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A Review of the Literature on Endometriosis in the male including the prostate gland, testis, urinary bladder and inguinal region: An Update

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Abstract

Endometriosis is well known and understood to occur in females but not in the male. The aim of the study was to review the literature on endometriosis in the male by searching various internet data sources. Fourteen cases of endometriosis have been reported in men so far. Endometriosis in the male tends to arise in the remnant of the Müllerian duct. Majority of cases tend to ensue, treatment for adenocarcinoma of the prostate gland by means oestrogens. The disease could develop in relation to chromosomal abnormalities that affect: (a) the Müllerian inhibitory substance (MIS) / MIS gene, (b) abnormalities of the secretion of Müllerian inhibitory substance, (c) dysgenesis of the gonads, (d) dysgenesis of the testes) that are associated with Müllerian duct abnormalities, (e) dysgenesis of the kidney that are associated with Müllerian duct abnormalities. The disease may present with haematuria (prostate); lump in the inguinal region, voiding and or irritating lower urinary tract symptoms; loin discomfort, a palpable lump in the testis or epididymis. Microscopic examination of endometriosis lesions of the testis and Müllerian duct remnant areas would tend to show the ensuing characteristics: A true cyst which has been lined by cuboidal or columnar epithelium that contains hemosiderin laden macrophages within the stroma, Haemorrhagic fluid tends to be seen in the lumens, Even though endometrial tissue may not be predominant or conspicuous, careful examination of the specimen would usually reveal presence of endometrial stroma; On immunohistochemistry studies the epithelium on staining for cytokeratins stain positively with: CK7, CK8, CK18, Oestrogen receptors (ER), Progesterone receptors (PR), Epithelial membrane antigen, Calretinin, Cytokeratin 5/6, The stromal cells, exhibit positive staining for: Oestrogen receptor (ER), PR, CD10. Calretinin (Some of the cells may be positive) Complete excision of endometriotic lesions in the male tend to lead to cure and no recurrence. Endometriosis is rare but can occur in the male patient therefore clinicians should have a high index of suspicion for the disease.

Key Words: Endometriosis of prostate; endometriosis of testis; endometriosis in the male; Male endometriosis; Mullerian inhibitory substance; the Müllerian inhibitory substance; Oestrogen receptors (ER), Progesterone receptors (PR)

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Introduction

The occurrence of endometriosis in the male is very rare and it is likely that a number of clinicians would unfamiliar with the disease. It would be perturbing to a man to be told that he has endometriosis. The ensuing review article on endometriosis in the male is divided into two parts (A) Overview and (B) Miscellaneous narrations and discussions from reported cases.

Aim: To review the literature on endometriosis in the male

Method: Various internet data bases were searched including Google, Google scholar, Educus, and PUB MED. The searched words that were used included: Endometriosis in male; Endometriosis of prostate gland; endometriosis of male bladder; endometriosis of testis, endometriosis of epididymis; endometriosis

of male inguinal region. Thirty references were used to write the article.

Results / Literature Review

(A) Overview

Definition

Sites

Endometriosis in the male is extremely rare but has been sporadically reported in the prostate gland, epididymis and sites of the Müllerian duct remnant [1]

Aetiology and Pathophysiology

- It has been stated that endometriosis in the male tends to arise in the remnant of the Müllerian duct including the prostatic utricle and in the appendix testis. [1]
- It has been stated that majority of cases of endometriosis affecting the male genital tract tend to ensue treatment for adenocarcinoma of the prostate gland by means oestrogens. [1]
- It has also been documented that endometriosis in the male could develop in relation to chromosomal abnormalities that affect: (a) the Müllerian inhibitory substance (MIS) / MIS gene, (b) abnormalities of the secretion of Müllerian inhibitory substance, (c) dysgenesis of the gonads, (d) dysgenesis of the testes) that are associated with Müllerian duct abnormalities, (e) dysgenesis of the kidney that are associated with Müllerian duct abnormalities. [1]

Presentation

- Endometriosis of the prostate and endometriosis affecting the male are rare therefore there is not a lot of literature on their presentation; however, it has been stated that endometriosis of the prostate and endometriosis-like lesions affecting the male genital tract or Müllerian duct remnant sites tend to be incidental findings. [1]
- Endometriosis of the prostate / prostatic urethra may present with haematuria as was reported at a stage in a case reported by Beckman et al. [2]
- Endometriosis lump in the inguinal hernia region may present as a lump in the inguinal region
- Endometriosis lesion in the prostate/prostatic urethra, or within the urinary bladder may present with voiding and or irritating lower urinary tract symptoms
- Endometriosis obstructing the ureter in a male may present with loin discomfort
- A history of treatment with oestrogens may or may not be associated with endometriosis in the male.
- Endometriosis affecting the testis or epididymis may present as a palpable lump in the testis or epididymis.

Investigations

Urine microscopy culture and sensitivity

- Urine microscopy, urine culture and sensitivity would be undertaken in cases of endometriosis in the male as part of the general assessment of a patient with endometriosis but the results would not be diagnostic of endometriosis.

Haematological investigations

- Full blood count and coagulation screen are useful tests in the general assessment of a male patient with endometriosis; however, the results would not be diagnostic of endometriosis. In cases of acute infections the white cell count would tend to be raised and there may be lymphocytosis in cases of chronic granulomatous inflammations that may mimic endometriosis or which could be considered in the differential diagnosis of endometriosis in a male.

Biochemistry investigations

- Serum urea and electrolytes, bone profile, liver function tests and serum glucose are general tests that are used in the general assessment of a male patient with any lump or mass that could be considered as a differential diagnosis of endometriosis but none of them are diagnostic of endometriosis. Furthermore, if endometriosis localized to the uretero-vesical junction area causes ureteric obstruction and the impairment in renal function the results of the renal function test would reflect the state of kidney injury (acute or chronic).
- Serum Beta Human Chorionic Gonadotrophin, (ß-HCG), Lactic dehydrogenase, (LDH) and alpha fetoprotein (AFP) tests are useful tests that are carried out in cases of lumps or tumours related to the testis for testicular tumours that could perhaps mimic endometriosis affecting the testis and epididymis area but in cases of endometriosis on the whole the results should be normal.

- Serum prostate-specific antigen is a common test undertaken as part of the assessment of patients with prostatic lumps / enlargements and in cases of endometriosis involving the prostate and prostatic urethra the results should be normal unless there is an associated adenocarcinoma of the prostate. Additionally, serum PSA is a test that is routinely carried out in the follow-up assessment of patients undergoing various treatments and follow-up protocols for adenocarcinoma of the prostate gland including patients on oestrogen therapy; nevertheless, the serum PSA results would not be diagnostic of endometriosis.

Radiological investigations

Ultrasound scan

- Ultrasound scan of the renal tract, abdomen and pelvis is useful in the assessment of masses in the abdomen and pelvis and would define whether the mass is solid or cystic as well as the position of the mass. It would also identify whether or not there is hydronephrosis or not.
- In the rare situation where there is hydronephrosis associated with ureteric obstruction by endometriosis per-cutaneous nephrostomy can be inserted under ultra-sound scan guidance and also via the percutaneous nephrostomy an ante-grade ureteric stent can be inserted temporarily to improve renal function prior to excision of the endometriosis lesion and pursuant to the excision of the endometriosis lesion nephrostogram can be undertaken and if there is no further obstruction the nephrostomy tube can be removed. In cases of previous ureteric obstruction by endometriosis, ultrasound scan of renal tract, abdomen and pelvis can be periodically undertaken to confirm there is no recurrent endometriosis lesion and there is no subsequent development of hydronephrosis.

Computed Tomography Scan

- Computed tomography (CT) Scan of abdomen and pelvis is useful for the assessment of endometriosis lesions of the pelvis and this would define the position, size and characteristics of the lesion as well as the number of lesions which would help the surgeon plan the surgical excision of the endometriosis lesion(s). CT scan of abdomen and pelvis can be used in the follow-up assessment of the patient to establish whether or not there is recurrence of the endometriosis lesion locally or elsewhere.

Magnetic Resonance Imaging Scan

- Magnetic Resonance Imaging (MRI) Scan of abdomen and pelvis is useful for the assessment of endometriosis lesions of the pelvis and this would define the position, size and characteristics of the lesion as well as the number of lesions which would help the surgeon plan the surgical excision of the endometriosis lesion(s). MRI Scan of abdomen and pelvis can be used in the follow-up assessment of the patient to establish whether or not there is recurrence of the endometriosis lesion locally or elsewhere.

Treatment

Complete excision of the endometriosis lesion in a male irrespective of site tend to lead to cure without any recurrence or long term sequels in view of this surgeons should endeavour when practicable to ensure they achieve complete excision of the lesion when histological confirmation of the diagnosis has been established.

Macroscopic features

- Gross examination of endometriosis lesion affecting the epididymis, para-testicular region or testis would tend to show haemorrhagic cysts that may be small, single to multiple, uniloculated, or multiloculated [1]
- Resected endometriosis lesion of the prostate / prostatic urethra may be seen as an abnormal red-tan tissue [2]

Microscopic features

Microscopic examination of endometriosis lesions of the testis and Müllerian duct remnant areas would tend to show the ensuing characteristics:

- A true cyst which has been lined by cuboidal or columnar epithelium which contains hemosiderin laden macrophages within the stroma [1]
- Haemorrhagic fluid tends to be seen in the lumens. [1]
- Even though endometrial tissue may not be predominant or conspicuous, careful examination of the specimen would usually reveal presence of endometrial stroma.

Immunohistochemistry

Endometriosis lesion of the prostate or the Müllerian duct related sites would on immunohistochemistry

studies tend to exhibit the ensuing staining characteristics: [1]

- With regard to epithelium staining for cytokeratins they would stain positively with:
  - CK7 [1] [3]
  - CK8 [1]
  - CK18 [1]
  - Oestrogen receptors (ER) [3]
  - Progesterone receptors (PR) [3]
  - Epithelial membrane antigen [3]
  - Calretinin [3]
  - Cytokeratin 5/6 [3]
- With regard to the staining of stromal cells, the stromal cells tend to exhibit positive staining for:
  - Oestrogen receptor (ER) [1]
  - PR [1] [3]
  - CD10 [1] [3]
  - Calretinin (Some of the cells may be positive) [3]

Differential Diagnoses

Some of the differential diagnosis of endometriosis of the prostate, testis/epididymis/seminal vesicles, pelvic, retroperitoneal, and abdominal wall endometriosis include:

- Teratoma, especially when seminoma is present in the testis [3]
- Inflammatory lesions of the prostate gland including inflammatory myofibroblastic tumour of the prostate gland, chronic prostatitis, brucellosis of the prostate, sarcoidosis of the prostate gland and other rare miscellaneous lesions of the prostate gland
- Haemorrhagic lesions of prostate and testis / epididymis [4] [5] [6] [7] [8] [9] [10] [11]
- Genitourinary chlamydia infections
- Rare tumours of the urinary bladder
- Rare tumours of the abdominal wall and pelvis as well of the inguinal region

Outcome

- Complete excision of endometriosis lesion in the male should lead to cure and improvement of symptoms and usually it would be expected that there would be no recurrence.

(B) Miscellaneous narrations from reported cases

Oliker and Harris [12] in 1971 reported a case of endometriosis of the urinary bladder in a male patient who had been treated with oestrogen therapy for adenocarcinoma of the prostate gland after radical prostatectomy and bilateral orchidectomy.

Pinkert et al. [13] in 1979 reported a man who had been treated with oestrogen for many years pursuant to radical prostatectomy and orchidectomy which he had had for adenocarcinoma of the prostate gland. The endometriosis had involved the patient's urinary bladder. Pinkert et al. [12] reported that in 1955 the patient who was then aged 50 years-old was found on digital rectal examination to have a small hard nodule in the right supero-lateral aspect of the posterior lobe of his prostate gland. Following frozen section histological confirmation of his prostate biopsy, he underwent radical perineal prostatectomy and seminal vesiculectomy. Microscopic examination of the prostatectomy specimen confirmed adenocarcinoma of the prostate gland of the acinar type. He had remained asymptomatic for five years with no evidence of recurrent disease. In 1968 (13 years later), he developed perforated appendix, intestinal obstruction and pelvic abscess for which he had drainage of the pelvic abscess. He represented the same year with rectal bleeding, pain and intermittent diarrhoea for which he had sigmoidoscopy which revealed a nodule in the anterior wall of the rectum. He underwent excisional biopsy of the nodule and histological examination of the lesion showed metastatic adenocarcinoma which was histologically identical to his previous adenocarcinoma of the prostate gland. He underwent bilateral orchidectomy and histological examination of the specimen showed decreased spermatogenesis which was adjudged to be consistent with the patient's age. He was commenced on oestrogen therapy and he received 12 mg of chlorotrianisene (TACE). In 1972 (about 17 years after his initial presentation) he represented with visible haematuria for which he underwent cystoscopy that revealed a sub-mucosal tumour nodule on the left side of the trigone. The lesion was resected and microscopic examination of the specimen showed adenocarcinoma of the prostate that had infiltrated in between the urinary bladder wall muscle bundles and along the regional nerves. Additionally the microscopic examination showed islands of large glands within an areolar stroma which was adjudged to look identical to hyperplastic endometrium but at

the time of initial examination the pathologist had overlooked a diagnosis of endometriosis hence the dosage of Chlorotrianisene (TACE) was increased to 24 mg. In July 1975 (about 20 years after the patient's initial presentation) he represented with recurrent visible haematuria. He had intravenous urogram which showed obstruction at the vesicoureteric junction. He underwent cystoscopy which showed a large ulcerated lesion in the trigone of the urinary bladder that occluded the left ureteric orifice which prevented catheterization of the left ureter. Histological examination of biopsy specimen of the lesion showed foci of endometriosis that involved the muscle wall of the urinary bladder and it had extended into the ulcerated mucosal surface of the urinary bladder. The previously resected bladder lesion was re-examined histologically and it confirmed presence of endometriosis. Subsequently he underwent resection of the entire residual lesion in the urinary bladder. Histological examination of the specimen showed larger areas of endometriosis as well as ulcerative cystitis but no evidence of residual adenocarcinoma of the prostate gland. The TACE medication was discontinued and the patient was put on one tablet per day of Brevicon which is an oral contraceptive which has been used in the treatment of female endometriosis. Nevertheless he continued to have intermittent haematuria that had culminated in a haemorrhagic episode associated with acute urinary retention for which he had cystoscopy and diathermy of a bleeding area in the left side of the trigone of the urinary bladder and the Brevicon medication was stopped four months later and following this he had been asymptomatic. In 1976 (about 21 years after his initial presentation) he underwent cystoscopy which showed an intact urinary bladder mucosa and it also did show that the mass in the region of the left ureter had reduced remarkably in size but no biopsy was taken. Pinkert et al. [13] stated that only one other case of endometriosis in the male had been reported in a man who had also been treated with oestrogen in a similar manner.

Schrodt et al. [14] in 1980 reported tissue which was histologically indistinguishable from endometrium from the urinary bladder of a 73-year-old man. They reported that the lesion had involved the right ureterovesical junction which had been responsible for right sided hydronephrosis in the patient. They also reported that the patient had been on oestrogen treatment for adenocarcinoma of the prostate gland for 5 years preceding the diagnosis of endometriosis. They also stated that prior to the report of their case of endometriosis in the male, there were two reports of endometriosis in the male and each of the two previously reported men with endometriosis had also received oestrogen therapy.

Beckman et al. [2] reported a case of endometriosis of the prostate gland in which the endometriosis was reported to have occurred in the prostate gland of a 78-year-old man after he had had a long course of oestrogen treatment. It had been stated that presence of endometrial tissue in the region of the urethral crest constitutes a potential histogenic and therapeutic implications for some of the neoplasms which arise in the area of the urethral crest. Beckman et al. [2] reported a man who was aged 72 years in 1959, and who had been married for 16 years without having a child, who was found on clinical examination to have mild enlargement of the prostate gland with areas of fixation of the prostate as well as a distinctly hard nodule in the left lobe of prostate. He was diagnosed clinically as having adenocarcinoma of the prostate gland. He had chromosomal analysis and based upon the features of the chromosomal studies he was adjudged to have Barr chromatin negativity and his karyotype was 46, XY. For the ensuing 5 years and 9 months, he had been taking oestrogen medication for his clinically diagnosed prostate cancer in the form of oestrogen chlotrimisene (TACE) 12 mg twice daily. The prostate gland on follow-up assessment had been found to have decreased in size somewhat; nevertheless, the nodule on the left lobe had been noted to have remained distinctly firm even though its consistency had decreased. There was no evidence of any metastatic disease. Six years later, when he was aged 78 years he presented with visible haematuria for which he had cystoscopy which showed a small raised area immediately proximal to his internal urethral meatus. Trans-urethral resection of the urethral lesion and the adjacent prostate gland was undertaken with resection of 7 grams of tissue. Macroscopic examination of the resected raised area showed abnormal looking red-tan tissue and the alteration was observed to have extended into the prostate gland. Microscopic examination of the specimen showed multiple zones of well-defined endometriosis situated adjacent to adjacent areas of the prostate, and one fourth of the examined resected chips had contained endometriosis. The microscopic examination of the resected specimens also showed that the regions of the endometriosis had exhibited a typical appearance which consisted of distinctly endometrial type glands in a basophilic small cell endometrial stroma. Immunohistochemistry studies

of the specimen showed that the glands of the endometriosis stained negatively for prostate specific antigen (PSA); however, the prostate glands stained strongly positively for PSA. Furthermore, microscopic examination of the specimen also showed that some of the glands were dilated cystic fashion that mimics cystic hyperplasia which is seen pursuant to stimulation of the endometrium with prolonged oestrogen treatment. Many of the glands that were surrounded by endometrial stroma were lined by cuboidal endometrial glandular cells which had merged into columnar cells that had somewhat mimicked glandular cells of the prostate gland. Nevertheless, immunohistochemistry study of the gland that were most suggestive of transformation showed negativity for prostate-specific antigen. Microscopic examination of the specimen also showed the endometrial stroma to be moderately well demarcated from the adjacent stroma of the prostate, and furthermore, the endometrial stroma looked distinctly different from the stroma of the prostate with a chronic inflammatory infiltrate. The nuclei of the endometria were slightly oval, contained less dense chromatin in comparison with the nuclei of lymphocytes, and they were about twice the diameter of the lymphocytes. Haemorrhages were noted in the stroma, which were adjudged to be recent and perhaps related to the surgical resection; nevertheless, there was no evidence of any haemorrhage within the adjacent prostate. An occasional small deposit of hemosiderin was found in the endometrial stroma. The microscopic features of the prostate depicted that of benign prostatic hyperplasia. Microscopic examination of the specimen did not reveal presence of adenocarcinoma in anywhere. Beckman et al. [2] additionally reported that with regard to the patient despite having been on oestrogen treatment, there had never been a histologically proven documented adenocarcinoma of the prostate gland. Based upon the aforementioned findings they were of the opinion that the firm nodule that had been palpable in the patient was most likely an area of unusually firm benign prostatic hyperplasia or perhaps it was a calculus within the prostate gland. The patient died at the age of 91 years (13 years after the diagnosis of the endometriosis) but no autopsy was carried out.

. Martin and Hauck [15] reported an 83-year-old man who was found to have endometriosis mass of the lower abdominal wall. He had over a period of ten years received 25 mg of TACE therapy pursuant to a diagnosed carcinoma of the prostate gland. He underwent a second trans-urethral resection of prostate (TURP) and histological examination of the resected prostate confirmed adenocarcinoma. Pursuant to the second trans-urethral resection of his prostate gland he was continued on the TACE treatment. He had chromosomal analysis which demonstrated a normal male phenotype. After a prolonged course of treatment, he was reviewed regularly until his death in 1979. There was no evidence of recurrence of his abdominal wall mass but he continued to have persistent low-grade adenocarcinoma of the prostate gland. His death was related to cardiovascular disease and this was not related to his carcinoma of the prostate gland.

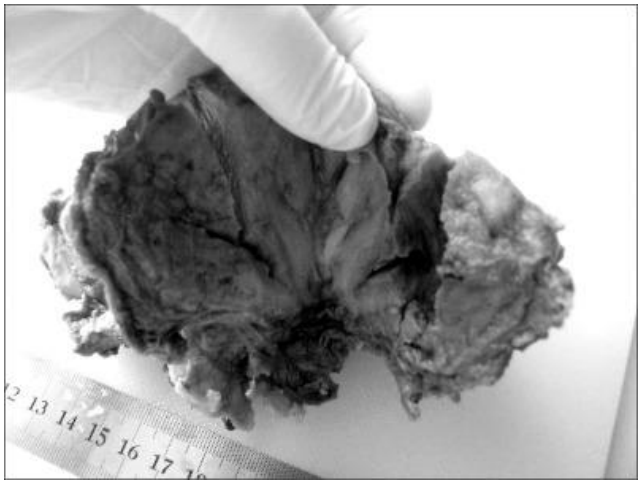


Illustration 1: Uterus-like mass beside the deferent ductus, holded area in the figure. Reproduced from [17] Simsek G, Bulus H, Tas A, Koklu S, Yilmaz S B, Coskun A. An Unusual Cause of Inguinal Hernia in a Male Patient: Endomtriosis. Gut and Liver 2012 Apr; 6(2): 284 – 285 DOI: http://dx.doi.org/10.5009/gnl.2012.6.2.284 with copy right © 2012 by the Korean Society of Gastroenterology, the Korean Society of Gastrointestinal endoscopy, the Korean Society of Neuro-gastroenterology and Motility, Korean College of Helicobacter and Upper Gastrointestinal Research, Korean Association for the Study of Intestinal Diseases, the Korean Association for the study of the Liver, Korean Pancreatobiliary Association, and Korean Society of Gastrointestinal Cancer. This is an Open Access Article distributed under the terms of the Creative Commons Attribution Non-Commercial License (http://creativecommons.org/licences/by-nc/3.0) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

Young and Scully [16] in 1986 described three new and eleven previously reported cases of testicular or para-testicular tumours that mimicked ovarian tumours of common epithelial type. They reported that the 14 tumours had occurred in patients

whose ages had ranged between 11 years and 68 years with a mean an average age of 47 years. The exact location of the tumours was known in the cases of 12 patients as follows: 5 had involved the testicular parenchyma primarily, 3 had involved the tunica vaginalis, and 4 had involved the para-testicular tissue. Five of the tumours were serous, four of which adjudged to be in the borderline category. Four of the tumours were Brenner tumours, and in one case it was admixed with an adenomatoid tumour. The rest of the tumours consisted of single examples of mucinous cystadenoma, mucinous cystadenocarcinoma, endometrioid adenoacanthoma, clear cell adenocarcinoma, and a benign tumour of mixed cell types. Young and Scully [16] stated that follow-up data was available in eight of the cases and the follow-up had ranged between 3 months and 14 years and that only one tumour which was the case of clear cell adenocarcinoma was known to have been clinically malignant. Furthermore, there was a fourth lesion in their article which was a para-testicular mass that had been composed of endometrial glands and stroma and smooth muscle. This lesion (endometriosis) was found in an 82-year-old man who had been treated with oestrogen for adenocarcinoma of the prostate gland.

Simsek et al. [17] in 2012 reported the first case of endometriosis which arose in the inguinal region, near the ductus deferens. They reported a 49-year-old man who had presented with a left inguinal hernia. He had undergone three previous operations in that area. He also had infertility without an explanation of its cause. He was found on examination to have normal external genitalia. He underwent surgical exploration for his provisionally diagnosed hernia and during the procedure, a mass was found beside the spermatic cord and this was excised for pathological examination. On gross examination, the mass was found to measure 8 cm x 7 cm x 6 cm (see illustration 1). On dissection the mass was found to contain a cystic cavity and the ductus deferens was found. The cyst wall was found to contain small lumens which were considered to be small vessels. Microscopic examination of the specimen showed that the cyst was lined with columnar epithelial cells and simple tubular invaginations that showed the same type of cell lining and the cellular stroma had exhibited typical features of endometrial mucosa. Furthermore, the microscopic examination also showed smooth muscle proliferation and some endometrial glands and stroma in those areas (see illustration 2).

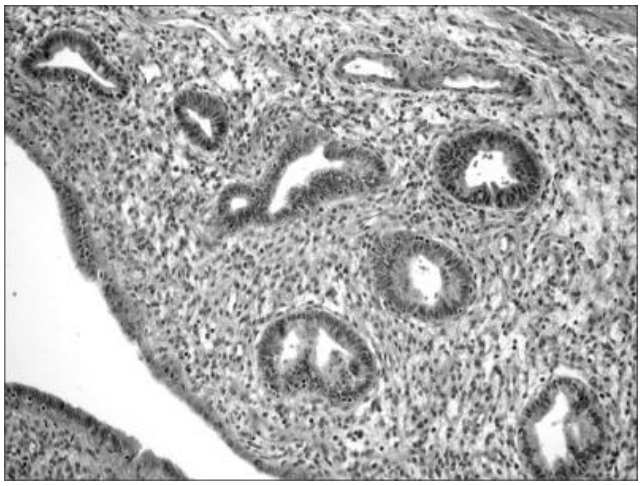


Illustration 2: High power field of endometriotic area (H&E stain, 200). Reproduced from [17] Simsek G, Bulus H, Tas A, Koklu S, Yilmaz S B, Coskun A. An Unusual Cause of Inguinal Hernia in a Male Patient: Endometriosis. Gut and Liver 2012 Apr; 6(2): 284 – 285 DOI: http://dx.doi.org/10.5009/gnl.2012.6.2.284 with copy right © 2012 by the Korean Society of Gastroenterology, the Korean Society of Gastrointestinal endoscopy, the Korean Society of Neuro-gastroenterology and Motility, Korean College of Helicobacter and Upper Gastrointestinal Research, Korean Association for the Study of Intestinal Diseases, the Korean Association for the study of the Liver, Korean Pancreatobiliary Association, and Korean Society of Gastrointestinal Cancer. This is an Open Access Article distributed under the terms of the Creative Commons Attribution Non-Commercial License (http://creativecommons.org/licences/by-nc/3.0) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

Simsek et al. [17] stated the following:

- With regard theories related to the pathogenesis of endometriosis in the female they had found three theories (a) transplantation theory, (b) coelomic theory, (c) embryologic cell rest theory [18] [19]
- The transplantation theory has been stated to be impossible in the male in view of the absence of a source of menstrual material. [13]
- The coelomic metaplasia could ensue inflammatory and hormonal influences and this theory could offer an explanation with regard to endometriosis in women who have Müllerian agenesis and who have absence of uterus or it could offer an explanation of the pathogenesis of

endometriosis that are occasionally reported in men. [20]

- With regard to the third theory, the induction theory which has been pivoted upon the induction of embryologic cell rests. Cell rests could persist between the appendices testes and the utricle. It would be expected that the aforementioned Müllerian cell rests would be located along the course the course of the ejaculatory ducts and the ductus deferens. Based upon these explanations. [13] It was their view that the induction theory may have played a role in their patient's development of endometriosis.
- Additionally, their patient had taken a drug which is named, Fertilin, for maturation and activation of his sperm. Fertilin consists of clomiphene citrate has anti-oestrogenic effect by means of binding oestrogen receptors in the hypothalamus and in the hypophysis. [21] The patient's case would contrast other cases of endometriosis reported in men in the literature. For this reason, it may be that an additional contributory factor may have played a role in their patient developing endometriosis.

Gonzalez et al. [22] stated that male endometriosis and endomyometrium which is also called "uterus-like mass" are two unusual presentations of endometriosis. Gonzalez et al. [22] reported a 52-year-old man who had complained having a stabbing pelvic pain. He had computed tomography (CT) scan which had shown a mass in his right inguinal area at the site of his previous inguinal hernia repair. The lesion was noted to be tubular in shape, with a thick muscular wall and a central blood like lumen. Gonzalez et al. [22] reported that microscopic examination of the excised did show layers of concentric smooth muscle, with endometrial glands and stroma lining lumen. Gonzalez et al. [22] further reported that the histopathological, immunohistochemistry, as well as molecular studies undertaken had confirmed the lesion to be that of endometriosis. Gonzalez et al. [22] iterated that a number of postulates have been promulgated with regard to aetiology of both endomyometriosis and male endometriosis including (a) remnant rests of Müllerian tissue and (b) metaplasia. Gonzalez et al. [22] also intimated that some reported cases of endometriosis in the male had been linked typically with oestrogen treatment for adenocarcinoma of the prostate gland. Additionally, Gonzalez et al. [22] reported that their patient had a history of cirrhosis of the liver and had taken spironolactone, which they conjectured might have led to an alteration of the patient's hormonal state which might have interacted with reactive/metaplastic process at the site of the patient's previous hernia repair site.

Jabr and Mani [23] reported a 52-year-old man who had a history of cirrhosis secondary to hepatitis and a past history of having had banding of oesophageal varices and a right inguinal herniorrhaphy twice over a period of two years. He had a mesh hernia repair during his second operation seven months preceding his presentation. He was seen on emergency basis with a 3-weeks history of right sided lower abdominal and pelvic pain which got worse on getting up from a supine position and which was not improved following bowel movements. The pain slightly got worse on voiding. He was found on examination to be tender in his lower abdomen and over his supra-pubic region as well as over the area of his hernia repair site. There was no clinical evidence hernia recurrence. He had a computed tomography (CT) scan which revealed cirrhosis of the liver, and an extra-peritoneal soft tissue mass which measured 2.6 cm x 2.3 cm in the pelvis adjacent to his urinary bladder and which had extended into his right inguinal canal. Based upon the CT scan findings, a provisional diagnosis of Meckel's diverticulum was made; however, he had a radio-nuclear Meckel's scan which was negative and which had excluded Meckel's diverticulum. He underwent laparoscopy which revealed a cystic mass that was adherent to his urinary bladder and the right inguinal area. The mass was next excised completely and the urinary bladder repaired. The excised mass measured 4.5 cm x 2.5 cm in diameter. Macroscopic examination of the specimen showed tissue with a diameter of 1.5 cm which was filled with old blood and surrounded by a thick muscular mass. Microscopic examination of the specimen showed the mass to consist of thick, smooth muscle fibres which had in a concentric fashion encompassed a central lumen bordered by endometrium-like glands and stroma. Immunohistochemistry studies of the specimen showed positive staining for oestrogen and progesterone receptors in the glandular component and for CD10 in the stroma which was adjudged to be consistent with endometriosis. His pain resolved completely pursuant to excision of the mass.

Giannarini et al. [24] stated in 2006 that endometriosis of the genito-urinary in the male sex is very rare and that there had been only 6 previously reported cases of endometriosis in men. Giannarini et al. [24] reported a 27-year-old man who presented with scrotal pain who was diagnosed as having a cystic endometriosis of the epididymis which they stated was the first reported case of cystic

endometriosis of the epididymis. The patient had never received any treatment with oestrogen.

Fukunaga [25] reported a 69-year-old man with adenocarcinoma of the prostate gland who had been under hormonal treatment for his carcinoma for 9 years who was found to have a multiloculated cyst in his left para-testis that measured 5.2 cm x 3.1 cm x 3.0 cm. Histological examination of the excised cyst showed that the cyst was lined by a single layer of cells that were cuboidal in appearance. The microscopic examination also showed a few glands scattered in the underlying stroma which had consisted of closely packed round or oval cells which had scanty cytoplasm mimicking endometrial stromal cells, as numerous capillaries as well as hemosiderin deposits. Immunohistochemistry studies of the specimen showed that the lining of the cysts and glands were positively stained for CAM5.2, vimentin, and calretinin, but they were negative for CD10, oestrogen, and progesterone receptors, which was considered to have indicated mesothelial profiles. Immunohistochemistry studies had also shown that the stromal cells had stained positively with vimentin, oestrogen, progesterone-receptors and CD10. The aforementioned findings were adjudged to be diagnostic of stromal endometriosis associated with the tunica vaginalis testis. Fukunaga [25] stated that Mullerian or endometriotic metaplasia and stromal hyperplasia under the influence of prolonged oestrogen treatment would be the mechanism behind the development of the endometriosis.

Taguchie and Enomoto, [26] reported a 74-year-old man who had undergone radical prostatectomy for localized carcinoma of the prostate gland 5 years preceding his presentation with visible haematuria. He had received treatment with leuprorelin and ethynylestradiol for 5 years as adjuvant treatment for a metastatic Gleason 4 + 3 = 7 with tertiary pattern 5 adenocarcinoma of the prostate gland which was staged pT3bN1M0. His serum PSA had been undetectable. He had cystoscopy which revealed a 3 cm solid mass situated over the left ureteric orifice in the urinary bladder. His urine cytology was negative and did not reveal any malignant cells. He had computed tomography (CT) scan which showed a mass that was reported to be suspicious of an invasive urinary bladder cancer. He underwent trans-urethral resection of the bladder tumour and histological examination of the specimen revealed papillary tubular glands that were scattered in the underlying stroma. The stroma consisted of closely packed small round or ovoid cells that had scant cytoplasm which mimicked endometrial stromal cells, associated with abundant capillaries and arterioles. There was no evidence of atypia, or mitosis within the stromal or glandular cells. The urothelium had normal cells. Immunohistochemistry studies showed that the stromal cells were positively stained for CD10, oestrogen receptor (ER), and progesterone receptor (PGr); but on the other hand the glandular cells stained negatively for CD10 and positively stained for oestrogen receptor (ER) and progesterone receptor (PR). Furthermore, the stromal cells and the glandular cells both were negative negatively stained for prostate specific antigen (PSA). A diagnosis of endometriosis of the urinary bladder was made based upon the histopathological and immunohistochemistry characteristics of the urinary bladder lesion. Six months pursuant to withdrawal of the ethinyl estradiol (EE), the tumour was noted to have shrunk both at cystoscopy and also based upon a repeated CT scan finding and the serum PSA was also noted not to be elevated. Taguchi and Enomoto [26] stated that endometriosis in the male is extremely rare and to their knowledge only eight cases of endometriosis in the male had been reported. Taguchi and Enomoto [26] stated the following: (a) Out of the 8 previously reported cases of endometriosis in men, 7 of the cases had been associated with prolonged administration of oestrogen treatment for carcinoma of the prostate gland [12] [25] and the only reported case of endometriosis in a male not associated with oestrogen treatment was reported in a 27-year-old man who was healthy and asymptomatic. [24] Taguchi and Enomoto [26] also stated the following:

- During the process of the normal development of the male genital organ systems, the embryonic Müllerian ducts undergo regression under the influence of Müllerian-inhibiting substance which is produced by Sertoli cells and in the end only the cranial and caudal ends of the Müllerian ducts permits which results in the formation of the appendix testis, and prostatic utricle.
- Higashi et al. [27] had stated that pathological lesions that emanate from Müllerian duct remnants have been found in less than 1% of male adults and on the whole have tended to be asymptomatic.
- They had reviewed all the 9 reported cases of endometriosis in men inclusive of their reported case and had found out that majority of the endometriosis lesions had developed and had been localized along the route of the Müllerian duct which is almost that of the ductus deferens, including the verumontanum, the trigone of the urinary bladder, ureterovesical junction, the lateral wall of the urinary bladder, and the para-testicular region.

- There was nothing relating to their reported case that would indicate any disorder of sex development.
- There is the assumption that oestrogen treatment could lead to induction of endometrial differentiation within a persistent Müllerian duct remnant. Furthermore, two major postulates that had been promulgated for the development of female endometriosis include (a) metaplasia of coelomic tissue, and (b) implantation of refluxed endometrium. In their opinion, the development of endometriosis in the male might endorse the coelomic metaplastic postulate.
- There is no consensus relating to the treatment of endometriosis; however, theoretically it would be assumed that cessation of oestrogen treatment should lead to shrinkage of the endometriosis lesion.
- Their reported case is the first case report which had illustrated regression of endometriosis lesion in a male pursuant to cessation of oestrogen treatment.

Zámecnik and Hoštáková [3] reported a 46-year-old man who had undergone right radical orchidectomy for seminoma of the testis, based upon a clinical diagnosis of testicular tumour. Macroscopic examination of the orchidectomy specimen revealed a 10 cm seminoma which had occupied the entire testis. Additionally, on the medial aspect of the mediastinum testis, a slightly thickened tunica albuginea associated with a cyst that measured 0.7 cm, was found. The cyst was serous type and it was adjudged to be suggestive of a mesothelial inclusion cyst of the tunica vaginalis. The tumour was noted to have grown into the rete testis and into the epididymis. Furthermore, a 1.5 cm metastatic nodule was observed within the spermatic cord about 4 cm from the epididymis. Histopathological examination revealed the 7 mm cyst to be of the mesothelial type which was situated within the fibrous stroma immediately inferior to the mesothelium of the tunica vaginalis. A 4 mm focus of endometriosis was observed within part of the wall of the cyst. Within the area with features of endometriosis, the mesothelium did show transition into columnar epithelium which is a typical endometrial-type epithelium. The stroma underneath the epithelium was found to be cellular with characteristic endometriod features which did contrast with a fibrous paucicellular stroma underneath the mesothelium. A few cells which were scattered were also observed within the fibrous stroma as tends to be seen in a mesothelial inclusion cyst. The tumour was adjudged to have morphological features diagnostic of anaplastic seminoma. Immunohistochemistry studies also showed that the seminoma stained positively for CD119, PLAP, and OCT4. However, the immunohistochemistry studies of the tumour showed negative staining for EMA and CK. The histological examination also showed evidence of metastatic seminoma in the spermatic cord. Immunohistochemistry studies also showed that the endometrioid epithelium had expressed strong positive staining for oestrogen receptors (ER), progesterone receptors (PR), cytokeratin 7 (CK7), epithelial membrane antigen (EMA). Some of the cells the endometrioid cells stained positively for mesothelial markers calretinin and cytokeratin 5/6. The stromal endometrial cells stained positively for progesterone receptors (PR), and some of the stromal endometrial cells also stained positively for calretinin and endometrial stromal marker CD10. The mesothelium on immunohistochemistry stained negatively for EMA, and sex-steroid receptors, but they stained strongly positive for CK7 and mesothelial markers CK5/6 as well as for calretinin. A fibrous stroma that was beneath the mesothelium was found on immunohistochemistry study to contain some for CK5/6 positive, calretinin positive, and CK7 positive mesothelial cells. Zámecnik and Hoštáková [3] stated the following:

- Controversies exist regarding the pathogenesis of endometriosis and the main postulates related to the pathogenesis of endometriosis include (a) cell proliferation within Müllerian embryonic rests, (b) retrograde menstruation, (c) and coelomic metaplasia as iterated by Mai et al, [28] Sampson [29] and Meyer et al. [30]
- With regard to their reported case, they had not found any congenital Müllerian abnormalities or any persistent Müllerian duct remnants like appendix tests or paradidymis which could have supported the embryonic rest theory. Furthermore, considering the fact that their patient was a man, the retrograde menstruation pathogenesis postulate would be excluded. The endometrial-type epithelium of the patient had shown continuity with a mesothelial cell layer and that in their opinion, would give credence to the metaplastic pathogenesis postulate ascribed to endometriosis. It was also their opinion that the immunohistochemistry study characteristics of the epithelium with positive expression of oestrogen receptors (ER +), progesterone receptors (PRs), EMA, which is a typical endometrioid phenotype had retained in some cells an expression of mesothelial markers calretinin and CH5/6. They would interpret the

aforementioned pathology study findings as evidencing a residual mesothelial phenotype in view of the fact that metaplasia is a gradual process and as supportive of the endometriosis having originated from the mesothelium.

- Endometriosis in men tends to occur in patients who have a high serum oestrogen levels as a result of oestrogen treatment for adenocarcinoma of the prostate gland. It may be that the hormonal treatment does induce the development of steroid receptors emanating in endometrial differentiation. With regard to their patient, he had not been treated with oestrogens and he had never had steroid treatment.
- In men, endometriosis had previously been reported in the urinary bladder, the prostate gland, seminal vesicles, retroperitoneum, epididymis, and paratesticular tissues. [16] [25] [31] to be associated with high serum levels of oestrogen
- They could not surely ascertain what had triggered the endometriosis in their reported case. Nevertheless, it may be that a local effect of seminoma and / or of a tumour-associated inflammatory process might have induced the formation of a mesothelial inclusion cyst in their reported case.

Test	Test Value	Reference Range
Testosterone	4.47 ng /ml	Male: 3.0 – 10.6
Prolactin	24.2 ng/ml	Male 3 – 25 Female: 5 – 35
Follicle stimulating hormone	32 IU/L	Male: 4 – 10 Female: 10 – 20
Luteinising hormone	11.8 MIU/ml	Male: 1.0 – 7.0 Female: 2.0 – 2.5

Illustration 3: Hormonal profile of the patient Reproduced from [32] Savitri M N, Surekha B H, Namrata B M, Nikhil M M Persistent Mullerian Duct Syndrome with Ovarian Endometriosis A Rare Case Report. Journal of Clinical and Diagnostic Research 2016 Feb; 10(2): ED14 – ED15 DOI: 10.7860/JCDR/2016/16691.7266 The Journal and the authors (the original source of the figures) maintains the copy right and any future reproduction of the figures would require copy right permission from the original source. Copy right Permission to reproduce this table was obtained from Dr. Hemant Jain, Editor in Chief/JCDR

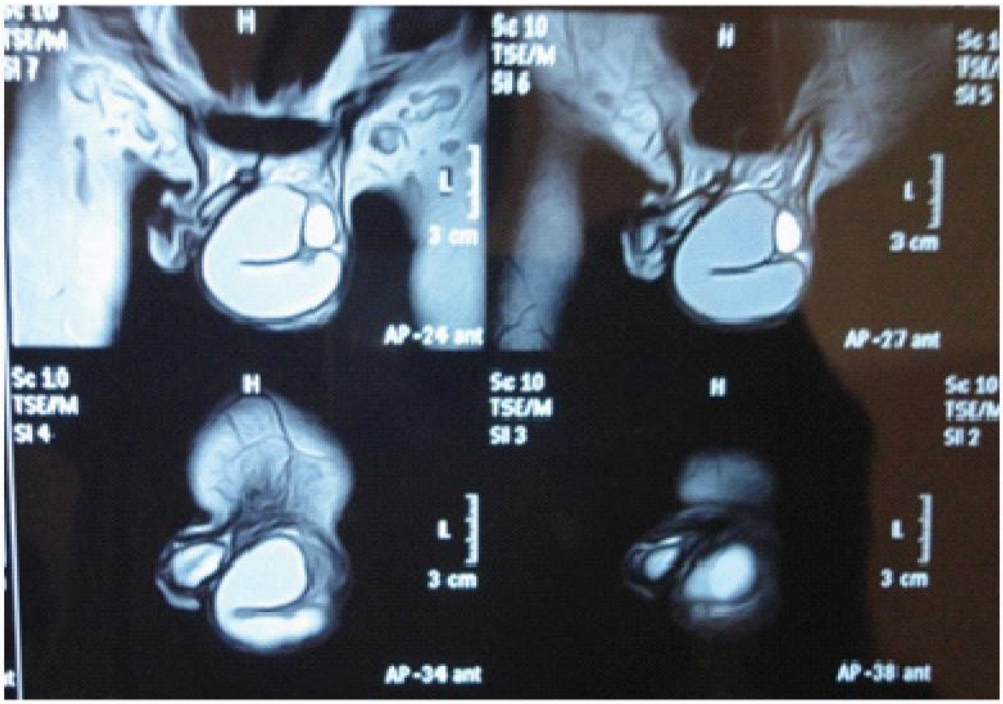
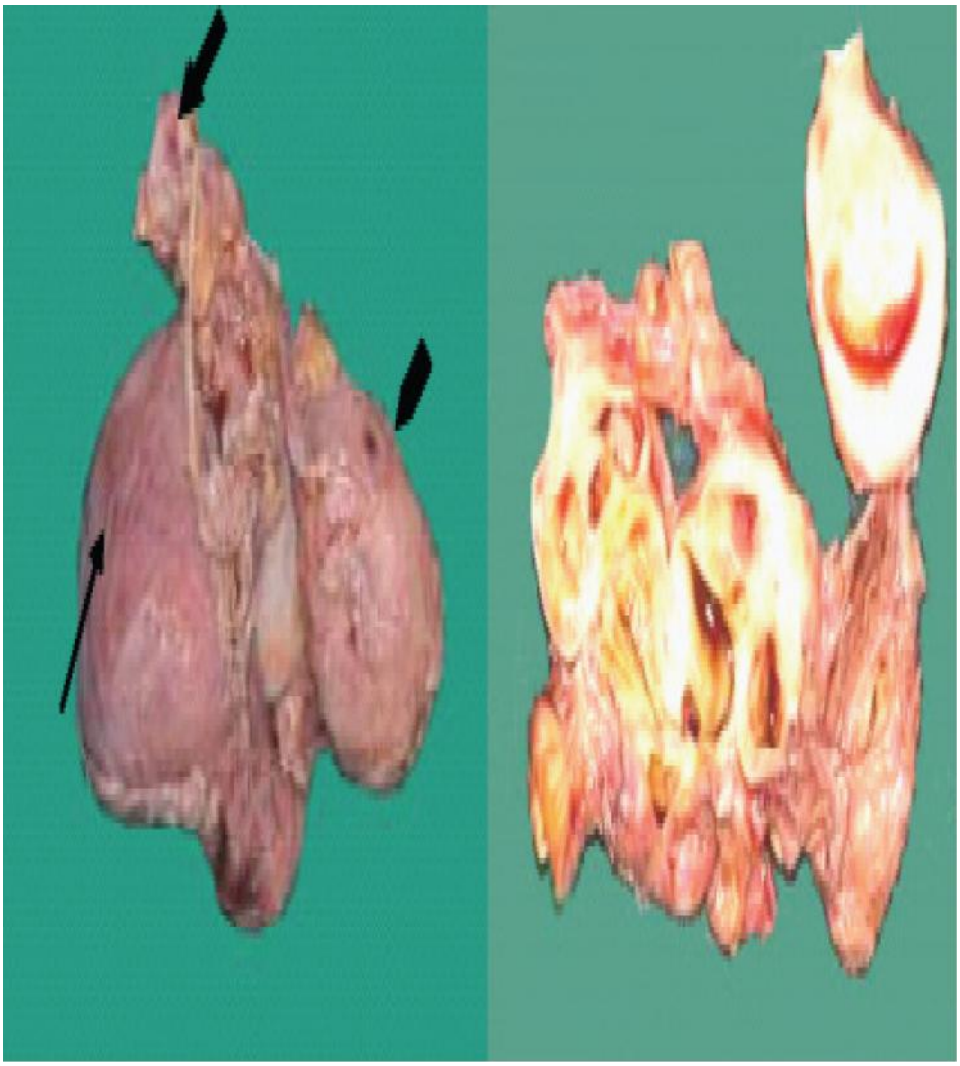


Illustration 4: MRI of left inguinoscrotal region (T1W1) show large multi loculated collection in between layers of tunica vaginalis suggestive of high cholesterol / fat content – with features of chylocele. Reproduced from: [32] Savitri M N, Surekha B H, Namrata B M, Nikhil M M Persistent Mullerian Duct Syndrome with Ovarian Endometriosis A Rare Case Report. Journal of Clinical and Diagnostic Research 2016 Feb; 10(2): ED14 – ED15 DOI: 10.7860/JCDR/2016/16691.7266 The Journal and the authors (the original source of the figures) maintains the copy right and any future - reproduction of the figures would require copy right permission from the original source. Copy right permission to reproduce this figure was obtained from Dr. Hemant Jain, Editor in Chief/JCDR



(A) ` (B)

Illustration 5a: Gross photograph shows cystic mass (thin arrow) tubular structure (thick arrow) and solid nodule (arrow head)

Illustration 5b: Cut surface of the specimen showing multi loculated cyst with altered blood and solid nodule with slit like space and attached tubular structure. Reproduced from: [32] Savitri M N, Surekha B H, Namrata B M, Nikhil M M Persistent Mullerian Duct Syndrome with Ovarian Endometriosis A Rare Case Report. Journal of Clinical and Diagnostic Research 2016 Feb; 10(2): ED14 – ED15 DOI: 10.7860/JCDR/2016/16691.7266 The Journal and the authors (the original source of the figures) maintains the copy right and any future reproduction of the figures would require copy right permission from the original source. Copy right permission to reproduce this figure was obtained from Dr. Hemant Jain, Editor in Chief/JCDR

Savitri et al. [32] reported a 19-year-old young man who presented with a 15 days history of left hemi-scrotal swelling. He had a history of cryptorchidism for which he underwent orchidopexy prior to his presentation. He was found on examination to have bilateral gynaecomastia and left inguinoscrotal swelling otherwise his examination was normal. His serum testosterone and prolactin levels were within normal range but his serum follicle stimulating hormone (FSH) and luteinising hormone (LH) levels were raised at 32 IU/L (normal range 4- 10 IU/L for male and 10 -20 for female) and 11.8 MIU/ml (normal range 1.1 – 7.0 MIU/ml for male and 2.0 to 2.5 MIU/ml) respectively (see illustration 3). He had ultrasound scan of the inguinal area and scrotum and the features were adjudged to have been consistent with liquefaction of the left testis which had been replaced by a hypo-echoic collection that was interpreted as suggestive of an abscess / pyocele of the testis. He had magnetic resonance imaging (MRI) scan which showed a big multi-loculated cystic collection between the layers of the tunica vaginalis which was adjudged to be suggestive of high cholesterol / fat content that represented chylocele. He underwent exploration of the inguinal canal expecting to find pus; however, a solid and cystic mass associated with an attached tubular structure was encountered and excised (see illustration 4). Macroscopic examination of the specimen showed a predominantly cystic mass together with a solid grey white area and a tubular structure. The cystic mass was found to measure 10 cm x 8 cm x 2 cm and the solid grey white nodule was found to measure 6 cm x 4 cm x 1.5 cm. The attached tubular structure was noted to have measured 10 cm x 2 cm (see illustration 5a). Furthermore, upon sectioning of the specimen a multiloculated structure which was filled with altered blood and the adjacent solid area was noted to have a slit-like space as well as the attached tubular

structure. Microscopic examination of the solid specimen revealed endometrial glands that had been embedded within compact stroma that was associated with surrounding normal myometrial tissue (see illustration 6a). Microscopic examination of multiple sections of the tubular structure revealed fallopian tube mucosa which had been composed of many delicate plical folds (see illustration 6b). Microscopic examination of sections of the cystic mass revealed ovarian tissue with cystic follicles, endometrial glands as well as stroma containing hemosiderin laden macrophages within the stroma of the ovary which was adjudged to represent ovarian endometriosis (see illustrations 7a and 7b). Despite a thorough search of the specimen there was no evidence of a malignant lesion and there was no evidence of testicular tissue and based upon the aforementioned pathological findings a diagnosis of persistent Müllerian duct syndrome (PMDS) with ovarian endometriosis was made. The patient was lost to follow-up. Savitri et al. [32] stated the following:

- Persistent Müllerian Duct Syndrome (PMDS) is an uncommon form of internal male pseudo-hermaphroditism which is characterized by presence of derivatives of Müllerian ducts including the fallopian tubes, uterus, and upper part of the vagina in a genotypic (46 XY) male.
- Odi et al. [33] had suggested that persistent Müllerian duct syndrome develops as a result of lack of anti-Müllerian hormone or due to defective functioning of anti-Müllerian hormone type II receptors [1]
- Sichani et al. [34] had stated that persistent Müllerian duct syndrome (PMDS) could occur sporadically or it may occur as an inherited X-linked autosomal dominant or autosomal recessive patterned condition
- Suresh et al. [35] as well as Prakash et al. [36] had stated that Nilson was the first to describe persistent Müllerian duct syndrome (PMDS) in 1939.
- Their review of the literature had shown that 150 cases of persistent Müllerian duct syndrome (PMDS) had been reported so far in the literature up to the time of publication of their paper in February 2016 and majority of these cases had been reported in Western Europe and Middle East male adults. Furthermore their case was the first case of ovarian endometriosis in a patient with persistent Müllerian duct syndrome (PMDS) and that their literature search had not revealed any previously reported cases.

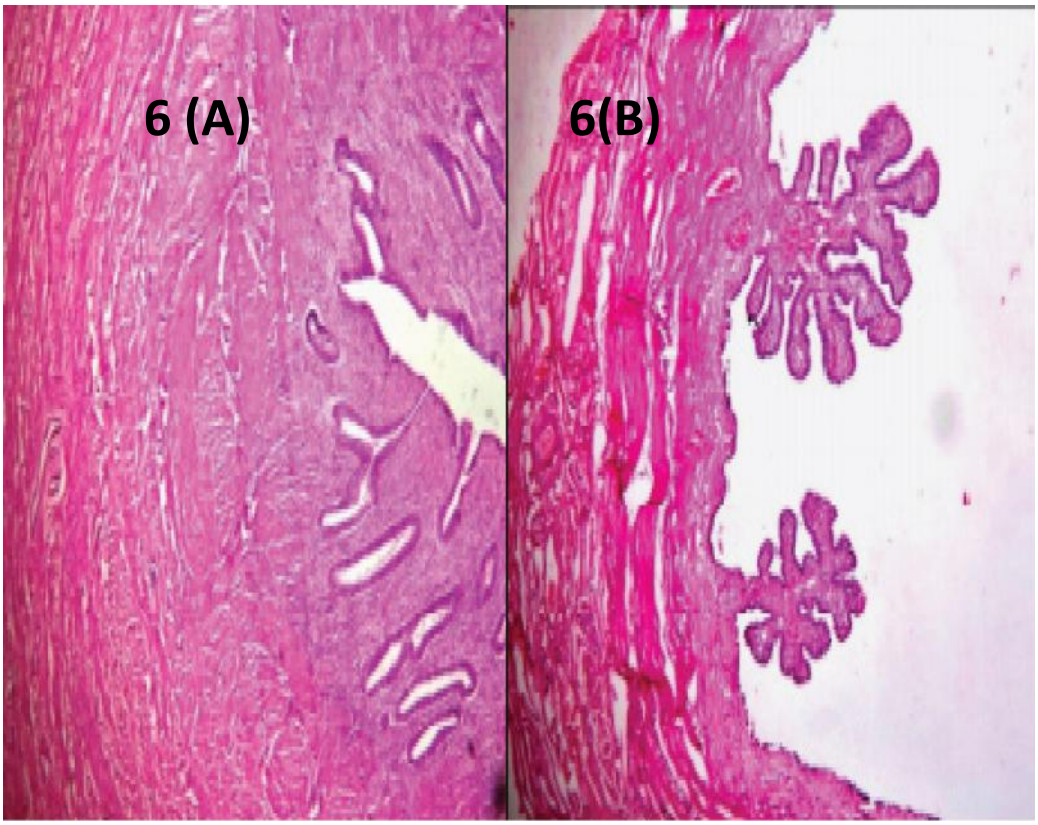


Illustration 6a: Photomicrograph of solid nodule showing endometrial glands in compact stroma and myometrium (H&E 100X). **Illustration 6b** Photomicrograph of tubular structure showing fallopian tube mucosa with plical folds (H&E 400X) Reproduced from: [32] Savitri M N, Surekha B H, Namrata B M, Nikhil M M Persistent Mullerian Duct Syndrome with Ovarian Endometriosis A Rare Case Report. Journal of Clinical and Diagnostic Research 2016 Feb; 10(2): ED14 – ED15 DOI: 10.7860/JCDR/2016/16691.7266 The Journal and the authors (the original source of the figures) maintains the copy right and any future reproduction of the figures would require copy right permission from the original source. Copy right for this figure was obtained from Dr. Hemant Jain, Editor in Chief/JCDR

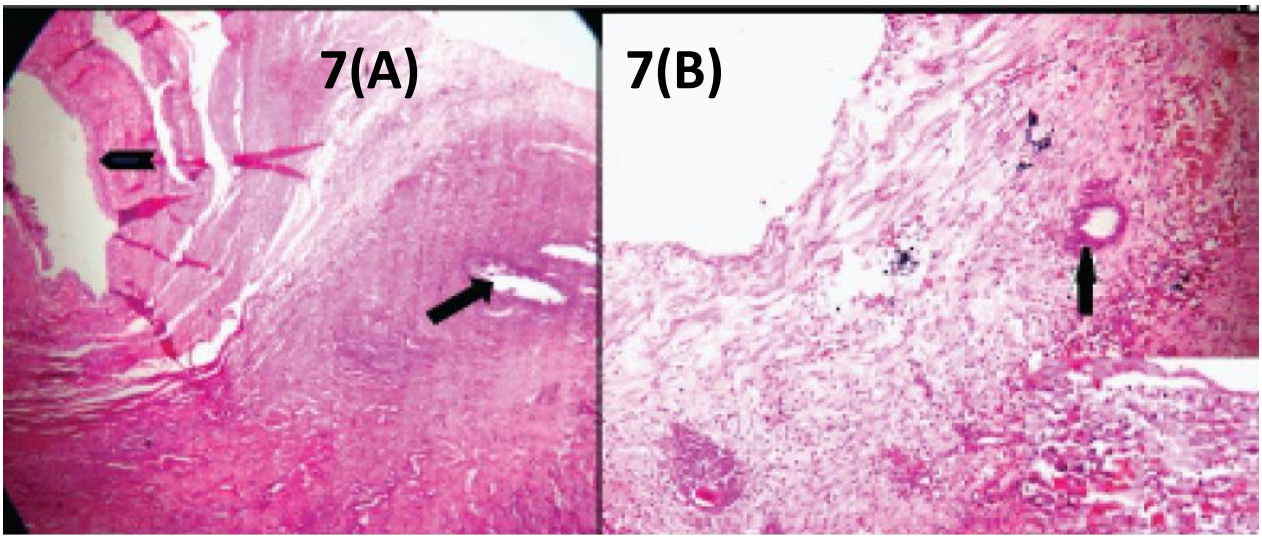


Illustration 7a: Photomicrograph of cystic mass showing cystic follicle of ovary (arrow head) with presence of endometrial stroma (arrow). **Illustration 7b:** Photomicrograph of cystic mass showing endometrial gland. Inset shows hemosiderin laden macrophages. Reproduced from: [32] Savitri M N, Surekha B H, Namrata B M, Nikhil M M Persistent Mullerian Duct Syndrome with Ovarian Endometriosis A Rare Case Report. Journal of Clinical and Diagnostic Research 2016 Feb; 10(2): ED14 – ED15 DOI: 10.7860/JCDR/2016/16691.7266 The Journal and the authors (the original source of the figures) maintains the copy right and any future reproduction of the figures would require copy right permission from the original source. Copy right for this figure was obtained from Dr. Hemant Jain, Editor in Chief/JCDR

Illustration 8: A table of reported cases of endometriosis in the male including the sites / organs involved

Reference	Age, site/organ	History of previous treatment with oestrogen
Oliker & Harris [12] 1971	Age details not available to author, urinary bladder	Yes he had had oestrogen therapy
Pinkert et al. [13] 1979	73 years, urinary bladder at vesico-ureteric junction causing hydronephrosis	Had radical prostatectomy & orchidectomy & oestrogen therapy for many years for adenocarcinoma of prostate
Schrodt et al. [14] 1980	73 years, urinary bladder at right vesico-ureteric origin causing right hydronephrosis	History of adenocarcinoma of prostate and was on oestrogen for 5 years
Beckman et al [2] 1985	78 years, in the region of the urethral crest	There was a long history of oestrogen therapy
Martin & Hauck [15] 1985	83 years, lower abdominal wall	History of 25 mg TACE treatment for 10 years for presumed adenocarcinoma of prostate which was subsequently confirmed by histological examination of resected prostate and TACE treatment continued until diagnosis of endometriosis made
Young & Scully [16] 1986	82 years, para-testicular mass	The patient had undergone oestrogen treatment for adenocarcinoma of the prostate gland for a period of time preceding the diagnosis of endometriosis.
Simsek et al. [17] 2012	49 years, left inguinal canal diagnosed pre-operatively as recurrent inguinal hernia	He had a history of infertility but no previous oestrogen therapy
Gonzalez et al. [22] 2014	52 years, right inguinal canal at previous herniorrhapy site	No history of oestrogen therapy in documentation
Jabr & Mani [23] 2014	52 years, and an extra-peritoneal soft tissue mass which measured 2.6 cm x 2.3 cm in the pelvis	He had a history of cirrhosis secondary to hepatitis and a past history of having had banding of oesophageal varices and a right inguinal herniorrhaphy twice over a

	adjacent to his urinary bladder and which had extended into his right inguinal canal	period of two years. No documentation of a history of oestrogen therapy.
Giannarini et al. [24] 2006	27 years, intra-scrotal epididymal cystic mass	No history of treatment with oestrogen
Fukunaga [25] 2012	69 years, who was found to have a multiloculated cyst in his left para-testis that measured 5.2 cm x 3.1 cm x 3.0 cm.	He had been having hormonal treatment for adenocarcinoma of prostate for 9 years
Taguchi & Enamoto [26] 2012	74 years, presented with haematuria. He had cystoscopy which revealed a 3 cm solid mass situated over the left ureteric orifice in the urinary bladder. He had computed tomography (CT) scan which showed a mass that was reported to be suspicious of an invasive urinary bladder cancer.	He had undergone radical prostatectomy for localized carcinoma of the prostate gland 5 years preceding his presentation with visible haematuria. He had received treatment with leuprorelin and ethynylestradiol for 5 years as adjuvant treatment for a metastatic Gleason 4 + 3 = 7 with tertiary pattern 5 adenocarcinoma of the prostate gland which was staged pT3bN1M0. His serum PSA had been undetectable.
Zamanecnik & Hostakova [3] 2013	46 years old, endometriosis as part of right tunica albuginea cystic lesion found associated with seminoma of right testis in an orchidectomy specimen for testicular tumour	There was no history of oestrogen therapy.
Savitri et al. [32] 2016	19 years, left hemi-scrotal swelling. He was found on examination to have bilateral gynaecomastia and left inguinoscrotal swelling, Ultrasound scan showed liquefaction of the left testis which had been replaced by a hypo-echoic collection that was interpreted as suggestive of an abscess / pyocele of the testis. He had magnetic resonance imaging (MRI) scan which showed a big multi-loculated cystic collection between the layers of the tunica vaginalis which was adjudged to be suggestive of high cholesterol / fat content that represented chylocele. Exploration of the inguinal canal revealed a solid and cystic mass associated with an attached tubular structure was encountered and excised	He had a history of cryptorchidism for which he underwent orchidopexy prior to his presentation. He had bilateral gynaecomastia His serum testosterone and prolactin levels were within normal range but his serum follicle stimulating hormone (FSH) and luteinising hormone (LH) levels were raised at 32 IU/L (normal range 4- 10 IU/L for male and 10 -20 for female) and 11.8 MIU/ml (normal range 1.1 – 7.0 MIU/ml for male and 2.0 to 2.5 MIU/ml) respectively, There was no history of oestrogen therapy

To the knowledge of the author only fourteen cases of endometriosis in the male have been reported in the literature in patients whose ages have ranged between 19 years and 83 years and these have been most commonly reported in the 70 to 80 years age group (see illustrations 8 and 9a and 9b). It could be conjecturally be argued that there is the possibility that endometriosis in the male may be underreported

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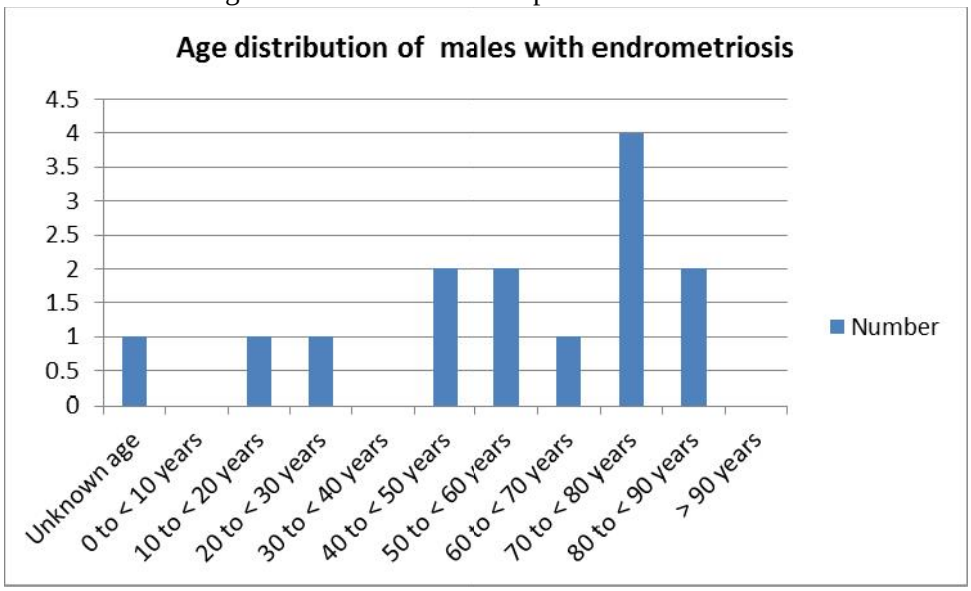
especially because men have not been known to have endometriosis and that a high index of suspicion does not exist in the minds of pathologists who examine resected lesions of patients who have subsequently had haematuria after being on treatment with oestrogen for a long time for adenocarcinoma of the prostate gland.

Illustration 9 a: Age distribution of males reported to have endometriosis

Age groups	Number
Age group not available to author	1
0 to < 10 years	0
10 to <20 years	1
20 to < 30 years	1
30 to < 40 years	0
40 to < 50 years	2
50 to < 60 years	2
60 to < 70 years	1
70 to < 80 years	4
80 to < 90 years	2
90 years and above	0

Illustration 9 b: Age distribution of males reported to have endometriosis



Conclusions

Endometriosis is rare but can occur in the male patient therefore clinicians should have a high index of suspicion for the disease. Microscopy examination and immunohistochemistry studies are required to establish the correct diagnosis of endometriosis in the male and this would enable the surgeon to undertake the appropriate surgical management to ensure complete excision of the lesion.

Conflict of Interest: None

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