CASE REPORT

No Longer an African Child's Disease: The First Indonesian Case Report of Adeloye-Odeku Disease, A Rare Anterior Fontanelle Cyst

Kharisma Ridho Husodo[1,2], Akhmad Suryonurafif[1,3]

[1.] Departement of Neurosurgery, Yogyakarta Islamic Hospital PDHI (Persaudaraan Djama'ah Haji Indonesia), Yogyakarta 55571, Indonesia
[2.] Faculty of Medicine, Brawijaya University, Malang 65145, East Java, Indonesia
[3.] Academic Hospital of Gadjah Mada University, Yogyakarta 55291, Indonesia

ABSTRACT

Adeloye-Odeku disease was initially reported as an African congenital cyst in 1971, but this disease is now been documented worldwide. Adeloye-Odeku disease is classified as a congenital subgaleal inclusion dermoid cyst of the anterior fontanelle region. Mostly, this disease occurs in infants or children. This is the first case report of Adeloye-Odeku disease in Indonesia. The case report of a 5-month girl is presented with a lump that had developed since birth. The characteristic of lump was soft, non-tender, dome-shaped, and positive transillumination results. A computed tomography (CT) scan was carried out, with the result of a subgaleal dermoid inclusion cyst in the anterior fontanelle region. The anterior fontanelle appeared widened without intracranial expansion . Surgical excision of the cyst was performed under general anesthesia successfully. Histopathologic examination showed dermoid cysts without malignancy features. There were no complications reported when the patient attended the follow-up visit at the outpatient clinic. This is consistent with the existing literature on Adeloye-Odeku disease. *Malaysian Journal of Medicine and Health Sciences (*2024) 20(4): 403-406. doi:10.47836/mjmhs20.4.51

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Corresponding Author:
Kharisma Ridho Husodo, MD
Email: kharisma48ridho@gmail.com
Tel: +6285743930870

INTRODUCTION

Two neurosurgeons from Nigeria, Adeloye and Odeku, first described this congenital disease in 1971 and named it Adeloye-Odeku disease (1). This disease is a rare lesion that develops at birth with the characteristic of a lump located at the anterior fontanelle, whose size ranges from 10 to 30 mm (2). The worldwide occurrence of congenital dermoid cysts of the anterior fontanelle has since been shown in publications from Nigeria, Europe, Japan, China , Turkey, Brazil, and others (2) There are no case reports of this disease in Indonesia.

The cysts of Adeloye-Odeku disease are benign lesions originating from epithelium or dermal structures along the midline during the embryonic development of the neural tube. The cysts usually present as a lump in the first months of life and then grow bigger over time due to internal desquamation (3).Based on histopathology, most of the cysts can be either dermoid or epidermoid, but the first one is more common (4). Dermoid cysts are described as the presence of hair follicles, sweat, and sebaceous glands in a pathologic anatomy examination (4).

Some radiology workups that are helpful in the diagnosis of the disease are computed tomography (CT) scans or magnetic resonance imaging (MRI) before treatment decisions. A CT scan is needed to evaluate the characteristic of the lump and the involvement of the underlying bone and intracranial structures (4). MRI has superiority to inspect the soft tissue, but it is inferior to CT scans in inspecting bones. The differential diagnosis of this disease includes encephalocele, meningocele, sebaceous cyst, hemangioma, lipoma, and lymphangioma (3).

Most of these cases are treated surgically by excision to take the cyst out through a transverse incision (1). In this paper, we report a 5-month-old female who developed a subgaleal inclusion cyst with successful surgical treatment.

CASE REPORT

A five-month-old female child visited an outpatient clinic with a complaint of a lump in the head over the anterior fontanelle. The diameter of the lump was 4 cm. The lump appeared at birth and became prominent after one month, and it was getting bigger at the time of presentation. The characteristic of the lump was soft and immobile, and the colour was the same with the skin around it (Figure 1). The transillumination of the lump was positive, which indicated liquid inside the lump. There was no report from parents about suspected pain or discomfort associated with the lump. Any history of trauma was denied by the patient's parents. There was no systemic complaint. A family history of tumours or congenital anomalies was absent. During pregnancy, there were no disorders experienced by the patient's mother.

Figure 1: Clinical appearance of the cyst

There was no abnormality in the physical and neurological examinations. A non-contrast CT scan was carried out, with the result showing a subgaleal dermoid inclusion cyst in the anterior fontanelle region. The size was 4,2 x 4,1 x 2,6 cm. The anterior fontanelle appeared widened without intracranial expansion (Figure 2). An MRI examination also showed a dermoid cyst (Figure 3). Laboratory and baby-gramme x-ray examinations were within the normal limit.

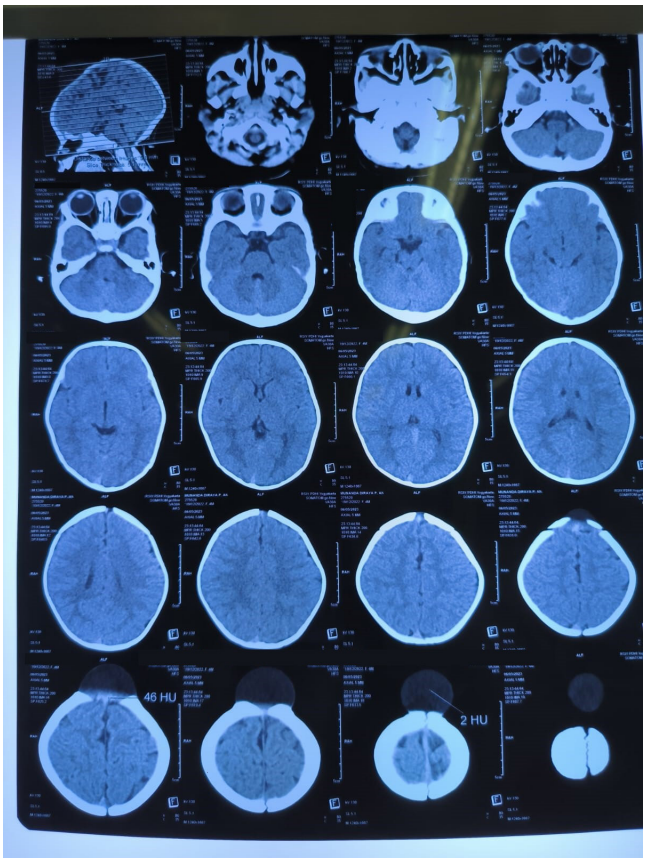

Figure 2: Non-Contrast Head CT scan of the patient

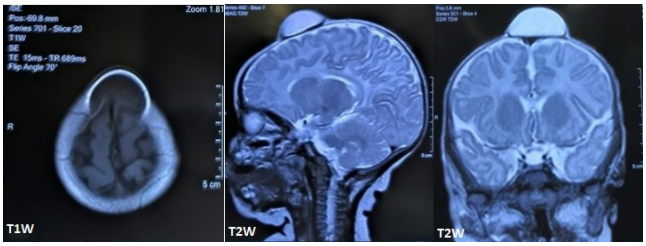

Figure 3: Head MRI examination of the patient

The surgery was performed under general anaesthesia by making a coronal incision over the lump. A blunt dissection of the cyst was done from the underlying tissues. The lump contained clear liquid with a white layer of hair and an adipose layer inside (Figure 4). After the cyst was taken out, the dura and cranium of the patient were intact, and there was no intracranial abnormality noted. The redundant scalp was trimmed to fit for closure. Wound suturing used Vicryl for the subgaleal layer and nylon for the skin layer. The cyst was then delivered for pathology and anatomy examination. The microscopic feature of the sample showed that the tissue was cyst-containing skin tissue and its adnexa with keratin. Malignant characteristics were not found. Histopathology concluded that it was a dermoid cyst.

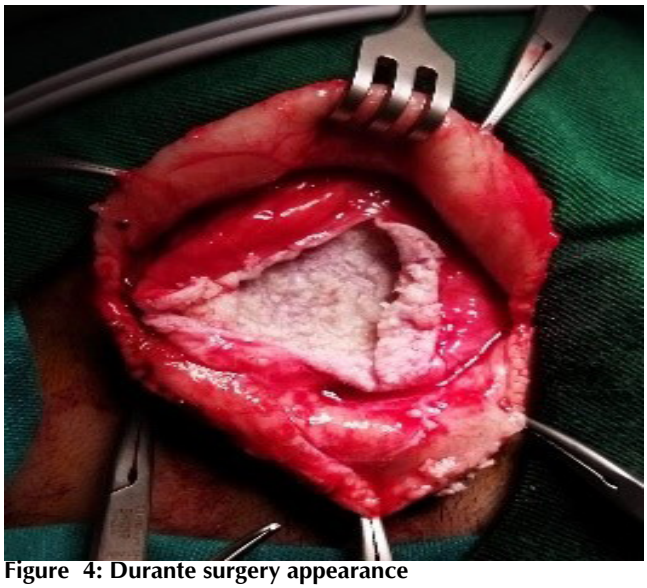

Figure 4: Durante surgery appearance

The patient was discharged two days after surgery without any complications. On the first follow-up visit at the outpatient clinic, the parents did not report any complaints. The post-surgery wound was clean, with no pus or clots. Then the suture was removed at a second follow-up visit at an outpatient clinic, 6 days after surgery. Six months after surgery, the patient had no complications such as recurring lumps, surgical site infections or complaints of pain.

DISCUSSION

Initially, Adeloye-Odeku disease was introduced as a rare congenital disorder in African children (1). It accounts for 0.1–0.2% of all cranial tumors and 0.2% of all inclusion cysts (3) Since then, several reports have been made from areas outside the Africa Region; such as Saudi Arabia, Turkey, Europe, Japan, and the Indian region. This case report is from the Southeast Asian Region, Indonesia (3). Therefore, this disease is a rare lesion that has been shown to be global and not African, as was initially thought (2).

From the latest literature, it can be concluded that Adeloye-odeku disease can occur at any age, however it typically manifests between the ages of three and six months during the first year of life (4). Female patients have a higher ration than male patients, with a comparison 2:1 (3) In this case, the patient has the majority criteria for the theory of the disease. This cyst characteristically manifests at birth. It is slow growing, non-tender, absence of intracranial extension, and being covered by skin (3). The transillumination test was positive in this patient as in the most case reports of Adeloye-Odeku disease (2).

A head CT scan of this patient showed a subgaleal dermoid cyst over the anterior fontanelle without intracranial expansion or bone erosion. MRI of the patient also support the result that the dermoid cyst is consistent with previous reports where it is hypointense on T1W and hyperintense on T2W (5). These radiology examinations are required to evaluate the cyst and ensure the involvement of bone or fontanelle and whether communication between the cyst and intracranial is present (4). However, in our opinion, a head CT scan is sufficient for diagnosis and preparation before surgery.

Histopathology showed that the patient lump was a dermoid cyst. This finding is in accordance with previous reports of Adeloye-Odeku disease, which showed the cyst wall was connective tissue lined by stratified squamous epithelium accompanied by sebaceous glands and hair follicles in the adnexial layer (4). In some cases, the conclusion of the pathologic anatomy examination was an epidemoid cyst, which is rarer than a dermoid cyst (1).

Treatment of choice is surgical excision, which was done for the index patient (4). The surgical procedure involves enucleation of the cyst through a transverse scalp incision (1). The surgery should be performed carefully to avoid injury to the underlying dura if fontanella is still patent (5). Then the redundant scalp may be excised to make the wound closure tight and fit aesthetically, as was done in this patient (1). The complications of this disease are rare; dural tears and post-operative leptomeningeal cyst formation were reported in a few cases (5).

CONCLUSION

The Adeloye Odeku disease has not been previously documented in Indonesia. This is the report of a 5-month old child, with an anterior fontanelle dermoid cyst consistent with the disease, which was successfully excised. This further supports the existing literature that the AdeloyeOdeku cyst is not an only African child disease.

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