Epidermoid cysts compose less than 1% of spinal cord tumors. Calcified intraspinal epidermoid cysts are rare, with only one case reported to date. Here, we report the case of an epidermoid cyst with calcification located at the cauda equina level; reported data on calcified intraspinal epidermoid cysts have also been summarized.

A 24-year-old woman presented with bilateral soreness in the gluteal regions and numbness on the lateral side of the left foot for approximately 3 months. A cauda equina tumor was diagnosed, and she visited our hospital for thorough examination and treatment. Neurological examination revealed no abnormality except for hyperalgesia in the left S1, 2, and 3 regions; plain radiographs revealed no bone destruction or malalignment. There was no history of surgical procedures, including lumbar puncture, that could have caused skin tissue to stray into spinal canal and no physical findings of skin sinuses suggestive of congenital disease.

Computed tomography revealed calcification in the spinal canal at the level of the lower endplate of the fifth lumbar vertebra (Fig. 1A, 1B); computed tomography myelography revealed a circular mass at this level and an oval mass caudal to it (Fig. 1C). The calcified area was located between the two masses (black arrow in Fig. 1C). Magnetic resonance imaging (MRI) revealed a bifurcated intradural extra-medullary tumor, demonstrating an iso signal intensity on T1-weighted images (Fig. 1D); its cephalic and caudal parts showed hypo (white triangles in Fig. 1E, 1F) and hyperintensity (white arrows in Fig. 1E, 1G) signals, respectively, on T2-weighted images. Contrast-enhanced MRI revealed homogeneous and heterogeneous contrast effects in the cephalic (white triangles in Fig. 1H, 1I) and caudal parts of the tumor, respectively (white arrows in Fig. 1H, 1J). A definitive diagnosis could not be made on the basis of preoperative imaging findings, and the tumor was removed for diagnostic and therapeutic purposes. After laminotomy of the L5 and S1, a bifurcated tumor with a membrane was found upon incising the dura; there were no adhesions between the tumor and dura. Dissection was first performed around the caudal tumor, as it was considerably large. The cephalic and caudal tumors was divided into two at the connective tissue between the cephalic and caudal tumors, and all tissues were removed with relative ease (Fig. 2A, 2B). The tumor was a keratoma-filled cyst lined by squamous epithelium (Fig. 2C) and was diagnosed as an epidermoid cyst due to the presence of cholesterol granulomas in some parts (Fig. 2D).

Epidermoid cysts are a benign tumor lined by a thin film of squamous epithelium containing cystic components filled with shed epithelial cells and cholesterol crystals. Congenital epidermoid cysts are believed to originate from stray epidermal cells in the skin, while acquired epidermoid cysts result from epidermal cell penetration into the skin secondary to lumbar puncture, surgery, or trauma. Ten cases of epidermoid cysts with calcification have been reported; only one was in the spinal canal. The previous and present cases are similar in terms of the sex of the patient and tumor location but differ with respect to the MRI findings (Table 1). Heterotopic calcification may explain the occurrence of calcification in an epidermoid cyst; lipids contained in the skin presumably stray into the dural canal and are later saponified in the weakly alkaline cerebrospinal fluid, leading to calcium salt deposition in the same area. In this case, calcification was observed in the small cross-sectional area between the two masses, suggesting the saponification of the surface in contact with the cerebrospinal fluid.
We experienced an epidermoid cyst with calcification located at the level of the cauda equina. Accompanying calcification should be considered during preoperative diagnosis and surgical planning of epidermoid cysts.

Conflicts of Interest: The authors declare that there are no relevant conflicts of interest.

Sources of Funding: None

Acknowledgement: The authors thank all the staff at the Jikei University School of Medicine, Kashiwa Hospital, for their kind support.

Author Contributions: S.K., C.U., and T.I. designed the research; S.K., C.U., T.I., T.I., A.S., T.K., D.A., S.A., S.K., S.O., S.S., and M.S. analyzed the data; S.K. wrote the paper; C.U. and M.S. supervised the study. All authors read and approved the final manuscript.
Figure 2. Gross and microscopic appearance of the tumor. (A) Photograph showing the caudal (left) and cephalic (right) parts of the tumor. (B) Photograph showing the tissue leading to the caudal part of the tumor. The cephalic and caudal parts of the tumor were originally connected via this tissue. Calcified tissue was present in the same area. Microscopic images showing (C) a cyst lined by squamous epithelium (black arrows) and (D) cholesterol granuloma (arrowheads) (hematoxylin & eosin staining).

Table 1. Reported Studies in English regarding Epidermoid Cyst with Calcification.

| Investigator          | Sex  | Age (years) | Symptoms                                      | Location          | MRI features                                                                 | Resection extent | Follow-up period | Recurrence |
|-----------------------|------|-------------|-----------------------------------------------|-------------------|------------------------------------------------------------------------------|------------------|-----------------|------------|
| Agrawal et al., 2019¹ | Female | 32          | Progressive spastic paraparesis with incontinence | L1–L3 levels      | T1WI, NA; T2WI, cerebrospinal fluid-like signal with hypointense lesion      | GTR              | 6 months        | NER        |
| Present patient       | Female | 24          | Soreness in both buttocks and numbness on the lateral side of the left foot | L5–S1 levels      | T1WI, iso signal; T2WI, low signal for the cephalic tumor, high signal for most of the caudal tumor; Gd-enhanced T1WI, homogeneous contrast effect for the cephalic tumor and a heterogeneous contrast effect for the caudal tumor | GTR              | 2 years         | NER        |

MRI, magnetic resonance imaging; NA, not available; GTR, gross total resection; T1WI, T1-weighted images; T2WI, T2-weighted images; Gd, gadolinium; NER, no evidence of recurrence

Ethical Approval: Ethical approval was waived by the ethics committee since this was a case report. All performed procedures were part of the routine care. The study was conducted in accordance with the principles of the Declaration of Helsinki and the laws and regulations of Japan.

Informed Consent: Informed consent for publication was obtained from the patient regarding the publication of her clinical data and photographs.
References

1. Roux A, Mercier C, Larbrisseau A, et al. Intramedullary epidermoid cysts of the spinal cord: case report. J Neurosurg. 1992;76(3):528-33.
2. Agrawal M, Gour SSK, Borkar SA. Unusual calcification in intramedullary epidermoid cyst. World Neurosurg. 2019;126:99-100.
3. Smirniotopoulos JG, Chiechi MV. Teratomas, dermoids, and epidermoids of the head and neck. Radiographics. 1995;15(6):1437-55.
4. Miyake S, Kobayashi N, Murai N, et al. Acquired lumbar epidermoid cyst in an adult. Neurol Med Chir. 2005;45(5):277-9.
5. Greenwood JE, Tan JL, Ming JC, et al. Alkalis and skin. J Burn Care Res. 2016;37(2):135-41.
6. Ikeda DM. Breast imaging: the requisites. 2nd ed. Philadelphia (United States): Elsevier/Mosby; c2011. Chapter 4, Mammographic and ultrasound analysis of breast masses; p. 109-11.