

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: http://Elsevier.com/locate/radcr



Case Report

Large presacral epidermoid cyst in an asymptomatic woman

Kyosuke Izumi MD^a, Satoshi Tsutsumi MD^{a,*}, Takeshi Hara MD^b, Hisato Ishii MD^a, Masanori Ito MD^a, Yukimasa Yasumoto MD^a

ARTICLE INFO

Article history:
Received 15 May 2017
Received in revised form 9 July 2017
Accepted 27 July 2017
Available online

Keywords: Presacral cyst Epidermoid cyst Asymptomatic DWI

ABSTRACT

An epidermoid cyst is an infrequent entity among cysts found in the presacral region, frequently coexistent with a meningocele. Diffusion-weighted imaging is known to be a useful diagnostic measure for differentiating presacral epidermoid cysts. Here, we present a large but asymptomatic case found in the presacral region. Epidermoid cysts should be considered in patients with presacral cysts.

© 2017 the Authors. Published by Elsevier Inc. under copyright license from the University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

Introduction

An epidermoid cyst is an infrequent entity among cystic pathologies found in the presacral region, which includes benign tumors represented by epidermoid, dermoid, and enteric cysts, and malignant pathologies represented by teratoma, teratocarcinoma, and yolk sac tumor [1]. An epidermoid cyst frequently coexists with a meningocele, the spina bifida, thick filum terminale, and lipoma. Also, an epidermoid cyst can coexist with anorectal malformation manifesting hemisacral agenesis [2–6]. Here, we present an asymptomatic case of a large epidermoid cyst in the presacral region. This cyst interfered with the treatment of a markedly compressing cervical cancer.

Case presentation

A 34-year-old woman had been followed up for 8 years at the Department of Gynecology for an asymptomatic pelvic mass. No anorectal or urological abnormalities were identified in this patient. Episodes of recurrent meningitis were not noted. At the time, the patient was diagnosed with a cervical cancer by tissue biopsy that required surgical resection. However, as the uterus was markedly compressed and deformed by the pelvic mass, it was difficult to examine the extent of the cervical cancer. A lumbar magnetic resonance image of the patient suggested a possible association between the pelvic mass and the sacral spinal sac. The patient was referred to our department

Competing Interests: The authors declare no competing interests concerning either the materials and methods or the findings presented in this study.

E-mail address: shotaro@juntendo-urayasu.jp (S. Tsutsumi). https://doi.org/10.1016/j.radcr.2017.07.017

^a Department of Neurological Surgery, Juntendo University Urayasu Hospital, 2-1-1 Tomioka, Urayasu, 279-0021 Chiba, Japan

^bDepartment of Neurological Surgery, Juntendo University School of medicine, Tokyo, Japan

^{*} Corresponding author.

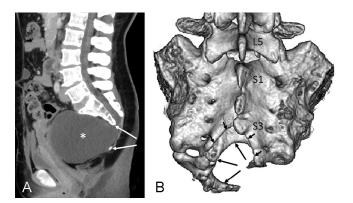


Fig. 1 – (A) Sagittal CT scan showing a well-demarcated hypodense mass in the presacral region (asterisk) 12×10 cm in maximal dimension accompanying chronic-appearing, well-marginated bony defects in the ventral sacrum (arrows). (B) Dorsal view from a 3-dimensional CT scan showing a spina bifida below the S3 level (short arrows) and the scimitar outline in the sacrum (long arrows). CT, computed tomography.

at this time. At presentation, the neurologic examination of the patient indicated no abnormal findings. Superficial findings in the lumbosacral region were normal. Computed tomography scans indicated a well-demarcated hypodense mass in the presacral region that was 12×10 cm in maximal dimension. The hypodense mass accompanied chronic-appearing, wellmarginated bony defects in the ventral sacrum. Threedimensional computed tomography scans indicated a spina bifida below the S3 and the scimitar outline in the sacrum (Fig. 1). T1-weighted magnetic resonance imaging showed an entirely capsulized large cyst that appeared heterogeneously hypointense. The cyst appeared heterogeneously hyperintense on T2-weighted images, although its intensity was lower than that of cerebrospinal fluid. A thick filum terminale continuous with the cyst wall and a membranous structure dividing the cyst from the sacral subarachnoid space were also noted (Fig. 2). On diffusion-weighted imaging (DWI), the cyst

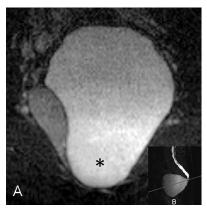




Fig. 2 – Sagittal T1- and T2-weighted magnetic resonance images showing a capsulized large cyst. The cyst appears heterogeneously hypointense on T1-weighted imaging (A) and heterogeneously hyperintense on T2-weighted imaging (B), but appears less intense than cerebrospinal fluid on T2-weighted imaging. Note that a thick filum terminale (arrow, B) is continuous with the cyst wall and a membranous structure separates the cyst from the sacral subarachnoid space (asterisk, A and B).

contents appeared hyperintense in the posterior portion and hypointense in the anterior portion (Fig. 3A). On the apparent diffusion coefficient map, the former presented as an iso signal, whereas the latter presented as mixed signals (Fig. 3B).

A surgery using a posterior sacral approach was carried out based on a presumptive diagnosis of a presacral meningocele. Laminectomy of the dysraphic S3 followed by incision in the transitional area between the normal and the pathologic dura exposed the thick filum terminale. The filum was cut at the S4 level to untether the cord. The defect in the ventral sacrum was then circumferentially dissected. The dissection revealed a membranous structure dividing



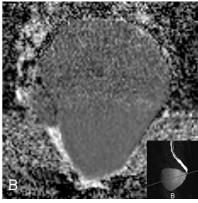


Fig. 3 – (A) Axial diffusion-weighted magnetic resonance image at the neck level of the lesion (inset) appearing hyperintense in the posterior portion and hypointense in the anterior portion (asterisk). (B) Axial apparent diffusion coefficient map at the same level showing the former as an iso signal and the latter as mixed signals.

the sacral sac from the cyst. Solid pearly tissue was observed upon incision of the membrane instead of fluid contents. This tissue was radically resected and histologically verified as an epidermoid cyst.

Discussion

Developmental cysts arising in the presacral region include variable benign and malignant tumors. These cysts often mimic adnexal cysts on imaging examinations. As a result, definitive diagnosis is commonly made intraoperatively [1]. Of note, reported cases of epidermoid cyst have been coexistent with meningoceles in variable proportions [2–6]. In contrast, the natural history of epidermoid cysts arising in the presacral region is elusive. In addition, when and how to treat asymptomatic cases are not defined.

A previous study reported that DWI can be used to differentiate perineural cysts from other slow-growing cysts [7]. Consistently, the presacral epidermoid cyst in the present case was well-characterized by DWI. The peculiar appearance of the cyst on DWI, with a hyperintense posterior portion and a hypointense anterior portion, may indicate a long-standing and continuous keratin deposition in the anterior direction. Presacral cysts often mimic adnexal cysts. In addition, most of the reported cases of epidermoid cyst in this region, including the present case, have been observed in women with a broad range of ages [2–6]. Therefore, DWI should be included in routine sequences when examining women with presacral cysts.

Conclusions

Epidermoid cyst should be considered when encountering female patients with cysts in the presacral region. DWI is a useful diagnostic measure for the detection of presacral epidermoid cysts.

REFERENCES

- [1] Dahan H, Arrivé L, Wendum D, Docou le Pointe H, Djouhri H, Tubiana JM. Retrorectal developmental cysts in adults: clinical and radiologic-histopathologic review, differential diagnosis, and treatment. Radiographics 2001;21:575–84.
- [2] Haga Y, Cho H, Shinoda S, Masuzawa T. Recurrent meningitis associated with complete Currarino triad in an adult—case report. Neurol Med Chir (Tokyo) 2003;43:505–8.
- [3] Horii T, Tsuchiya H, Tomita K. Presacral tumor associated with the Currarino triad in an adolescent. Arch Orthop Trauma Surg 2001;121:114–6.
- [4] Kansal R, Mahore A, Dange N, Kukreja S. Epidermoid cyst inside anterior sacral meningocele in an adult patient of Currarino syndrome manifesting with meningitis. Turk Neurosurg 2012;22:659–61.
- [5] Shamoto H, Yoshida Y, Shirane R, Yoshimoto T. Anterior sacral meningocele completely occupied by an epidermoid tumor. Childs Nerv Syst 1999;15:209–11.
- [6] Nakamura S, Wakamatsu K, Tsubokawa T, Moriyasu N. Sacral epidermoid cyst communicating with the spinal CSF canal. Childs Brain 1980;6:103–11.
- [7] Manara R, Severino M, Mandari R, Mattisi G, Dal Pozzo S, Carollo C. Chronic cystic lesion of the sacrum: characterisation with diffusion-weighted MR imaging. Radiol Med 2008;113:739–46.