SKILL AND TALENT

Vesiculectomy with laparoscopic partial prostatectomy in the treatment of primary adenocarcinoma of the seminal vesicle with carcinomatous transformation of the ejaculatory duct

J.C. Angulo a,∗, I. Romero a, P. Cabrera a, J. González a, J.M. Rodríguez-Barbero b, C. Núñez-Mora a

a Servicio de Urología, Fundación para la Investigación Biomédica, Hospital Universitario de Getafe, Universidad Europea de Madrid, Madrid, Spain
b Servicio de Anatomía Patológica, Fundación para la Investigación Biomédica, Hospital Universitario de Getafe, Universidad Europea de Madrid, Madrid, Spain

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Seminal vesicle; Clear cell adenocarcinoma; Surgical treatment; Partial prostatectomy; Central area; Seminal vesicle cyst; CA-125; Azoospermia

Abstract
Introduction: Primary adenocarcinoma of the seminal vesicle is an extremely rare condition. Some cases have been described in relation to congenital seminal vesicle cysts, which is often also associated with agenesis or ipsilateral renal disgenesis. The rareness of this type of lesions makes it difficult to plan a regulated surgical approach for them, although they are often treated by simple exeresis or exenteration, depending on their stage at the beginning.

Materials and methods: We present a new surgical technique that consists of radical vesiculectomy associated with laparoscopic partial prostatectomy (total segmentary) of the central area to successfully treat primary seminal vesicle adenocarcinoma in a young man who was diagnosed through an azoospermia study.

Results: A study of the scan (MRI) with diffusion and the transrectal biopsy of the mass allowed us to make a thorough preoperative evaluation of the case, confirming the malignity and precociousness of the lesion. The laparoscopic approach allowed us to perform a pelvic lymphadenectomy and transperitoneal exeresis, including the central prostate area and suture of the posterior face of the urethra at the height of the apex of the prostate. The wall of the seminal cyst lesion confirmed infiltrating clear cell adenocarcinoma and non-invasive adenocarcinoma in the prostate segment of the central gland in the light of the ejaculatory conduct with ‘‘in situ’’ growth. Thus, the surgical specimen allowed radical exeresis with negative margins, guaranteeing minimally invasive surgery with preservation of continence and erection.

∗ Corresponding author.
E-mail addresses: jangulo@futurnet.es, jangulo.hugf@salud.madrid.org (J.C. Angulo).

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**Introduction**

Primary tumors of the seminal vesicle (adenocarcinoma, sarcoma and lymphoma) are very rare lesions, which are often initiated by nonspecific symptoms. Primary adenocarcinoma of the seminal vesicle usually occurs in patients aged more than 50 years. Detection by means of transrectal ultrasound, contrast CT or MRI of solid papillary mass and elevation of serum CA-125 suggests the presence of these lesions, of which, at most, only several dozen cases have been reported with good clinical and histopathological correlation.

The histopathological pattern that is characteristic of seminal vesicle adenocarcinoma comprises fine structures of complex anastomosing papillary branching, which consists mainly of clear cuboidal cells or partially granular cells with some "hobnail" cells of round nuclei and occasional mitotic figures. In the light, these cells often produce Alcian blue epithelial mucin and positive mucicarmine. Immunohistochemical staining with CA-125 is typically positive while CEA, AFP, PSA and PAP are negative. This pattern helps to exclude other undifferentiated lesions of prostate or rectal origin with secondary infiltration of the seminal vesicle.

The treatment of these lesions is determined by the level of extension to nearby organs and by the patient’s age. Already in 1967, Smith et al. recommended the radical inclusion of the prostate in the specimen, because the ejaculatory duct is often affected. It therefore seems reasonable to propose radical prostatevesiculectomy in these patients when the tumor appears localized, especially when prostate invasion is suspected. However, some cases were treated with isolated vesiculectomy with good clinical evolution. At times it was also necessary to consider cystectomy. Radiation therapy seems to have been indicated as palliative treatment in very advanced cases with very bad prognosis. Other authors prescribed adjuvant therapy with
Fig. 1 Retrovesical cyst with parietal exophytic growth in MRI. A. Transverse T2 showing intracyst protein content. B. ADC map parietal lesion suggestive of malignancy. C. Sagittal T2-weighted MRI showing relationship between the mass, the ejaculatory duct and the central prostate, as well as the rejected urethra and bladder.

antiandrogens or even orchiectomy, although there is no scientific basis for this.

Materials and methods

We report the case of a 29-year-old male who consulted for azoospermia and isolated microhematuria. An ultrasound revealed a seminal vesicle cyst with a 5 cm diameter, with an appearance of partially solid contents in its interior and absence of the right kidney. Palpation revealed a small prostate, with normal consistency and a palpable soft mass on it. The testes and vas deferens were normal. Cystoscopy confirmed the absence of the right ureteral meatus.

An abdominal CT scan revealed a structure of approximately 15 mm in diameter, in right para-aortic position, which could be related to dysplastic kidney. In the lower pelvis, cranially with respect to the prostate, we observed a preferably cystic ovoid lesion with a diameter of approximately 6 cm, but with a 2 cm solid area next to the wall. Given the renal dysplasia and the associated ureteral agenesis, these findings are consistent with cystic lesion dependent on seminal vesicle associated with congenital renal anomaly.

The pelvic MRI revealed retrovesical cystic formation in theoretical location of the seminal vesicles, measuring 5.9 cm × 7.4 cm. By means of a diffusion map (ADC), the study corroborated the existence of a solid papillary lesion, with behavior suggestive of malignancy in the anterolateral wall of the cyst, measuring 1.8 cm (Fig. 1). With the clinical impression that this was a malignant papillary lesion that had developed on the wall of a seminal vesicle cyst associated with renal agenesis and right renal dysplasia, we performed a transrectal ultrasound guided biopsy, which revealed atypical glandular proliferation, although it was not possible to properly evaluate the infiltrative nature of the lesion in the sample submitted. The neoformation comprised tubules of various sizes and forms, covered with very flattened cells with large nuclei and dense and irregular chromat in that protruded into the tubular lumen and stained CK7 intensely and CA-125 focally; CK20, PSA and PAP were negative. The most cellular portion of the sample expressed a proliferative Ki-67 index and overexpressed p53 in more than 25% of the cells. Given these findings, we ruled out prostate or urothelial origin and suspected seminal vesicle origin or that of other Müllerian remnants. Urine cytology was negative, and serum PSA was 0.46 ng/ml.

With the suspicion that it was an adenocarcinoma on the wall of the seminal vesicle cyst, subjected to degeneration on malformative embryonic area, we considered the possibility of performing laparoscopic vesiculectomy, and also decided to perform pelvic lymphadenectomy and excision of the central portion of the gland to completely resect the margin of the ejaculatory ducts (Fig. 2).

Surgical technique

By means of a laparoscopic transperitoneal approach using 4 trocars, we opened the Douglas pouch; we identified, dissected, ligated and sectioned both vas deferens; we dissected the cyst and the seminal vesicles up to the prostatic base, subsequently advancing the dissection of the ejaculatory duct up to the prostatic urethra where we sectioned it. The specimen included the central portion of the prostate for histopathological analysis. We proceeded to extract the specimen in a laparoscopic bag and then closed the ventral surface of the prostatic urethra with 3-0 Monocryl continuous suture; we performed hemostasis of the surgical site, removed the trocars and closed by plane (Fig. 2).

Histopathological diagnosis

The resected cystic lesion showed an irregular outer surface, covered by adipose tissue and both vas deferens severed at the surface. The specimen was accompanied by a nodular area of 2.5 cm × 1.5 cm in diameter, consisting of tissue with an elastic consistency that tallied with the central prostate area (Fig. 3). During the histopathological study, we opened the cyst and found a hematic deposit associated with irregular papillary growth of clear cells with large, hyperchromatic nuclei...
Vesiculectomy with prostatectomy for treating primary adenocarcinoma of the seminal vesicle

Fig. 2 Description of the vesiculectomy and partial prostatectomy technique. A. Retrovesical cyst dissection after opening the peritoneum and Denonvillers. B. Ligation and section of the lateral pedicles of the cyst. C. Opening of the prostate up to the urethra, including verumontanum and central prostate in the resection specimen. D. Closure of the prostatic urethra and surgical site after removal of the specimen.

Fig. 3 A. Topographical relationship between bladder and prostate with seminal vesicle cyst and ejaculatory duct. B. Macroscopic specimen of seminal cyst with vegetative neoformation and tissue corresponding to the central part of the prostate (on the right).
Fig. 4 Neoplastic lesion consisting of: A. adenocarcinoma with mixed tubular growth pattern, B. papillary and C. solid with predominance of clear cells (hematoxylin–eosin, 100×).

and prominent nucleoli, as well as moderate mitotic activity. In several areas the pattern was cribriform and/or tubular, becoming solid (Fig. 4). One tumoral area was calcified, suggesting its slow growth. In several areas, tubular structures of the neoplasia infiltrated the cyst wall, which comprised fibrous and muscular tissue; however, its surface was invasion-free and the remitted prostatic parenchyma that comprised the nodular area accompanying the cyst was also invasion-free. We found perineural and intraneural tumor invasion in the cyst wall. Likewise, the ejaculatory ducts had noninvasive malignant epithelial lining and the appearance of “intraductal” or in situ growth.

From a purely histological viewpoint, the neoplasia was conspicuous due to the predominance of cells with clear cytoplasm (Fig. 4), evidencing glycogen inside the cell in the PAS staining. The tumor was positive for cytokeratin AE1-AE3, cytokeratin 8-18 and cytokeratin 7. It was also intensely positive for epithelial membrane antigen (EMA) and racemase, and focally positive for CA-125. The proliferative index estimated with Ki-67 was moderate, around 15–20% in the most active foci. On the other hand, CK20, CK 8-18, CK 7, calretinin and PSA were negative.

It was therefore an invasive clear cell papillary adenocarcinoma that had grown on the wall of a seminal vesicle cyst. The margins were negative, but the ejaculatory duct inside the partial prostatectomy specimen, which corresponded to the central part of the prostate, presented diffuse adenocarcinoma in situ, which almost reached the verumontanum, but did not manifest in the urethral lumen. The cystic structure drained into a single ejaculatory duct, which in turn emptied at the height of the verumontanum. We included the entire central portion of the prostate in the specimen (Fig. 3). Retrospectively, in the MRI, we observed the presence of a large caliber ejaculatory duct, draining into the urethra and the actual urethra anterior, connected to the bladder and displaced by the mass (Fig. 1).

Results

Despite our definitive findings of intraductal extension and perineural invasion in the specimen, the patient received no adjuvant treatment, as the surgical margins were negative. Postoperative recovery was excellent, showing continence and conserved erectile function. Thirteen months after an exhaustive follow-up, the patient is disease-free and has not presented any sequela arising from the surgery, although he requires IPDE to maintain erectile function. During the clinical follow-up, we performed uroflowmetry, cystoscopy, urine cytology and MRI without pathologic findings.

Discussion

The persistence of Müllerian structures occurs when the paramesonephric ducts do not appropriately return in the male embryo, generating cysts in the prostatic utricle and/or Müllerian ducts.9 These remnants are often diagnosed as retrovesical mass in men in the second or third decade of life, when they produce hemospermia and ejaculatory duct obstruction. Some cases have been described of carcinomatous degeneration in the thickness of such cysts, such that both the seminal vesicles and the vas deferens empty into this cystic mass. These are usually adenocarcinomas of tubulopapillary clear cells, which bring to mind adenocarcinoma in the uterus or ovary.10–13

Clear cell adenocarcinoma has also been described in the prostate utricle of adolescents and young men, which appears as a solid mass and/or cyst in the prostate midline under the bladder neck and is connected to the posterior urethra by means of a narrow canalicular segment.14 This type of tumors has a common cellularity described as “hobnail”. It is not entirely clear whether they originate from Wolffian remnants, Müllerian remnants or paraurethral glands. The manner in which this type of rare lesions with poor prognosis is often diagnosed is because they produce
hematuria and obstructive symptoms of urinary flow or even urinary retention.\textsuperscript{15} Their usual immunohistochemical profile is racemase, positive CK7 and CK20 variable CA-125 and negative p63.\textsuperscript{16} From a histogenetic point of view, clear cell adenocarcinoma of the urinary tract is urothelial in origin, although there is some similarity with some Müllerian tumors, including female and very rarely male.\textsuperscript{16}

Conceptually, the neoplasia we treated in this young man at the end of the third decade of his life was a primary clear cell adenocarcinoma of the seminal vesicle with tubulopapillary growth. There are similar cases that are also based on congenital cysts of the seminal vesicle and are also associated with renal agenesis\textsuperscript{5} or with ureteral ectopia.\textsuperscript{17} It is most likely some kind of genetic disorder of unknown etiology that affects the genesis of this rare tumor. The dysgenetic condition manifested in the formation of the seminal cyst, of the coinciding dysplasia or agenesis, and its development at such an early age, supports the existence of a genetic-molecular explanation that is unknown today.

Despite the high proliferative rate of the tumor, its limited malignancy is determined by the fact that this degeneration occurs within the cyst, where it becomes focally invasive, although most of this lesion has a non-invasive parietal component that resembles intraductal carcinoma of the breast or ovary carcinoma \textit{in situ}. The involvement of the whole epithelium of the ejaculatory duct in the thickness of the central area of the prostate confirms this diffuse or multifocal nature of the disease, but diagnosed at an early stage, if it is compared with the few cases of seminal vesicle adenocarcinoma classically described.\textsuperscript{5}

This case exemplifies how it is possible to carry out a radical laparoscopic approach in this type of lesion, which must necessarily include pelvic lymphadenectomy and resection of the portion of prostate specimen housing the ejaculatory duct; in short, a total segmental partial prostatectomy. This ingenious situation, vesiculectomy with laparoscopic partial prostatectomy is a new technique described here for the first time, and which should be considered as the first choice in all the rare cases of seminal vesicle adenocarcinoma diagnosed early. The incidental appearance of this patient, who was studied for azoospermia, and the application of new methods of diagnostic imaging such as diffusion MRI allowed early diagnosis of the lesion and the approach of successful minimally invasive surgery of the tumor, including resection of the carcinomatous transformation of the ejaculatory duct.

**Conflict of interest**

The authors declare that they have no conflict of interest.

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**References**


