Pacinian corpuscles are specialized nerve endings that have a role in vibration and pressure sensation. They are concentrated in the dermis of the palmar surface of the hands and feet, as well as in the joints and tendons. Only a few pathological lesions containing pacinian corpuscles have been reported in the literature, the majority being pacinian neurofibromas. Other case reports have also documented lipomas containing muscle tissue, cartilage, bone, and respiratory epithelium. Only 3 reports in the literature have described mature pacinian corpuscles within a lipoma. Here we describe an asymptomatic variant of a complex intraspinal lumbosacral lipoma for which we have used the term “paciniolipoma.”

Case Report

History and Examination. A female newborn was found to have a soft-tissue swelling over the lumbosacral junction. There were no complications during pregnancy or childbirth. The baby had developed normally, and there were no concerns regarding sphincter function. She also had normal limb movements. She was the first-born child of Southeast Asian parents with no family history of dysraphism.

A 2 × 3–cm subcutaneous lipoma with no skin defect was seen at the lumbosacral region of the back. There was no cough or strain impulse, and no neurological deficit was identified. A whole-spine MR image obtained when the girl was 12 months of age showed dysraphism with a narrow intradural lipoma extending down from the dorsal cord at L-1 to L-3 and continuous with the subcutaneous lipoma through a defect in the dorsal elements at S1–2. The cord was tethered and the conus lay low at the L-3 level. The rest of the neuraxis was normal with a capacious lumbar canal and no mass effect from the lipoma (Fig. 1). At 15 months of age the patient underwent prophylactic surgery to untether the cord and improve cosmesis.

Operation. A longitudinal incision was made over the fatty lump, the subcutaneous fat was divided, and the subcutaneous lipoma was dissected out. The stalk extending into the canal through a bone defect in the spinal canal was identified at the level of S1–2, and a bilateral muscle strip was performed to expose abnormal dysplastic dorsal elements above the stalk as well as normal sacral laminae. A laminectomy was performed superior to the stalk to the level of L-4, as was a partial sacral laminectomy inferior to the lesion. The stalk was followed to where it entered the dura mater, which was then opened around it. The intradural lipoma was divided rostrally, and the remaining intradural portion was retracted up into the dural tube appropriately. A separate thickened

This article contains some figures that are displayed in color online but in black and white in the print edition.
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fatty filum was also seen and was coagulated and divided. Histological examination of the lipoma revealed mature adipose tissue with abundant pacinian corpuscles (Fig. 2). These corpuscles comprised elongated spindle-shaped cells arranged in multiple concentric circles. Between the pacinian corpuscles were scattered peripheral nerve twigs. Immunostaining for neurofilament protein showed a single axon at the center of the corpuscles. Cells positive

Fig. 1. Whole-spine sagittal and axial T1- (A and C) and T2-weighted (B and D) MR images. The spinal cord is low lying and terminates at around mid-L3 (A and B). There is an intradural lipoma attached to the posterior aspect of the lower spinal cord extending from L1–2 downward. There is spina bifida at S-2, and a fibrous tract extends from this region to the surface of the skin at the level of approximately L-4. The lipoma exerts no mass effect in the canal (C and D).

Fig. 2. Photomicrographs showing pacinian corpuscles. A: Pacinian corpuscles comprising multiple concentric layers of cells. H & E. B: Note the central dot-like axon at the center of the pacinian corpuscles. Immunoperoxidase stain for neurofilament protein. C: Note the S100-positive cells at the center of pacinian corpuscles. Immunoperoxidase stain for S100 protein. D: Elongated spindle-shaped cells forming the layers of the pacinian corpuscles are epithelial membrane antigen–positive perineural cells. Immunoperoxidase stain for epithelial membrane antigen. E: Low-power photomicrograph showing an overview of the lesion. Note the lipoma (clear cells) with collagenous septa and pacinian corpuscles (arrows). Bars = 100 mm.
Postoperative Course. Postoperatively the child made an uncomplicated recovery. At the 6-month follow-up, she had normal power and sensation in her lower limbs with no restriction in forward flexion and no disturbance in sphincter function.

Discussion

Congenital lipomas are well described in spinal dysraphism at the lumbosacral canal. Many of these lesions have contained fibrous connective tissue among normal adipose tissue, while other case reports have described a variety of heterotopic cells of ectodermal, mesodermal, or endodermal origin. In the case of myolipoma, the fatty stalk was seen to contract with diathermy, suggesting that the muscle fibers were functional. We do not know whether the pacinian corpuscles were functioning in our case. Clinically, our patient did not show any discomfort or reaction to palpation of the lump. No such comment was made in the other reports of similar histology.

An abnormal remnant of such tissues during caudal neural tube formation has been posited as an explanation for why lipoma variants can be found intradurally. Other theories postulate that premature disjunction during primary neurulation can leave the posterior endplates open, allowing aberrant cell migration of mesenchymal cells causing lipomyelomeningocele, and perhaps that histological variants are caused by other cell lineages also migrating with these mesenchymal cells, for example, neuroectodermal cells from which pacinian corpuscles arise, and mixing within the normal adipose tissue. Other tissue types seen within some lipomas may result from the abnormal persistence of pluripotent embryonic cells. Terms such as “myolipoma,” “paciniomyolipoma,” and “pacinioma” have been used to describe these entities accordingly.

We have described a fatty, cord-like mass connecting subcutaneous tissue to intradural structures of a dysraphic spine and consisting of mature pacinian corpuscles within adipose tissue, which demonstrate a curious histological appearance. Although previous lesions were presented as “pacinioma” by Bale and Kojc et al., we prefer to apply the term “paciniolipoma” because it emphasizes the intermixed components of pacinian corpuscles and lipoma in this lesion. The previous reports show similarities with ours in that the lipomatous mass appeared to be connected to the subcutaneous layer through a bony defect of the lower spine, one at the L-5 level and the others, as in our case, in the sacrococcygeal region. Of these reports, only one did not pierce the dura but was attached to the external surface of the dura. Only one case exhibited abnormal overlying skin with a red sacral papule.

We believe that this rare histological variant has no clinical manifestation, although the subjective sensation of “pressure” is difficult to obtain from infants during assessment. The discovery of future variants may help to clarify the etiology of this lesion.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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