

· CYSTOMYOMA OF SEMINAL VESICLE*

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THE RARITY of this lesion warrants description and discussion of every case. To our knowledge, no such observation has been reported in the Anglo-American literature.

Case Report.—Hosp. No. 152951: M. L., white, male, age 66, was admitted to the Beth Israel Hospital, May 6, 1943, because of a palpable mass in his left lower abdominal quadrant, which had been noticed two months previously. It occasioned him no pain or discomfort and he was not aware of its presence. The mass had not increased in size since it was discovered. There had been no abnormal gastro-intestinal symptoms. Appetite had been good, bowels regular, no blood had been noticed in the stool. There had been no weight loss, no dysuria, nocturia, hematuria or urgency. Neither had there been any symptoms of loss of cardiac reserve, the patient having carried on his usual occupation without interruption.

Physical Examination: The patient was thin, lungs clear to auscultation and percussion, pulse 80, regular sinus rhythm, no murmurs heard, blood pressure 138/80. The abdomen was scaphoid, soft, not distended. The liver edge was palpable 5 cm. below the costal margin; it was sharp and smooth. The spleen was not palpable. In the left lower quadrant there was an irregularly, round, hard nodular mass, about 8 x 8 cm. in diameter. It was movable but not freely so, and not tender. Rectal examination revealed no intrinsic lesion. Bimanual palpation with one finger in the rectum and the other hand on the abdomen demonstrated that the mass extended into the pelvis, almost completely filling it. It was fixed and immovable in the pelvis, but pressure by the abdominal hand was transmitted through the mass to the rectal finger. The prostate was in normal position and of normal consistency.

Barium enema showed no intrinsic colonic lesion, but the rectum and sigmoid were pushed to the right by the mass filling the pelvis.

Laboratory Data: Red blood cells 4,600,000, hemoglobin 88 per cent, white blood cells 6,200, with 69 per cent polymorphonuclear leukocytes. Urine: Specific gravity 1.012, glucose negative, albumen faint trace, white blood cells 18-20 per H. P. F., numerous urates.

Clinical Diagnosis: The original diagnosis rested between carcinoma of the rectum, with pelvic involvement; and retroperitoneal tumor, possibly fibrosarcoma or neurofibroma.

Operative Pathology: May 11, 1943 (Dr. Standard): The sigmoid was pushed to the right by a retroperitoneal, lobulated, hard mass which filled the pelvis and the entire hollow of the sacrum, pushing the bladder anteriorly. Portions of the tumor were cystic and these cysts contained dark, thick, chocolate-brown fluid. One such cyst was located deep in the hollow of the sacrum close to the area of the seminal vesicles, and the left vas deferens seemed to emerge from it. The entire tumor was well-encapsulated and could be enucleated along its lines of cleavage. The mass was roughly 15 x 13 cm. in diameter (Fig. 1).

Operative Technic: Left midline muscle-splitting incision. Rectus fascia was split and the rectus muscle displaced laterally. Peritoneum entered. Small intestine packed

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off under the upper angle of the wound. The posterior peritoneum along the left lateral surface of the tumor was incised and the tumor enucleated from its retroperitoneal bed. There was some spillage of dark brown fluid during the enucleation. There was a sharp line of cleavage between the major portion of the solid tumor and the cystic portion (3 x 4 cm.) which lay deep in the pelvis and from which the left vas deferens arose. The vas was ligated and cut from the cyst. (The other vas, which was found in the specimen, was not recognized during operation). The bladder was identified anteriorly and a catheter inserted. Clear urine was evacuated when the procedure was completed. Posterior peritoneum resutured over bed of tumor and abdomen closed in layers.

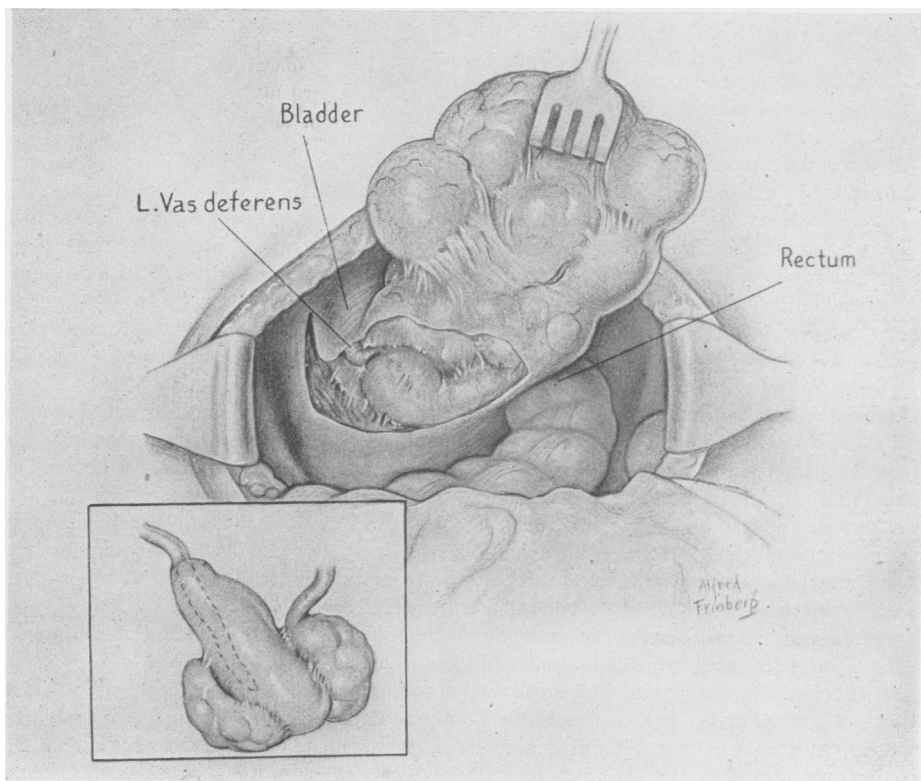


FIG. 1.—View of operative field (reconstructed), looking into the pelvis from above. Through a window in the posterior peritoneum one sees the left vas deferens and the adjacent cyst. The rectosigmoid has been pushed towards the right by the tumor. The retractor has pulled the myomatous mass out of the abdominal cavity.

(Insert)—The dotted line indicates the course of the ductus deferens within the cyst wall.

Postoperative Course: Uneventful. Wound healed *per primam*. Patient discharged 13 days after operation.

Follow-up: Three weeks after operation the patient reported noting "blood in his urine." The urine was brown; microscopically, it showed no red cells. The color of the urine was due to the same brownish, thick material which had been originally evacuated from the tumor. Only on two occasions, a few days apart, did the patient notice this discoloration of the urine.

Today, more than five months after operation, the patient is in perfect health.

Pathologic Examination.—Gross: No. 43184: The specimen was irregularly lobulated and firm. It consisted essentially of one somewhat kidney-shaped piece, 14 x 11 x 8

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cm., which was connected to another irregularly ovoid piece, 8.5 x 6 x 6 cm., by means of a pedicle-like structure. The specimen was surrounded by a hyperemic capsule in which many larger and smaller blood vessels were seen. Hyperemic membranous flaps were loosely attached to the outside. The one surface of the larger ovoid piece

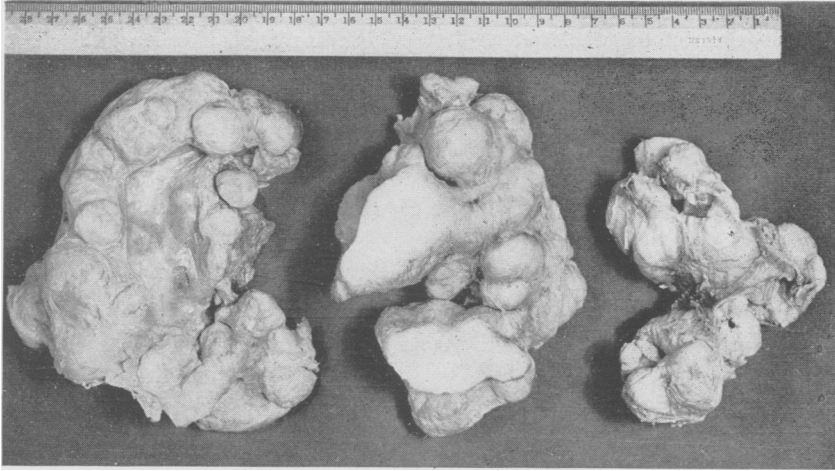


FIG. 2.—Gross specimen after fixation. Note the characteristic round myomatous nodules. The whitish areas in the middle specimen correspond to recently made cut-surfaces. The cavity is visible in the largest piece.

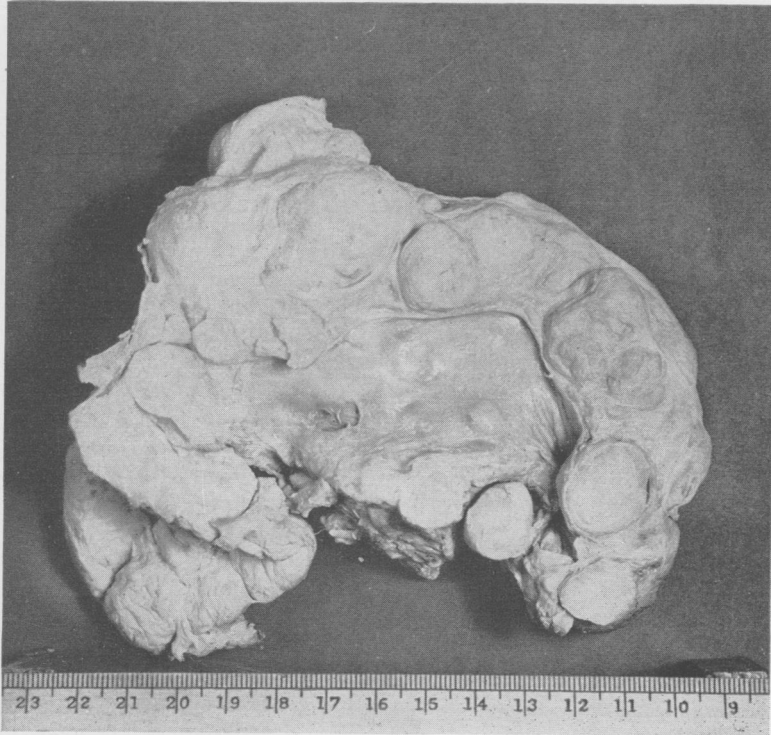


FIG. 3.—Close-up of largest piece. The high lights indicate the smooth lining in the cavity.

was flat, the other one irregularly convex. Both pieces were subdivided by grooves between which irregularly ovoid portions of different sizes protruded. A long section through the larger piece clearly showed many, more or less completely separated, nodules. The cut-surfaces could best be described as being very similar to those of myoma uteri.

Near the convex surface of the large piece a cavity was situated. Its volume was about one-fourth of the whole piece. It was crescent-shaped, it had a distinct smooth lining which was sharply demarcated from the surrounding tissue. The cavity had prolongations whose outlines corresponded to the contours of the whole piece. A transverse incision into the smaller piece opened a similar smaller cavity. Both cavities contained rather thin, dark reddish-brown fluid. (Figs. 2 and 3).

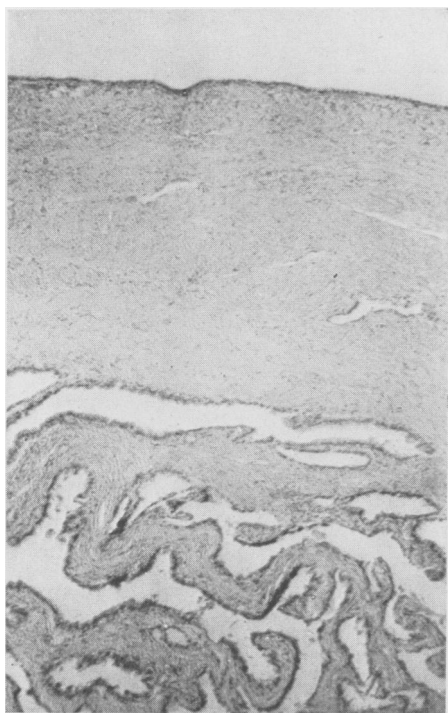


FIG. 4.—Characteristic structure of mucosa of seminal vesicle. The lining of the cyst appears in the upper portion of the specimen as a thin, dark contour.

A flat, soft, hollow specimen, 9 x 4 x about 1 cm., was received separately. It had torn prolongations and two ovoid thickenings. From an opening at one end dark, brownish fluid exuded. The lining of the cavity was smooth, partly pale, partly deep red. One of the prolongations contained a continuation of the cavity, about 1.2 cm. wide. The other protrusion was a thin, firm, cylindrical structure with very narrow lumen and firm elastic, obviously muscular, wall. Both looked like vas deferens. The outer aspect of the ovoid thickenings was suggestive of seminal vesicle; the cut-surfaces were characteristic (see insert to Fig. 1). The one seminal vesicle-like structure measured 3 x 1.5 x 1 cm., the other one 3.5 x 2 x 1.3 cm. They were separated by a piece of cyst wall, about 1 cm. wide. The cylindrical structures which entered this specimen really were the vasa deferentia. They were normal in thickness and slightly tortuous. They entered the wall of the cystic structure at an acute angle and ran mostly in the wall, with their lumina very near the surface. The average width of the lumen was 2.5 mm. About one centimeter from the one pole of one seminal vesicle the lumen was somewhat widened.

The probing of the ducts was done after fixation of the specimen. Through the one the probe passed easily, not through the other. There was, however, no reason to assume that the other duct did not communicate with the lumen of the cyst.

Microscopically, the picture of the solid tumor was that of fibromyoma. As is usual in such tumors, the ratio between muscle and connective tissue varied, and so did edema and inflammation, the latter not being severe anywhere. Elastic fibers were few except for the subepithelial layers. These layers ran parallel to the surface of the cavity and, thus, were more regularly arranged than the layers which formed the myoma proper. The subepithelial layers contained small amounts of light brown pigment. The cavity in the myoma was lined with a single layer of cylindrical cells. There were no glands. The cells did not contain mucin. Directly under the epithelium some large, obviously phagocytic, cells were situated.

In sections from different portions of the cystic specimen (Figs. 4 and 5) the microscopic pictures were those of seminal vesicle, ampulla or vas. The muscular walls of these structures were continuous. There was some pigment in epithelium and in muscle. Subepithelial phagocytic cells were present. There were a few calcifications in the mucosa. Sperm heads were found in the lumen. The characteristic subepithelial elastic structures were conspicuous.

Only four reports of such cases could be found; none in the English or American literature.

E. Emmerich, in 1910, reported the first one under the title "Enormous Cyst of Vas Deferens." The tumor was found accidentally during the autopsy of a 74-year-old man who had died of leukemia. No urologic symptoms had existed and no pelvic pain. Both seminal vesicles were normal, as were testicle and epididymis on both sides; the prostate gave the picture of ordinary

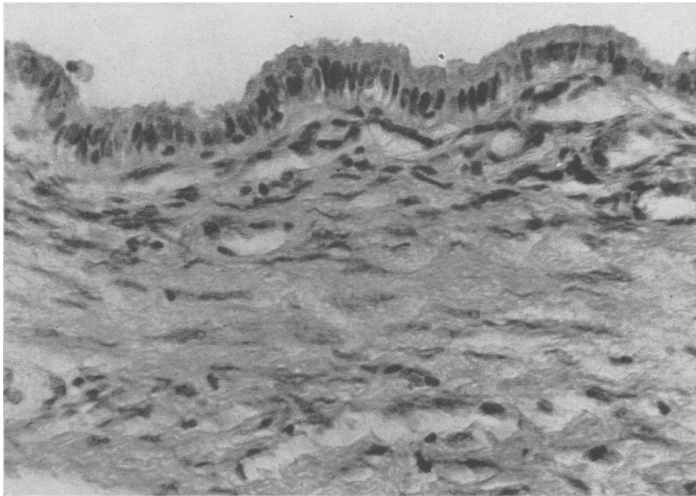


FIG. 5.—The cavity is lined with a single layer of regular high cylindrical epithelium.

hypertrophy. Right kidney and ureter were absent, there was no trace of a right ureteral opening in the bladder. The tumor (Fig. 6) arose from the left vas deferens, between ampulla and ejaculatory duct. It consisted of a round, firm, smooth, fist-sized mass which protruded from the small pelvis to the right of the midline. This mass was attached to the left ampulla towards the midline by means of a thick-walled hollow stalk, 6 cm. long. The lumen of the stalk communicated with the lumen of the ampulla and with the epithelium-lined cavity in the fist-sized mass. The wall of this cavity was only 2 mm. thick, that of the cylindrical portion was thicker; both consisted essentially of smooth muscle fibers. The epithelium was cylindrical or cuboidal. No spermatozoa were found.

Emmerich considered his case a cyst of the vas deferens, on the basis of a congenital malformation.

In 1912, Voelcker published a case of "Myoma of the Capsule of the

Seminal Vesicle"—this is the only case with clinical symptoms. Voelcker's patient was 56 years old; for more than 15 years he had suffered from pain in the perineum, he had consulted numerous surgeons, had been proctoscoped frequently, had been subjected to various procedures with no avail, and had, finally, become a morphine addict. On rectal examination a nontender swelling could be felt above the prostate. On bimanual palpation, in Trendelenburg position, under general anesthesia, it appeared as a movable, firm, smooth, spherical mass in the region of the seminal vesicle. The posterior wall of the bladder was pushed forward by this mass.

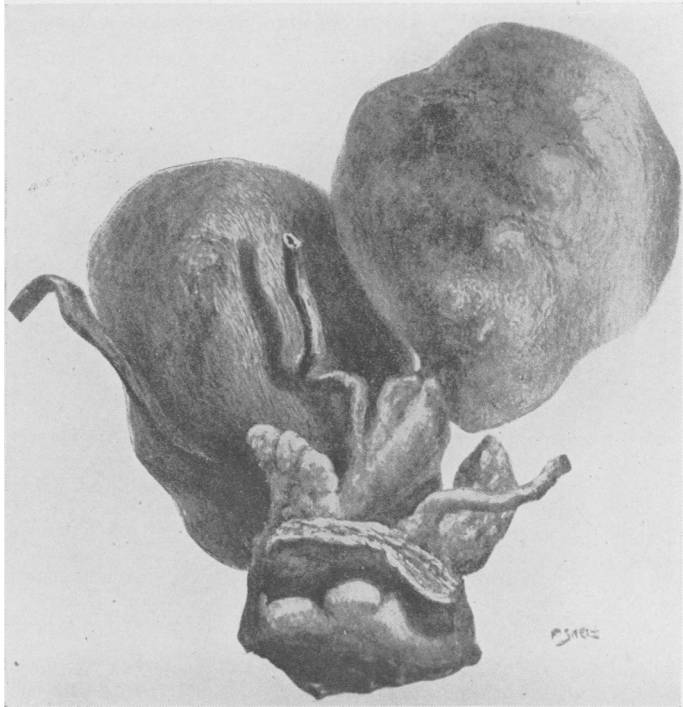


FIG. 6.—Emmerich's case. (*Zentralbl. f. allg. Path. u. path. Anat.*, 21, 673, 1910):
View from behind. The urinary bladder occupies the left half of the picture; the left ureter is normal. (The right ureter and kidney were absent in this patient). From the ampulla of the left vas deferens the tumor arises. Both seminal vesicles and the right vas deferens are normal.

Voelcker exposed the tumor by a paracoccygeal approach, severing part of the gluteus muscle, the tuberosacral ligament and the levator ani. After incision of the visceral pelvic fascia the rectum was pushed towards the midline and the smooth surface of the tumor came into view. Most of the tumor was shelled out; a small portion which was adherent to the bladder had to be left behind. The patient was discharged after three weeks. When seen six months later he was in perfect health.

The specimen was a fist-sized tumor which, grossly and microscopically, looked like an ordinary myoma. It contained a small cavity without epithelial

lining. Voelcker explained the tumor as a myoma originating from remnants of the müllerian duct. No illustration is given.

In the same year (1912) Ceelen described a huge "Fibromyoma of Seminal Vesicle." It was found, protruding retroperitoneally from the small pelvis, at the autopsy of a 67-year-old man who had died of cirrhosis of the liver. No mention is made of clinical symptoms. The tumor had the size of a baby's head; rectum and lower sigmoid were pressed against the sacrum. The thick-walled urinary bladder was pushed to the left and pressed against the anterior aspect of the tumor. The prostatic portion of the urethra was drawn out, the prostate, which was not hypertrophic, was elongated, with its posterior surface firmly pressed against the tumor. Both ureters were

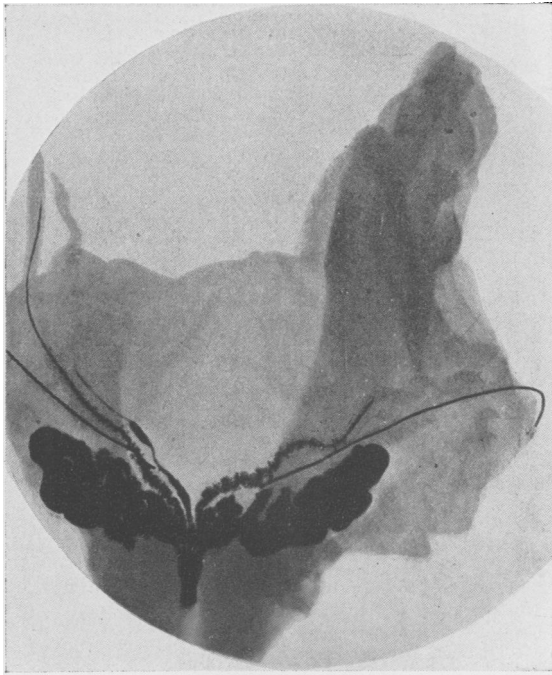


FIG. 7.—Voelcker. (*Chirurgie der Samenblasen*, 1912): Remnants of midportion of right müllerian duct. The genital tract is filled with collargol. Metal probes are inserted into the ureters.

compressed, their upper portions were as thick as a pencil. The right kidney was normal, the left kidney pelvis was distended, and the kidney substance only 1 cm. thick. The bladder mucosa was trabeculated, the ureteral openings could be probed easily. The colliculus was distinct. The right ejaculatory duct was missing, there were two lumina on the left side. The left seminal vesicle and vas deferens were normal. The right seminal vesicle, which appeared much smaller, was continuous with the tumor, the right vas was stretched over the tumor. The center of the mass was formed by a cavity with shaggy, partly necrotic wall; it contained dull grayish-red fluid with blood clots. The outer solid portion was 3-4 cm. thick, yellowish-gray and

grayish-white. The histologic picture was that of fibromyoma; some of the muscle cells contained pigment similar to that found normally in the muscle of the seminal vesicle. No illustration is given; and nothing is mentioned about histogenesis.

After a long interval (1930) a fourth case was reported, by Panà, under the title "Leiomyoma of Seminal Vesicle and Ductus Deferens, on the Basis of Malformation." This tumor also was found at autopsy. The 59-year-old man had suffered from tabes, syphilitic aortitis and chronic kidney disease. A pear-shaped retroperitoneal mass (8 x 7 x 5 cm., weighing 110 Gm.) was attached to the left seminal vesicle by means of a hollow pedicle, 9 cm. long, 1.5 cm. thick. The lumen of the pedicle was continuous with that of the seminal vesicle and with an epithelium-lined cavity in the pear-shaped mass. This cavity was lined with cuboidal epithelium; there were neither glands nor pigment. The gross and microscopic picture was that of myoma. There was a moderate right hydronephrosis caused by compression of the ureter. Panà interprets the pedicle as either left ampulla or hypoplastic left seminal vesicle. He considers the possibility of a müllerian remnant with hypertrophy of muscle.

Somewhat similar to these cases, but only as large as a cherry, was a cyst Luksch found at autopsy, in 1903, between the vasa deferentia of a 54-year-old man. The wall consisted of smooth muscle, the cavity was lined with a single layer of epithelial cells. Luksch considered it as müllerian.

COMMENT.—Large tumors in the female pelvis frequently are symptomless (myoma uteri, ovarian cystoma), because they may have grown slowly and need not interfere with important functions. The situation is the same with these tumors of the seminal vesicle or ampulla; thus, the fact that most of these tumors have been found accidentally is not so astonishing.

In Voelcker's case only, clinical symptoms (pelvic pain) were present. They appeared in the fourth decade, 15 years before operation. One may assume that the slowly growing myomatous tumor had then existed for some time previously. The three other tumors reported in the literature were found at autopsy of old men (74, 67 and 59 years). There is no way of knowing at what time of life they may have formed or had attained their final size. The same applies to our case, in which the detection of the tumor was hardly less accidental. One might point out the fact that these tumors, while based on malformation, became manifest late in life; but it would be futile to speculate about the causes of this seeming discrepancy.

These tumors resemble myoma uteri so closely that the idea of müllerian origin will present itself almost automatically. Emmerich speaks only about congenital malformation, not mentioning müllerian origin specifically; there is hardly any doubt in our mind that Emmerich's tumor is müllerian, in spite of Schneider's attempt at tracing it to ureteral muscle.

Voelcker, as mentioned, considered his tumor as a myoma originating from remnants of the müllerian duct. In his monograph on the surgery of the seminal vesicles Voelcker shows a beautiful roentgenogram of a müllerian

remnant in a 58-year-old man (Fig. 7). He feels that by filling specimens with contrast substance one might find more of these very rare malformations.

Ceelen gives no opinion concerning the genesis of his "Fibromyoma of Seminal Vesicle."

Luksch mentions the müllerian nature of the cyst, and Voelcker agrees with him.

Some of the smaller cysts of seminal vesicles (Schwarzwald) and some of the retrovesicular cysts, as mentioned by Priesel, are also explained as remnants of the midportion of the müllerian duct. The remnant of the proximal portion of the müllerian duct, the hydatid of Morgagni, is an almost regular finding; the remnant of the distal portion, the utriculus, belongs to normal anatomy, but the midportion usually disappears early in embryonic life, without leaving traces. Vestiges of it have been described (Priesel, Voelcker, Luksch). They are considered great rarities, and it is even more uncommon that they develop into a tumor, as in our case. It remains unknown under what exceptional conditions this happens.

There is no preference for the right or left side. The right kidney and ureter were absent in Emmerich's case. In Ceelen's case the right ejaculatory duct was missing, while two lumina were found on the left side. Panà's case showed no further malformations, neither did Voelcker's, nor did ours (as far as this can be determined in the living patient).

The tumor in our patient obviously originated from the point where ampulla and seminal vesicle meet.

SUMMARY

A large myomatous tumor, closely resembling uterine myoma, has been found attached to the ampulla or seminal vesicle of a 66-year-old man.

Remnants of the midportion of the müllerian duct are the source of these tumors.

Only four such cases have been reported, one of them in a living patient.

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