Giant benign tumor of the sacrum - A case report

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Summary

Introduction: A benign sacral tumor is a rare disease, especially with a giant tumor. We will present the clinical examination, imaging diagnosing images, and our experience in this case. *Case presentation*: We report a 25-year-old female with a massive sacral tumor, which had been developing for ten years. Clinical examination showed a 210 × 150 × 210mm mass originating from S2 into the sacrum with organ displacement. The biopsy confirmed that was a benign tumor. The surgery was performed with the anterior and posterior approach to remove the tumor and preserve sacroiliac joints. After 18 months, her recovery was smooth. However, she still presented irreversible damages to the bladder and rectal sphincter functions. *Conclusion:* En block resection of the giant benign sacral tumor with the anterior and posterior approaches is only administrated for a massive and complicated mass, which can only be performed in hospitals with advanced operating departments, anesthesiology, and aggressive supportive care.

Keywords: Sacrococcygeal bones, benign tumor.

1. Background

A benign sacral tumor accounts for 2% to 5% of primary sacral tumors in general, which usually happens at the age of 20 to 40 [1, 2]. The symptoms come on gradually. Thus, this disease often is diagnosed late when the size of the tumor becomes bigger. En bloc resection is the best option for small tumors. However, with bigger tumors causing mass effect to the vascular supplies of the sacrum, nerve system, and organs in the pelvic cavity, the tumor removing process is more challenging. Blood loss during operation and coagulation disorder due to longing fluid transfusion are reasons for the postoperative death. Besides, many complications such as pelvic contents damages and severe infection can affect the postoperative life quality. Surgeons must have a suitable strategy and a good combination with anesthesiologists to have a successful operation [3], [4], [5], [6]. This case report will present a successful operation on a giant benign sacral tumor and experience in the treatment process.

2. Case presentation

A female patient with a 50kg weight and 160cm height had found the tumor of ten years. The onset symptom was a dull pain in the sacrococcygeal region happening continuously for years. The patient also presented fecal and urinary incontinence, paresthesia, and physical insensibility in the perineum. Sexual dysfunction appeared because the tumor had developed near the pubis. We could not do a vaginal examination and a physical examination showed a giant sacrum tumor with hard density. Portosystemic collaterals were recognized on the sacral mass skin surface (Figure 1). The mass had developed and had a serious impact on the hypogastric region. The musculoskeletal, sensory system, and tendon reflexes of lower limbs were normal.

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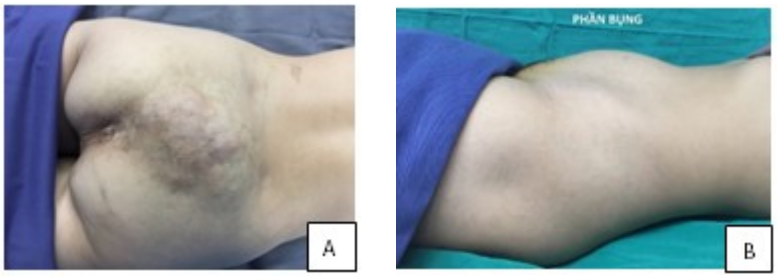

Figure 1. The tumor developed backward and portosystemic collaterals presented at the surface (A), the hypogastric region presented a bulging mass (B).

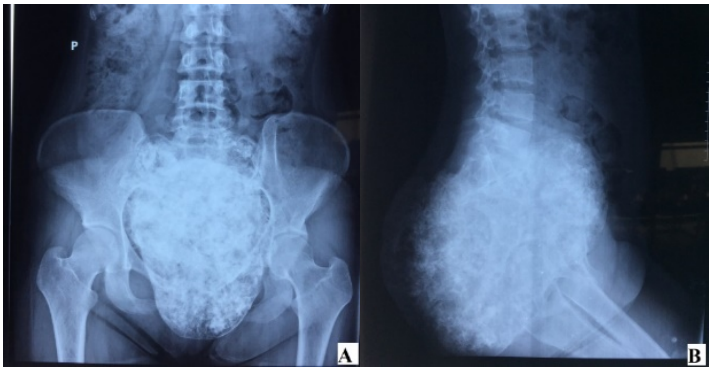

Figure 2. AP X-ray image (A) and lateral X-ray image (B) of the pelvis. A massive inconsistent radiodensity image caused a mass effect in the pelvis, and the sacrococcygeal bones are destroyed.

Imaging diagnostic tests including X-ray, computed tomography (CT), and magnetic resonance imaging (MRI) clarified that the sacrum had been destroyed from S2 to below. The mass presented an image of inconsistent radiodensity on both X-ray and CT images. CT scan with contrast showed that the tumor did not absorb the contrast material. The right ureter was compressed by the tumor. Thus, the right kidney was atrophied. MRI revealed a well-demarcated image, an inconsistent signal intensity of the sacral tumor, and the pelvic contents were pushed into the front and below the sacrococcygeal bones. No evidence of invasion of the mass to other organs nearby is found on CT and MRI images. DSA showed that big arteries in the pelvis were displaced by the mass and no big artery supply for the tumor. Cystoscopy and proctoscopy did not find any abnormal results.

Bone scan showed increased radiotracer of the sacrococcygeal region and no abnormal increased radiotracer in other localized regions. Biopsy taken confirmed the sponge bones were benign covering with connective tissues. No malignant images existed.

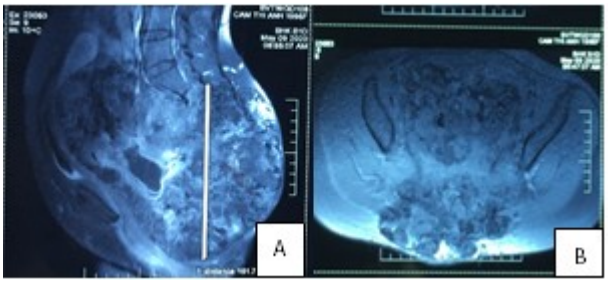

Figure 3. Inconsistent signal intensity of the tumor on AP MRI image (A) and lateral MRI image (B).

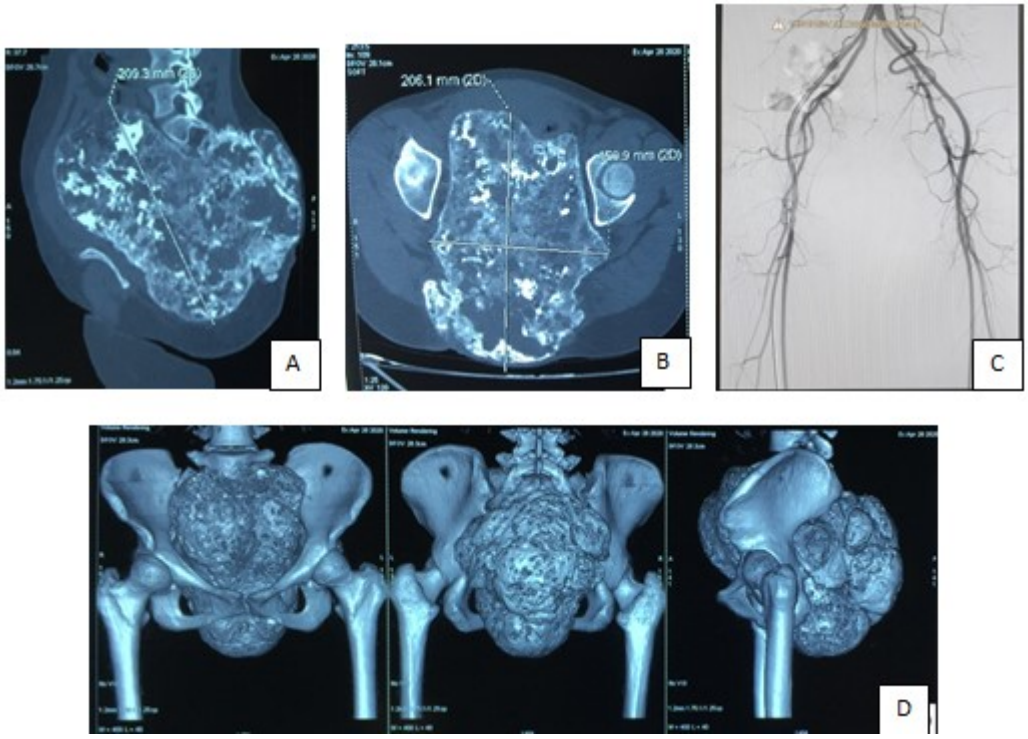

Figure 4. A, B: Sagittal and coronal views CT scan of the tumor. CT scanner: The tumor originated from S2 into the sacrum measuring 210 × 150 × 210mm in size, with a hyperdense material, a well-demarcated image including sacrococcygeal bone destruction. Organs in the pelvic were displaced into the ventral space and there was no evidence of encroachment. The right kidney was atrophied due to tumor compression. There was no image of lymph nodes in the pelvic region. C: DSA image of the tumor's supplied blood vessels. D: Three-dimensional image of the tumor on computer tomography.

The operation happened in March 2020 and was performed by many surgery experts in the abdomen, vascular, and spinal. We have exposed the tumor in two ways: The anterior approach and the posterior approach. With the anterior approach, the big arteries were exposed following upper and lower midline incisions. Then, the tumor was separated from other abdominal organs and chiseled to reduce its size. The tumor was exposed following the posterior gluteal incision line, which had the Mercedes shape. We removed the mass and exposed bilateral sacroiliac joints. The S1 nerve root was revealed clearly after removing the S1 vertebral arch. However, other nerve roots were still infiltrated by the mass. We chiseled the tumor through the sacroiliac joint and continued to break the sacrum on the same horizontal as the S1-S2 disc to collect the remaining tumor pieces. The S1 sacroiliac joint was still preserved to stabilize the bilateral sacroiliac joints. During the process of tumor removal with the posterior approach, we had to turn the patient over to check for abdominal bleeding because the vital signs dropped and vasopressors were used to raise these figures. The internal iliac vein and Sigmoid colon were damaged when breaking the tumor. Thus, surgeons had to cut off the Sigmoid colon and the appendix and perform an abdominal washout. The colostomy was placed at the right hypochondriac region, then we inserted two drains, and closed the abdomen. We turned the patient to the prone position and opened the *Sacrococcygeal* region damage. We exposed and cut off the attachments of gluteus muscles, and turned over the fasciocutaneous gluteal flaps to cover the defect due to removing the mass.

The operation lasted for 10 hours. The mass weight was 5,13kg. The patient was transfused 11000ml blood, including 3000ml self whole blood

taken from a Cell-saver machine system; 8000ml RBCs; 6000ml fresh plasma; 400ml factor VIII, and two units of platelets (3×10^9 platelets/ml per 1 unit). We had to check the results of a complete blood count test (CBC), artery blood gas, and coagulation every 15-30 minutes to control the acid-base balance, and serum electrolytes.

The blood loss was 1500ml/3h after the operation. That was the reason why we had to check the bleeding conditions. No evidence of bleeding was found in the abdomen and the gluteal region. The patient was transfused 2000ml of RBCs. The coagulation disorder was successfully controlled by ROTEM: 500ml fresh plasma and two units of platelets.

Periodical examination showed that bilateral lower limbs' function and sensory were normal. Mild pain in the right sacroiliac joint did not limit the patient's movement. Urinary incontinence still existed and the stoma worked well. After six months since the operation, the patient could have sexual activities.

Figure 5: A, B: Anterior and posterior incision line (Mercedes line)
C: The tumor was taken out; D: Postoperative CT-scanner image

3. Discussion

The surgeons must concern about the risks of massive bleeding and abdominal injuries when performing the procedure. The bigger of tumors, the bigger chance of bleeding when operating. The surgical decision was decided depending on many criteria: Age, intraoperative complications, and the patient's desire. Due to the risks that may happen during operation, the procedure is only allowed to perform at technologically advanced hospitals with surgical specialists in spinal, abdomen, vascular, and anesthesiology. We have predicted all possibilities that can happen and prepared solutions for each case. The preoperative imaging diagnostic tests clarified that there was no evidence of invasion of the tumor to other nearby organs. That was a plus point for our surgery decision.

The operation strategy with whether anterior or posterior approaches should always be considered when removing a massive sacrococcygeal tumor [7], [8], [9], 10]. The anterior incision should be done first to expose and cover arteries and abdominal contents during the chiseling process. The iliac veins are often be damaged because these walls are thinner than arteries. Internal iliac arteries should be preserved for supplying blood to the gluteal flap. Following posterior incision to remove S1 and S2 vertebral arches, dissect nerve roots before removing the tumor to maintain the nerve function as much as possible. Because the tumor has developed overlap the S2 spine and the patient had a symptom of urinary incontinence, we have decided to remove the S2 nerve root. Chiseling the tumor from the posterior approach made several impacts on the abdominal organs and large blood vessels. The internal iliac vein and Sigmoid colon wall were damaged because the posterior operation view was small and we did not cover the abdominal contents carefully. Thus, we had to close the incision temporarily and open the anterior incision to check for bleeding. Changing posture during an operation may cause damage to the patient (especially when the hemodynamic is unstable), extend the surgical time and increase the risks of infection. To avoid secondary bowel perforation and be more comfortable in postoperative monitoring, the damaged colon was cut off and the proximal stoma was used for colostomy. Lateral sacroiliac joint fixation is not required because both sacroiliac joints and the S1 spine are still maintained. Two fasciocutaneous gluteal flaps were used to cover the defect (due to the tumor removal) [11], [12], [13]. The gluteal flap rotation helped the wound recover faster and avoid abdominal hernia in the future [14], [15].

The biggest risk of removing sacral tumor surgery with prolonged time and a high volume of blood loss is coagulopathy. Coagulopathy

correction and adequate blood transfusion are the keys to successful surgery. For surgical removal of benign tumors, re-circulating blood transfusion through a Cell saver filter is the fastest and safest compensation method for circulating volume. Continuous tests in a certain period with correction of coagulation by ROTEM are effective and accurate methods. Coagulation disorders can be complex and long-lasting. Although major bleeding causes might not be found, surgical removal of hematoma can prevent endogenous coagulopathy factors.

After 18 months, postoperative examinations showed that the patient had no movement damage in the lower limbs, with stable sacroiliac joints. The quality of life has be significantly improved. The patient had no pain. She could have normal sexual activity, although bladder and rectal sphincter functions have irreversible damage.

4. Conclusion

Giant benign sacral tumor resection is challenging and risky to both the surgical team and the patient. The procedure should only be performed at advanced technological hospitals with well-experienced surgeons. Specific strategy and accurate complication predictions are the keys in this case.

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