patient with such kind of presentation.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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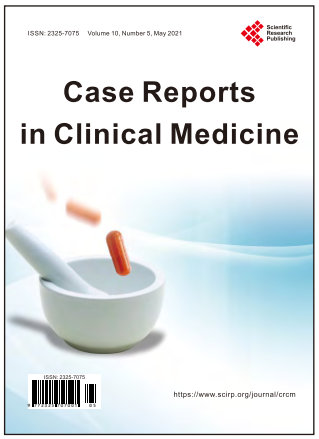

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