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Case Study

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SACRAL CHORDOMA: A RARE PRESENTATION AS GLUTEAL ABSCESS

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ABSTRACT

Sacral tumour is a rare clinical entity. The most common type of primary sacral tumour is chordoma. Sacral chordoma is a locally aggressive, malignant tumour originating from ectopic notochordal cells. Chordoma predominantly affects the axial skeleton such as sacrum, skull and other mobile segments of the spine. Sacral chordoma is a slow growing tumour arising at the midline of the lower sacrum that can locally invade the sacral bone and progressively increase in size expanding cranially and anteriorly, thus hindering its early diagnosis. Its diagnosis can be delayed because of its unclear clinical manifestations. Here, we report a case of sacral chordoma with a rare and unusual presentation as gluteal abscess.

KEYWORDS: chordoma, notochord, abscess.

INTRODUCTION

Sacral tumour is a rare clinical entity.[1] The most common type of primary sacral tumour is chordoma.[1,2] In 29-50% of all cases, this tumour involves the sacral region.[3] Sacral chordoma is a locally aggressive, malignant tumour originating from ectopic notochordal cells. Chordoma predominantly affects the axial skeleton such as sacrum, skull and other mobile segments of the spine. Sacral chordoma is a slow growing tumour arising at the midline of the lower sacrum that can locally invade the sacral bone and progressively increase in size expanding cranially and anteriorly. This sacral tumour if large enough can push abdominal and pelvic organs anteriorly. Metastasis is very rare even when the tumour is very large. In addition, the tumour rarely invades the rectum or genitourinary organs because it is confined by the thick presacral fascia and periosteum. Sacral chordoma affects males more than females and is more commonly found in middle age and elderly patients.[1]

Usually, clinical presentation is unclear with uncertain symptoms that often lead to a working diagnosis of another more frequent disease.[4] The slow growth rate of this tumour hinders its early diagnosis.[5]

CASE REPORT

- A 69 year old female patient was admitted at our hospital with the complaints of pain in bilateral gluteal region since last one month and swelling in bilateral gluteal region since last 15days. There was a history of backache along with the restriction of movements. There was no associated history of fever, trauma or any insect bite. History of multiple intramuscular injections especially of analgesics for the complaint of backache was found to be present. Patient was a chronic smoker with no history of any comorbidity. Patient was vitally stable. On local examinaion, there was found to be a swelling present in both sides of the gluteal region with raised local temperature. Tenderness was found to be present. The swelling was found to be cystic in consistency. Fluctuation test was found to be positive. There was found to be local erythema in that region. With a suspicion of gluteal abcess due to possible cause being intramuscular injections, ultrasonography of bilateral gluteal region was performed. It showed the presence of multiloculated, heterogenously hypodense areas, with thin septations, with low level echoes and foci within it, in bilateral gluteal region which was suggestive of first an abscess formation (loculated) or second an organised infective collection(loculated). Patient was further subjected to other routine investigations (Hb- 7.1g/dl, TLC-13,700/mcl, ESR=45mm/hr, blood urea=26mg/dl, serum creatinine=0.6mg/dl, coagulation profile=18.8sec, serum sodium=146.1mmol/L, potassium=3.9mmol/L, blood sugar=55mg/dl, INR=1.52) Fine needle aspiration cytology showed a few neutrophils admixed with few lymphocytes.

Incision and drainage was planned and about 2100 ml of reddish jelly like material was aspirated from right side and about 150 ml from left side.

- Post operatively, the jelly like material continued discharging from the incision site for 3 consecuetive days. There was a little relief in pain but the restriction of movements persisted for which ortho consultation was taken. Computerized tomography of the bilateral gluteal region was planned which showed a well-circumscribed lytic lesion with marginal sclerosis with areas of cystic necrosis and heamorrhage seen in sacrum, suggestive of sacral chordoma. patient was referred to the higher centre for further management(neurosurgical consultation) on his attendant s request amd so could not be further followed.

DISCUSSION

Chordoma is a rare, (4%) slow growing neoplasm arising from cellular remnants of notochord.[1,2] Due to its slow growth rate, the presentation is late and diagnosis is hindered. It usually occurs at 50 years of age. The most common site is sacrum and coccyx and it communicates with gluteal musculature which may be confused with gluteal abscess.[4] This is the possible cause of such presentation in our case. Usually, clinical presentation is unclear with uncertain symptoms that often lead to a working diagnosis of another more frequent disease.[4] It can locally invade the sacral bone and progressively increase in size expanding cranially and anteriorly, thus hindering its early diagnosis. Its diagnosis can be delayed because of its unclear clinical manifestations.[5]

Sacral chordoma is a locally aggressive malignant tumour that rarely metastasizes even if it presents late.[5] This is one of the malignant tumours that lack many features of malignancy on histopathological analysis such as mitosis, anaplasia and increase nuclear-cytoplasmic ratio. Most sacral chordoma occurred in middle age or elderly adults.[1]

The most common underlying cause of coccygodynia and mass formation in the natal cleft is pilonidal disease. The 3 characteristic findings that confirm the diagnosis are: 1) abscess or sinus, 2) drainage of fluid (usually purulent), and 3) midline skin pits. In case one or more of them are absent, we should think about the rare causes of the coccygodynia such as chordoma, giant cell tumour, intradural Schwannoma, perineural cyst, and intra-osseous lipoma.[6]

Surgical resection with wide margins is considered the treatment of choice for sacral chordoma because it improves local control and disease-free survival.[7-10] Local recurrence rates of 43% to 85% and metastases rates of 5% to 40% at 1 to 10 years have been reported for sacral chordomas.[8-11] When gluteal invasion is present, the risk of recurrence is reportedly higher and wider margins are important.[12]

Even though sacral chordoma is considered to be a benign tumour, it can evolve with time, representing the most common malignant tumour of the sacral region. Sometimes chordomas are diagnosed when they are already at an advanced stage with distant metastasis to the lymph nodes, liver, bones, lungs, brain, soft tissues and/or the peritoneal cavity.[13]

Sacrococcygeal chordomas are insidious tumors that are difficult to diagnose. Many patients are treated for an assortment of unrelated diagnoses before the correct diagnosis is made.[7]

CONCLUSION

Considering the above situation, we conclude that effective management of sacrococcygeal chordoma requires early diagnosis, accurate preoperative staging, definitive and adequate surgical resection with proven tumor-free cut margins, and close follow-up.

Conflicts of interest

None.

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