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PRESACRAL, RETRORECTAL DERMOID CYST IN A FEMALE PATIENT - CASE REPORT

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The retrorectal tumors are well defined, classified and understood pathological entities in the literature but in practice they represent very unusual and infrequent pathology. We are presenting a case from the group of dermoid congenital retrorectal cysts which at first manifested itself clinically as inflamatous retrorectal cyst that had spontaneously rupture in the postanal space with local (tumor, dolor, calor, rubor, function laesa and fluctuation) and systemic signs of infection (fever, rise temperature, leukocytosis). On physical examination there was typical postanal dimple which gives a picture of “double anus” on inspection. On DRE there was retrorectal soft tumor with compression of the anorectum. Diagnosis was confirmed with MRI and fistulography. After a palliative treatment for abscending cyst with incision, Penrose drainage and daily washings with antiseptic solutions the patient was transferred in specialized institution - the Clinic of Digestive surgery at the Medical Faculty in Skopje for definitive treatment. The operation was performed with the patient in jack-knife position with conventional preparing of the colon and prophylactic antibiotic regiment started preoperatively. An on table anoscopy was performed at first which sowed typical mammilla at the internal opening of the fistulous communication of the cyst with the rectum about 3 cm above the posterior crypt of Morgagni. We started with excision of the external opening, and preceded with whole excision of the pericystic granulomatous tissue about 14 cm in length till the presacral point. The fistulous communication was excised completely and the rectum was sutured in two layers with separate sutures. The wound was laid open and the patient was discharged on the 5-th postoperative day. About one month the wound was treated with daily washings with antiseptic solutions and after that one month with only water. After two mounts the defecation is normal, the wound is sealed and there are no signs of inflammation and secretion locally. The retrorectal tumors are difficult for treatment as well as for diagnosis where even punctional biopsy is not recommended so they should be treated in specialized institutions by experienced surgeons from the moment of diagnosis to the definitive surgical treatment.

Keywords: retrorectal cyst, excision, fistulous communication

PRESACRAL, RETRORECTAL DERMOID CYST IN A FEMALE PATIENT – CASE REPORT

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INTRODUCTION

The presacral or retrorectal space is a common site for embryologic remnants from which neoplasms and cysts may arise. This group of heterogeneous lesions is known as retrorectal tumors. The worldwide accepted classification of the retrorectal tumors is that proposed by Uhlig and Johnson is shown on **Table 1** [1, 4, 9, 7].The incidence is very rare. Cleveland Clinic reports 50 cases over a 55-year period. The Mayo Clinic estimates the incidence to be about 1 in 40000 hospital admissions. They reported 120 cases of which 66% were congenital, 12% neurogenic, 11% osseous and 11% were miscellaneous. Stewart and al. combined reports for a total of 301 retrorectal tumors of which 63% where congenital, 8% were inflammatory, 10% neurogenic, 7% osseous and 12% miscellaneous.

Congenital	Osseous
Developmental cysts (epidermoid, dermoid, and mucus-secreting cysts; teratoma)	Osteoma
Chordoma	Osteogenic sarcoma
Teratocarcinoma	Simple bone cyst
Adrenal rest tumor	Ewing's tumor
Anterior sacral meningocele	Chondromyosarcoma
Duplication of rectum	Aneurismal bone cyst
	Giant cell tumor
Inflammatory	**Miscellaneous**
Foreign body granuloma	Metastatic carcinoma
Perineal abscess	Liposarcoma
Internal fistula	Lymphangioma
Retrorectal abscess	Lipoma
Chronic infectious granuloma	Fibroma
	Fibrosarcoma
	Leiomyoma
	Leiomyosarcoma
	Hemangioma
	…

Table1

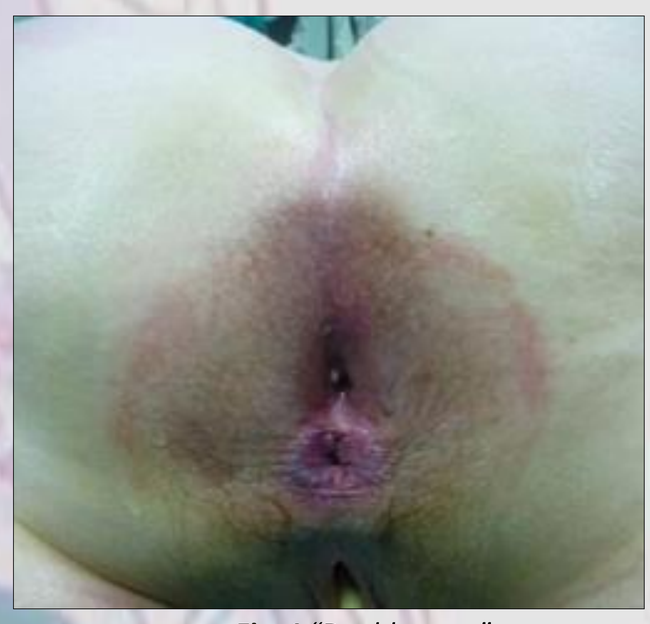

Fig. 1 *"Double anus"*

The symptoms of retrorectal tumors are mostly related to the size and complications such as malignisation [6] and infection. They are mainly : **pain**, local and systemic **signs of infection** and signs of **recurrent perianal suppuration**. Interference with pelvic outlet may lead to **constipation**, **incontinence** or **dystocia** [15]. **Disturbances to the bladder and urinary function** can be due to damage to the innervation, pressure on the bladder, urethra or ureters. **CNS manifestations** in form of headaches and recurrent episodes of meningitis are characteristic for anterior sacral meningocele.

Examination begins with **inspection** of the perineal area where one should look for characteristic **postanal dimple**. On **DRE** solid mass overlain with intact mucosa should be well recognized, whereas cystic lesions may be felt as mucosal folds and may be missed if they are not infected. The examination should proceed with: **plain film**, **sygmoidoscopy** and **CT scan** or **MRI**. **Barium enema** and **fistulography** then can be indicated. **Endorectal ultrasonography** is a very sensitive method in assessing the rectal wall involvement and pelvic flour muscle invasion.

The biopsy is only indicated if the lesion is inoperable because if the lesion is solid spreading of malignant cells may occurs, if the lesion is cystic infection may be spread and with anterior sacral meningocele meningitis my occurs.

Once the retrorectal tumor is diagnosed it should always be removed usually trough posterior approach with the patient in prone jack-knife for low lesions or infected cysts [8, 14] or abdominal approach with the patient in the lithotomy position for high lesions (above S4 on the imaging technics or when the upper border cannot be assessed on DRE) [1, 2, 3]

Epidermoid and dermoid cysts belong in the group of the developmental cysts as a part of the congenital lesion that account for more than 50% of all retrorectal tumors The majority of the developmental cysts is asymptomatic and may be missed on rectal examination due to low tension in the cyst. Epidermoid and dermoid cysts are result from defective closure of the ectodermal tube which results in inclusions of skin with or without accessory appendages. Both are lined with stratified squamous epithelium, well circumscribed with thin layer of connective tissue and fill with tick yellow-green fluid. The difference is that epidermoid cysts have no skin appendages. There is 30% rate of infection presenting as either as retrorectal abscess or mistakenly diagnosed as perianal fistula.

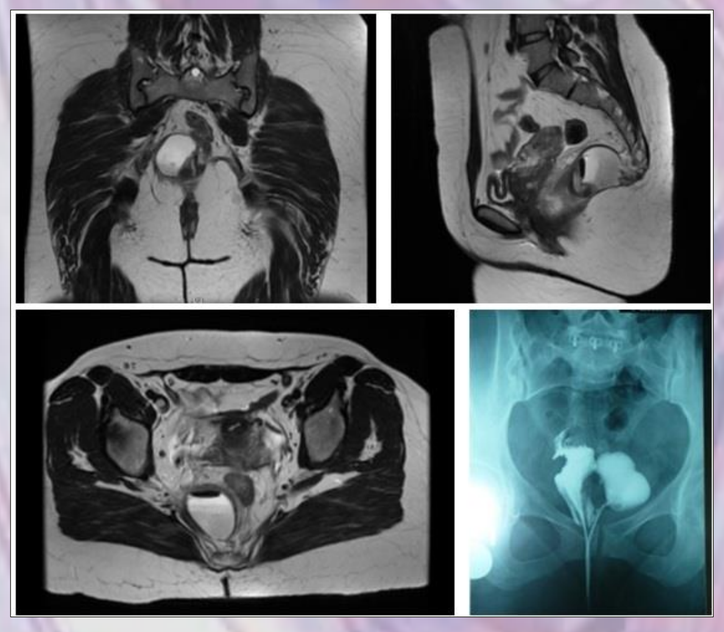

Fig. 2 *MRI and fistulography*

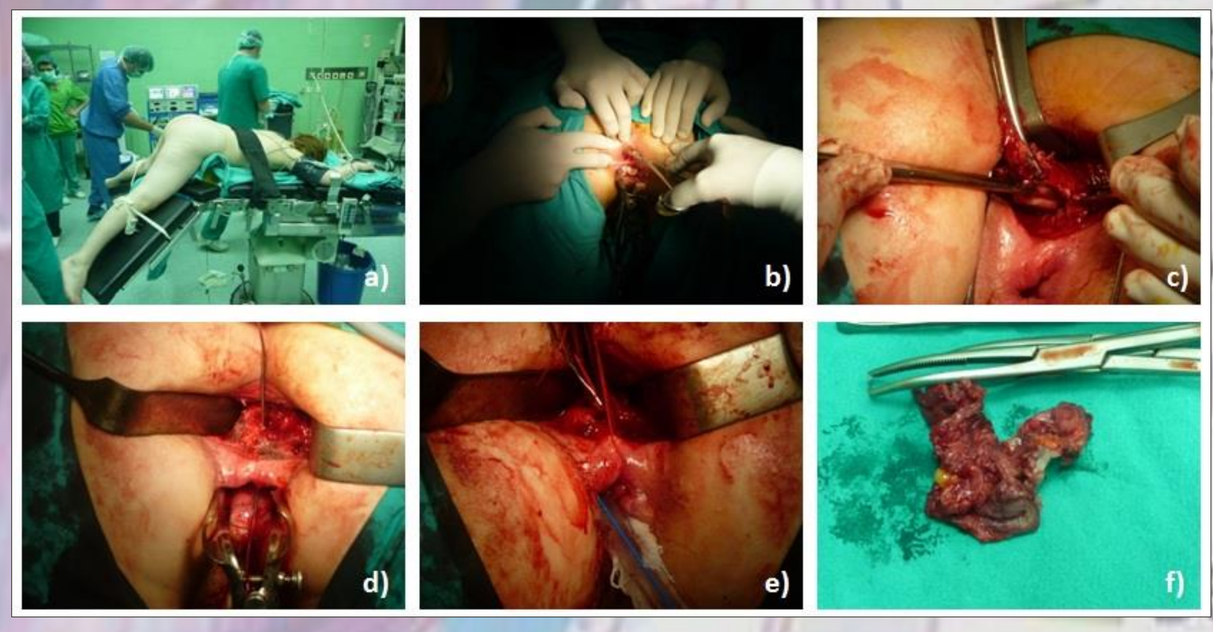

Fig. 3 *Course of the operation*

CASE REPORT

We are presenting 35 years old female patient who was first admitted to hospital with severe perianal and rectal pain, local signs of inflammation (tumor, dolor, calor, rubor, functio laesa and fluctuation) systemic signs of inflammation (fever, high temperature, leukocytosis) and typical so-called postanal dimple from which there was a leakage of suppurative exudate or pus. The DRE was very painful and soft tumor can be felt in the retrorectal space with intact mucosa over it. The symptoms were present about one week and the patient had no trouble before in her life. The leaking of pus started two days before admission when she felt slate relief. At first the condition was understood as retrorectal, pre-coccigeal abscess which had spontaneously ruptured in the postanal region. The patient was started on broad spectrum antibiotics and incision with evacuation of the pus from the retrorectal space was done through the spontaneous opening in the postanal dimple. Penrose drain was installed in the pre-coccigeal space which was changed dally along with dally washings with antiseptic solutions. After seven days of such treatment the local and systemic signs of inflammation were subsided as well as the suppuration and the patient was discharged. On the controls the patient was complaining on occasional perianal pain. On inspection of the perianal region there are two openings anal and epithelized postanal opening which make a picture of "double anus**" (Figure 1)**. On bimanual examination using DRE with probing of the postanal opening the probe goes deep in the postanal precoccigeal and presacral region. There is constant drainage of serosanguineous fluid from the postanal opening as a sign of fistula formation and persisting tumor in the presacral space on DRE with partial compression of the anorectum. In the meantime MRI and fistulography were done that showed cystic tumor in the retrorectal, presacral space with minor communication with rectum **(Figure 2)**. About 40 days after the first hospitalization the patient was send on the Clinic of Digestive Surgery at the Medical faculty – Skopje for definitive treatment. Preoperatively the colon was prepared conventionally and prophylactic antibiotic regimen was started. In the operating room the patient was put in prone jack-knife position **(Figure 3a)** and on explorative anoscopy the internal opening of a fistula in the anorectum about 3 cm above midline posterior crypt of Morgagni was registered. The operation was started with excision of the perineal opening of the cyst **(Figure 3b)**. The cyst is about 14 cm long and was liberated completely along with its capsule **(Figure 3c, 3f)** which enabled to visualize the fistulous canal which Leeds to the internal opening in the rectum **(Figure 3d, 3e).** The canal was excised with suturing the rectum in two layers first only the mucosa and then the muscle layer. The residual space after removing of the cyst was laid open and treated with daily washings with antiseptic solutions. The patohistological finding at our Institute of Pathology is: CYSTA EPITELIALIS CONGENITA INFLAMATA with all characteristics for dermoid cyst included in the text of the finding. After two mounts the process of defecation is normal, the wound is sealed without signs of inflammation and secretion locally although the postanal dimple still exists **(Figure 4).**

CONCLUSION

The retrorectal tumors are well defined, classified and understood pathological entities in the literature but in practice they represent very unusual and infrequent pathology [5, 10, 13].They are difficult for treatment as well as for diagnosis where even punctional biopsy is not recommended so they should be treated in specialized institutions by experienced surgeons from the moment of diagnosis to the definitive surgical treatment.

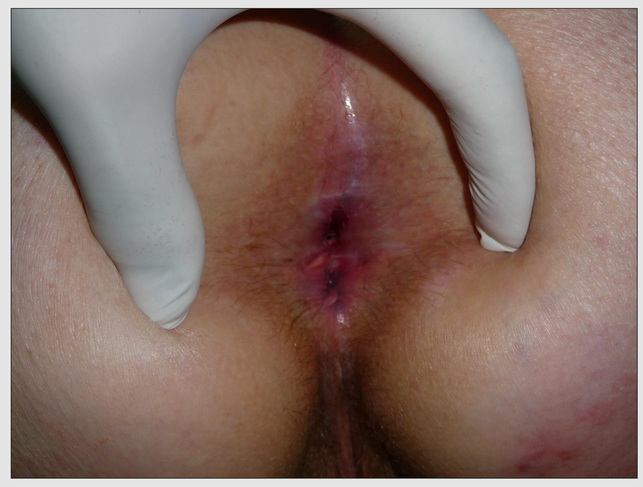

Fig. 4 *Current condition*

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