

Sacrococcygeal Approach for Presacral Tumors

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ABSTRACT

Aim:To identify sacrococcygeal approach as one of the best approach for complete excision of presacral tumors with minimum morbidity and mortality.

Method:In this study nine cases of presacral tumors are reported, seven females and two males, with mean age of (34.1 years), ranging from 17 to 58 years old, most of the cases are referred from other centers as cases of complicated sacrococcygeal pilonidal sinuses and retrorectal masses, five patients out of the seven had been operated before for the same complain and presented to us for recurrence of their symptoms after incomplete excision or misdiagnosed at the beginning ,they were admitted in Slemani Surgical Teaching Hospital and Soma Private Hospital with in a period of five years, after proper history and physical examination, investigations were requested and finally were diagnosed with histopathological result, These data were taken from the hospital's medical records and direct interview with the patients, consent was taken from all the patients regarding publishing their records in this series.

Result: sacrococcygeal approach resulted in complete resection of presacral tumors in all nine cases included in the study, with smooth post operative period and no recurrence recorded in the follow up period which was two years, pathological findings: fivemature cystic teratoma, one enterogenous cyst, one chondroma, and two tailgut cysts. No perioperative death.

Conclusion:the main treatment for presacral tumors is surgical, bowel preparation is necessary because sometimes the tumor invaded the rectum and the procedure may end with colostomy, in our study we approached all the cases through sacrococcygeal approach, complete excision of the tumor is necessary in order to prevent recurrence, daily dressing of the wound is mandatory to avoid secondary bacterial infection.

Keywords: Presacral tumors, sacrococcygeal approach, enterogenous

INTRODUCTION

Presacral tumors are rare. The incidence has been reported in literature to be between 1 in 40,000 to 1 in 63,000 hospital admissions¹. The sacrococcygeal area lies between the upper two thirds of the rectum and sacrum above the rectosacral fascia. It is bound by the rectum anteriorly, the presacral fascia posteriorly, and the endopelvic fascia laterally (lateral ligaments). The retrorectal space contains multiple embryologic remnants derived from a variety of tissues (neuroectoderm, notochord, and hindgut)². . The embryonic development and subsequent disappearance of the structures in this region aid in explaining the origin of these tumors³. The lesions are either solid or cystic, with developmental cysts being the most common retrorectal cystic lesions in adults, occurring mostly in middle-aged women⁴. Female to male ratio of 10:1⁵.

Although developmental cysts are often asymptomatic, patients may present with symptoms resulting from local mass effect (e.g., constipation, rectal fullness, lower abdominal pain, dysuria), with a palpable retrorectal mass at digital rectal examination⁴.

Due to its unusual presentation the diagnosis is challenging and requires a high level of suspicion, The cornerstone of diagnosis of presacral tumors is the digital rectal examination (DRE), with sensitivity approaching 97%⁶, Transrectal ultrasound imaging (TRUS), computed tomography (CT) and magnetic resonance imaging (MRI) provide assessment of their topography and relation to the

other anatomical structures of the pelvis minor (6), pelvic MRI is the most sensitive and specific imaging². Treatment is almost always surgical². Which include sacrococcygeal, transabdominal and combined abdominal-sacral approach⁷.

The main aim of the study is to identify sacrococcygeal approach as one of the best approach for complete excision of presacral tumors with minimum morbidity and mortality.

METHOD

Registration and ethics:The research registry number has been taken in accordance with the declaration of Helsinki- "Every research study involving human subjects must be registered in a publicly accessible database before recruitment of the first subject ".

The approval for conducting of this study was taken from the Scientific and Ethicalcommittee of kurdistani board for medical speciality

Study design:This study is a retrospective multicenter case series. The cases were consecutive.

Setting: The institutions in which the cases were managed were academic, community and private practice setting. All of them located in Kurdistan/Slemani. The patients were received and managed from February 2016 till May 2020. The data collection was done during this period.

Information was taken from patient data records, patients, and the supervising surgeons.

Participants: Inclusion criteria were all cases of presacral tumors admitted to our tertiary center. They were

diagnosed clinically and/or radiologically with MRI of pelvis or transperineal ultra sound. The socio-demographic and clinical data were taken from patients' medical records, health care providers and the patients themselves.

Pre-intervention consideration: All patients were prepared for general anesthesia and spinal anesthesia. They were checked prior to the operation for vital signs, hemoglobin level, and chest x.ray, and MRI of pelvis, mechanical and chemical bowel preparation done, they were examined and the area of proposed incision was marked

Types of intervention(s) deployed: Five patients underwent operation under general anesthesia (GA), four patients under spinal anesthesia (SA). In prone position. Through elliptical incision with excision of the coccyx exploration of pre sacral region was done. They were nil by mouth for six hours and received intravenous antibiotics. Mechanical and chemical bowel preparation was done for eight patients out of the nine patients.

Peri-intervention considerations: During the operation, the patients were followed up by continuous electrocardiography (ECG) monitoring. Intravenous fluid in form of crystalloid was given according to vital signs. Postoperatively after excision of the tumors the wounds were left open for secondary closure.

Who performed the procedures: The procedure was performed by specialist with a team composed of senior house officers and nurses.

Post-intervention consideration: The wounds were left open for secondary closure with daily dressing, initially the wounds were packed with gauze flavored with iodine and after 2 days the gauze were removed and daily dressing was started with normal saline. The diagnosis of all cases was confirmed by the result of the histopathological examination (HPE) of the excised specimen.

RESULTS

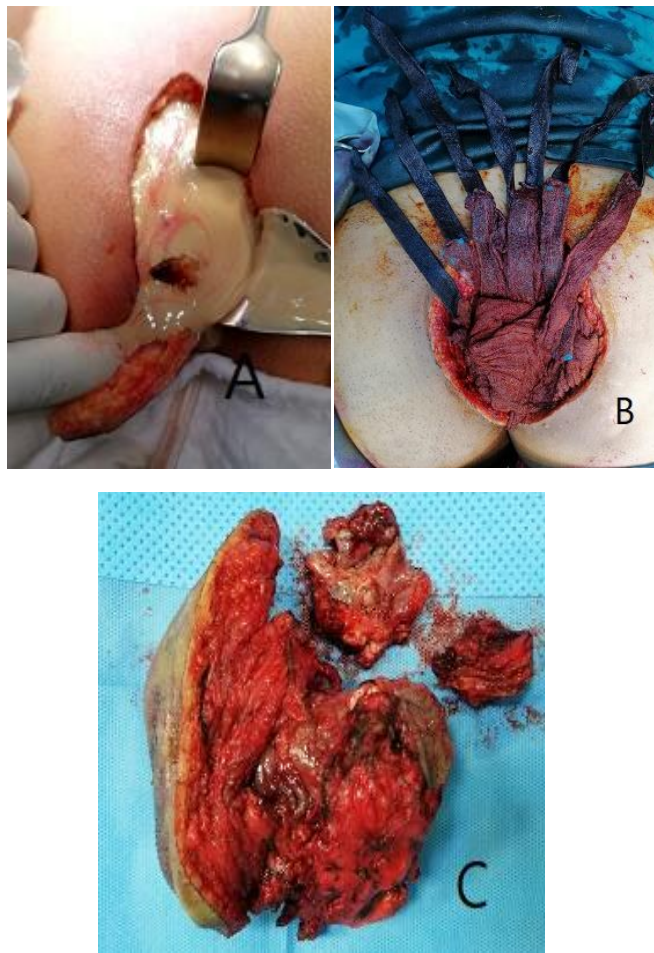
Participants: The series included nine patients, seven of them female and two of them male, ranging from seventeen to fifty eight years old, most of the cases were referred from other centers after recurrence of symptoms post operative intervention of presacral mass, surgery was performed for all of them with complete resection of the tumors, no recurrence in reported in the follow up period.

The diagnosis was suspected after performing radiological investigations of the pelvis and confirmed after final histopathological report returned.

Patient 1: A 24 year-old-male patient presented with low backpain for about three years duration, associated with discharge for the last one month, he had been operated when he was ten years old for a mass in the lower back at the coccygeal region, now he has recurrence of the mass and referred to our center, transperineal ultra sound was performed showing (large complex space occupying lesion occupying coccygeal region with both multiloculated cystic and solid component, the lesion extending deep in to retrosacral region, with no obvious relation to the anal canal) , after preparation of the patient under general anesthesia in prone position complete excision of the lesion was done and biopsy sent for HPE, wound left for secondary healing, after placing gauze flavored with iodine

(Figure 3-1 A,B,C), result of the biopsy was mature cystic teratoma.

Figure 1 (A) show drainage of the cyst content, (B) placing of gauze and leaving the wound for secondary healing, (C) biopsy of the cyst after complete excision.



Patient 2: A 17-year-old female patient presented with lower abdominal pain for about seven months duration, U/S of the abdomen was done and showed (Right adnexia contain (108 mm X 90 mm) thin wall cystic lesion which contain clear fluid & contain thin septa inside) the provisional diagnosis was para-ovarian cyst, she was operated through lower midline incision, both ovaries were normal and no pathology could be found, after referral to our center, MRI of the pelvis was done with oral and iv contrast which showed (large 102 mm X 94 mm presacral cystic lesion seen with pelvic extension pushing the uterus and urinary bladder anteriorly, no obvious intra spinal extension, overall picture in favor of anterior type sacrococcygeal cystic teratoma) (figure 3-2 A,B)

Operation was done by the surgical team, under general anesthesia, prone position, large presacral cystic lesion was found containing clear fluid, complete excision of the cyst wall was done (figure 3-3 A) and biopsy sent for HPE (figure 3-3B) which turned to be mature cystic teratoma, the

wound was left for secondary closure and during the follow up period the patient was well and no sign of recurrence.

Fig. 2: MRI of pelvis showing the cystic lesion. (A) Coronal view, (B) Sagittal view.

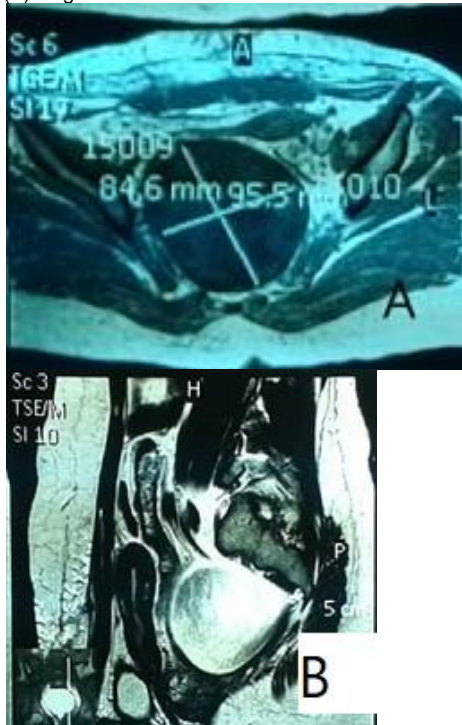


Figure 3 (A) Complete excision of the cyst wall

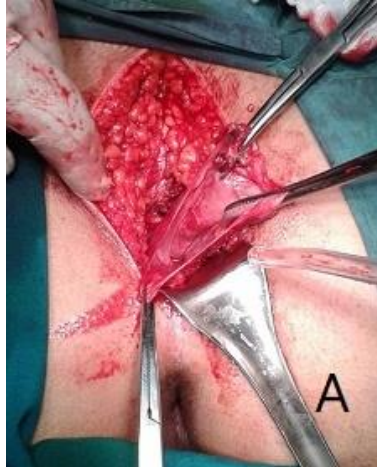
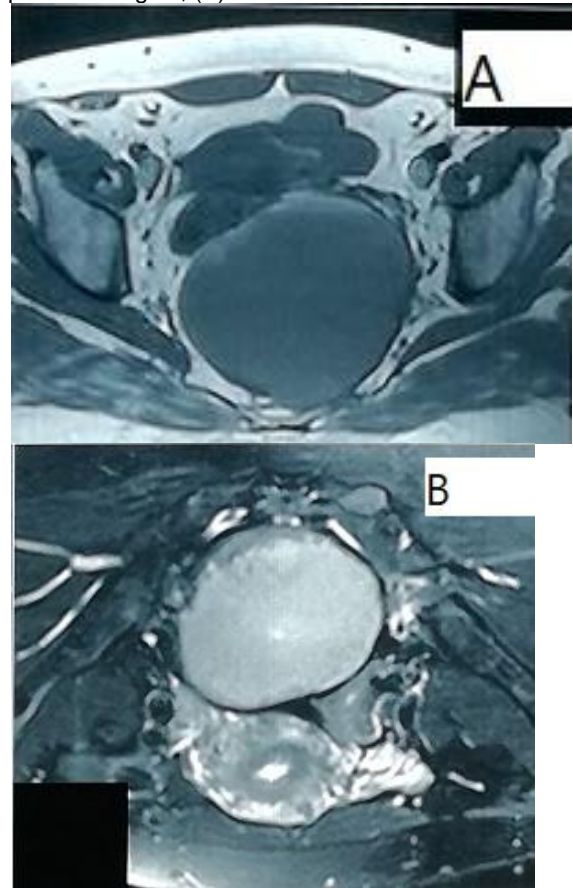


Figure 3 (B) biopsy of the tumor after complete excision.



Patient 3 : A 42-year-old ,unmarried ,female patient presented with long standing lower abdominal pain and lower back pain for about 4 years duration, no associated symptoms , she has history of thyroid disease before 15 years which was controlled by medication, past surgical history negative. Patient sent for MRI of pelvis which showed (anterior to the sacroccocygeal segment is a large cyst with proteinous content measuring 9 X8 X 7.5 cm , the cyst shows few filaments entering in-between sacral vertebrae) (figure5 A,B) . Operation was done under general anesthesia in prone position large fluid containing cystic lesion found, complete excision was done, wound left for secondary closure, in follow up visit the patient was well and within 2 years of follow up there was no sign recurrence, result of the biopsy was enterogenous cyst, no malignancy.

Figure5 : MRI of the pelvis (A) showing the lesion in the presacral region, (B) retro rectal extension.



Patient 4: A 58-year-old female patient presented with chronic low back pain for about six years duration. She had been operated before five years for the same complain as there was a mass in the pre sacral region. She was hypertensive on treatment, pre operative evaluation was done all were normal, MRI of the pelvis done with intravenous and oral contrast which showed (a large lobulated mass about 9 cm X 6 cm involving the coccyx and extra peritoneal soft tissue at presacral space pressing on and displacing the rectum and uterine cervix, extending

posteriorly to involve soft tissue and subcutaneous layer of gluteal region, the mass is homogenous hypointense on T1, bright on T2, showing avid heterogeneous enhancement on IV contrast, no associated regional lymphadenopathy) (figure 6 A,B). Operation was done under general anesthesia, semi prone position, complete excision of the mass was done, wound left for secondary healing, result of the histopathological report showed chordoma.

Figure 6 MRI study of pelvis (A,B) showing large lobulated mass involving the coccyx.



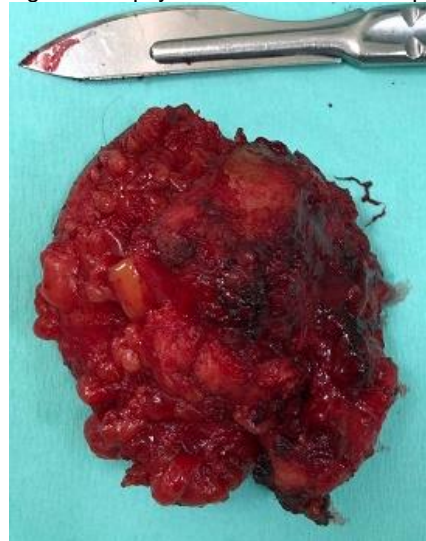
Patient 5: A 47-year-old male patient presented with recurrent attacks of pain in the lower back for the last four years, he has been operated four times for same condition, all the previous biopsies were benign after histopathological examination but not completely excised, after referral to our center MRI of the pelvis was requested which shows (evidence of small 17 mm X 5 mm residual cystic cavity at the site of the operation at the lower sacral region close to the tip of the coccyx, it shows marginal

enhancement and mild edema in the region, no relation to the pelvic wall musculature, no affection of the external anal sphincters, sinus formation seen to the skin from the cavity), after preparation of the patient under spinal anesthesia, prone position, complete excision of the lesion done, the wound was left open for secondary healing (figure 7), result of the biopsy (figure 3-8) was tailgut cyst (retrorectal cystic hamartoma), no sign of recurrence in two years follow up period.

Figure 3-7 intraoperative images showing the cystic lesion.



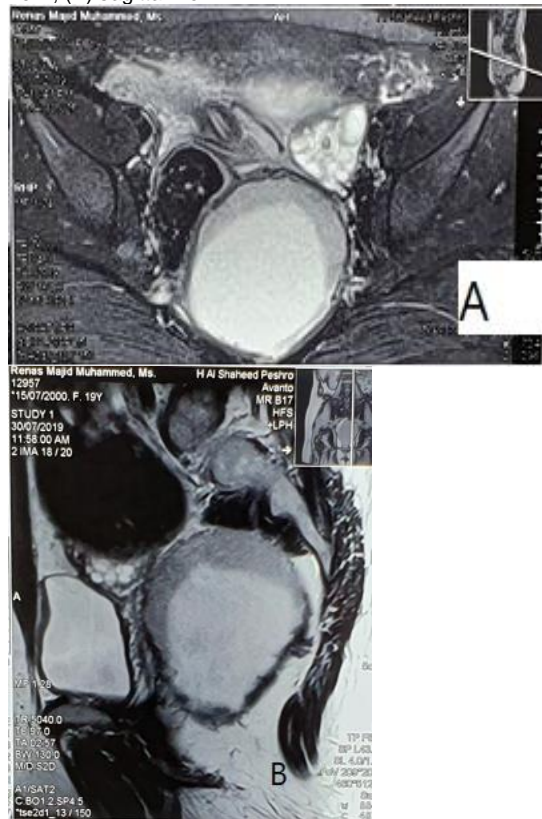
Figure 8 biopsy of the lesion after complete excision.



Patient 6: A 19-year-old female patient presented with chronic low abdominal pain and altered bowel motion for about two years duration, ultra sound of the abdomen was done showing (a 83 mm X 73 mm cystic lesion with internal debris with few septation adjacent to left ovary), after referral to our center the patient was sent for MRI of the pelvis which showed (large cystic lesion 92 mm X 88 mm X 86 mm with multilocular appearance displacing the uterus to the right side anteriorly, contains mostly serous fluid – T2

hyper intense & T1 hypointense, post contrast study showed slight enhancement of the wall, picture was of an extra peritoneal pre sacral cystic mass) (figure 9 A,B), under general anesthesia in semi prone position, through elliptical incision drainage of the cyst and complete excision of the wall was done, wound left for secondary healing, biopsy result was of mature cystic teratoma.

Figure-9 MRI images showing the large cystic lesion with few internal debris and few septation displacing the uterus.(A) coronal view , (B) sagittal view.



Patient 7: A 38-year-old female presented with low abdominal pain for about two years duration, she had been operated through mid line incision for an abdominal mass which turned to be presacral lesion, de-roofing done alone, after one year she had recurrence of her symptoms, ultra sound of the abdomen was done showing 120 mm X 100 mm X 80 mm midline presacral cystic structure (figure 3-10), MRI of the pelvis was requested which shows presacral cystic lesion of 90mm X80mmX7.5 mm with septation (figure 3-11), operation was done under spinal anesthesia, sacrococcygeal approach, elliptical incision done, complete excision of the cyst wall done and sent for HPE (figure 12), wound left for secondary healing, result of the biopsy was mature cystic teratoma.

Figure 10 ultrasound image showing 12X10X8 cm cystic structure



Figure 11 MRI of pelvis showing the cystic mass with septations.

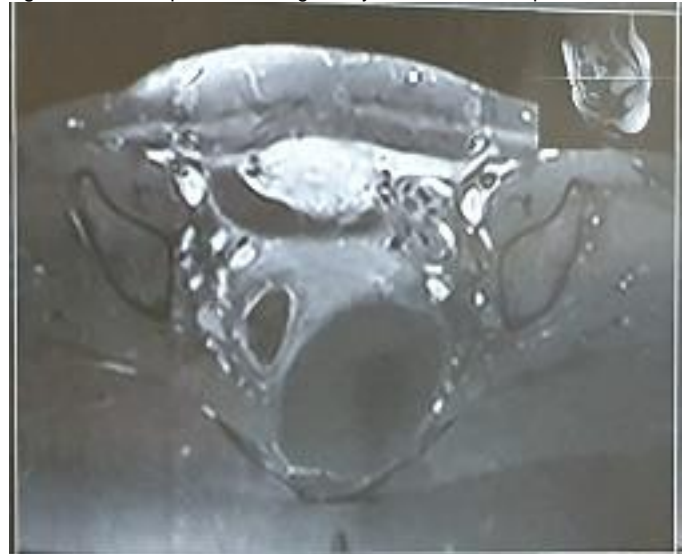
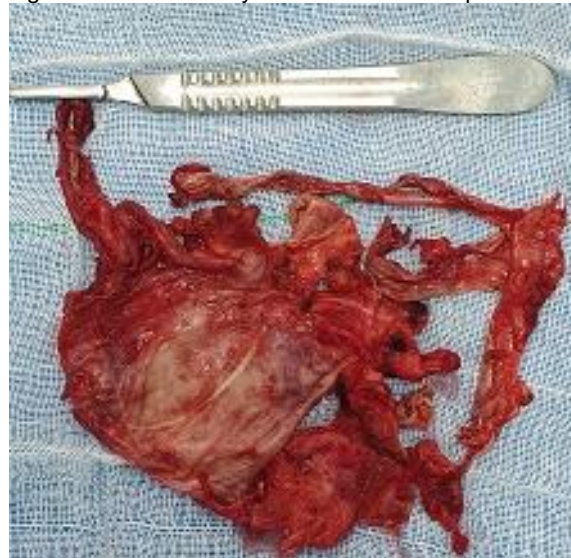


Figure 12 wall of the cystic lesion after complete excision.



Patient 8: A 22-year-old female presented with chronic low back pain and constipation for about two years duration, trans anal ultra sound shows a cystic like structure

measuring about 5 cm x 5 cm in the presacral region, operation was done under GA, prone position, sacrococcygeal approach, complete excision of the lesion done and sent for HPE (figure 13), wound left open for secondary healing, result of the biopsy was mature cystic teratoma, in the follow up period the patient was free of pain and no recurrence of symptoms in two years follow up.

Figure 13 Complete excision of the cystic lesion



Figure 14 (A) MRI sagittal view showing the cystic lesion posterior to the rectum, (B) biopsy of the cystic mass after complete excision.



Patient 9: A 40-year-old married female presented with chronic lower back pain for about three months duration, she had history of abortion before four months with intermittent vaginal bleeding, DRE was done there was a palpable mass in the posterior aspect, pelvic US was done which showed (left adnexial lesion about 8x7 cm with internal echos with few calcification foci, CT or MRI suggested), MRI was performed which showed (evidence of retro-rectal complex cystic lesion about 70x60x50 mm , not related to adnexia) (Figure 3-14 A) , decision was made for operation, under SA , prone position , through sacrococcygeal approach complete excision of the cystic lesion done which contained yellow color fluid material, biopsy sent for HPE (Figure 3-14 B), wound left open for secondary healing, result of the biopsy was tail gut cyst.

Result of HPEs:

Patient	Age	Gender	Result
1	24 year	Male	Mature cystic teratoma
2	17 year	Female	Mature cystic teratoma
3	42 year	Female	Enterogenous cyst
4	58 year	Female	Chordoma
5	47 year	Male	Tailgut cyst
6	19 year	Female	Mature cystic teratoma
7	38 year	Female	Mature cystic teratoma
8	22 year	Female	Mature cystic teratoma
9	40 year	Female	Tailgut cyst

Outcome and follow up: Within two years of follow up all patients were found to be cured from the disease, with no recurrence reported in any patient, the wounds were healed after leaving them for secondary closure.

Complications and adverse or unanticipated events: No any significant complication was reported.

DISCUSSION

Pre sacral tumors are rare conditions, incidence has been reported to be between 1 in 40,000 to 1 in 63,000 of those who are admitted in hospital¹, many sacrococcygeal tumors in adults are asymptomatic and often found on physical examination or by chance during imaging studies (8), most of our cases were misdiagnosed from the beginning, they presented with vague abdominal or lower back pain, and some of them operated on as ovarian cysts or complicated pilonidal sinus abscess in the coccygeal region then after referred to our center .

The congenital tumors are most common and arise from all three germ layers, these are more common in females, these may arise from ectoderm (dermoids and epidermoids), sequestration of hindgut remnants (tail gut cysts, rectal duplication cysts), neural tube defects (anterior sacral meningocele), and notochord (chordomas)¹. They are classified into three groups: 1/ benign, containing well-differentiated tissue; 2/ immature, without malignant features; and 3/ malignant (9). A careful digital rectal examination is diagnostic in 90%¹. After appropriate diagnostic interventions, complete surgical resection remains the primary and only satisfactory treatment; there are three approaches commonly used for resection; abdominal, transsacral, or a combined abdominosacral approach¹⁰. Coccyx should be excised en bloc only when involved with tumor or existence of doubtful malignant

potential¹⁰, Most of the available papers report neonatal retro-rectal tumors, our research include nine adult patients within a period of 3 years.

A research which was done in Mayo clinic collecting patients from 1960-1979 included 120 patients with a mean age of 43 years, with a female predominance associated with congenital cyst (15:1) and male predominance of chordomas (5:1)¹¹, the mean age of our patients was 34 years, with female predominance associated with congenital cysts (7:2), the biopsy result of 5 patients turned to be mature cystic teratoma, which they completely responded to the surgical excision and daily dressing.

Detailed history and complete physical examination is mandatory for reaching the diagnosis, comprehensive clinical assessment including digital rectal examination is essential to establish a diagnosis¹³, Computed tomography of the pelvis is widely used and is capable of differentiating solid from cystic lesions, vascular, adjacent organ involvement and bony destruction, Contrast enhanced MRI pelvis is the gold standard investigation of choice and the most sensitive and specific investigation also¹². Needle biopsy should be avoided because complications including meningitis after puncture of a meningocele, infection of a cystic tumor, malignant extension through the biopsy tract or bleeding can occur¹³. Only in cases of unresectable lesions, or in patients with significant comorbidity that precludes pelvic surgery, may biopsies be performed for histological diagnosis if required to indicate adjuvant or palliative therapy¹³, in our research we didn't perform needle biopsy for any of the cases.

In a study by Sean C. Glasgow in department of surgery/ Washington university, Thirty-four patients with retrorectal tumors were treated, the Surgical approach was anterior (n = 14), posterior (n = 11), and combined abdominoperineal (n = 9). Eleven patients required *en bloc* proctectomy. Patients undergoing posterior resection had lower blood loss and required fewer transfusions (14).all of our patients underwent posterior sacrococcygeal approach with complete excision of the tumors, no transfusion and no rectal injury reported.

CONCLUSION

Presacral tumors are uncommon and diagnosis requires high level of suspicion along with digital rectal examination in all suspected cases, when diagnosed sacrococcygeal approach provides a good way for complete resection of the tumor with minimal morbidity to the patient as there is decreased incidence of blood loss, rectal or adjacent organ injury, and no recurrence were reported in two years follow up period.

Recommendations

1. In all patients presenting with pilonidal sinus or abscesses in the natal cleft DRE should be done to exclude presacral lesions and exclude other pathologies.
2. In cases of recurrent abscess in the natal cleft, presacral teratoma should be suspected and further radiological work up be done.
3. MRI of the pelvis provides the best anatomical imaging.

4. Mechanical and chemical bowel preparations are mandatory before any operative intervention.
5. Sacrococcygeal approach provides an excellent way for complete excision of the tumor with minimum blood loss and co morbidity.

Conflicts of interest: There is no conflict to be declared.

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Ethical approval: Ethical approval has been taken from Kurdistan board for medical specialty.

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