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Case Report

# Bubble Over the Head: Adeloye-Odeku Disease in a Turkish Child-Case Report

## *Kafanın Üzerindeki Baloncuk: Bir Türk Çocuğunda Adeloye-Odeku Hastalığı-Olgu Sunumu*

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**ABSTRACT**

Adeloye-Odeku disease is composed of dermoid cyst over anterior fontanelle, first described in 1971. We present an 11-year-old girl with a soft, fluctuant swelling over bregma. The lesion content was isointense to cerebrospinal fluid on both T2W and FLAIRW images. There was a lytic area under the lesion, seen on CT. The lesion was totally excised. Histopathology confirmed the diagnosis. The case was unique; because a fibrous band was observed extending to superior sagittal sinus and it has never been reported before. Although lesions are sub-aponeurotic, because of this kind of fibrous band, a thorough examination with neuroimaging tools is very important for planning of surgery.

**KEYWORDS:** Bregma, Anterior fontanelle, Dermoid cyst, Neuroimaging, Adeloye-Odeku disease

**ÖZ**

Adeloye-Odeku hastalığı, anterior fontanel üzerinde yerleşimli dermoid kistleri tanımlar ve ilk kez 1971 yılında literatüre girmiştir. Bregma üzerinde yumuşak ve hareketli bir şişlik tespit ettiğimiz, 11 yaşındaki bir kız olguyu sunmaktayız. Lezyon içeriği T2 ve FLAIR ağırlıklı MR incelemelerinde beyin-omurilik sıvısı ile izointensti. Beyin tomografisinde lezyonun altındaki kalvaryumda kemik defekt vardı. Lezyon tamamen çıkarıldı ve histopatolojik tanısı dermoid kist olarak geldi. Bu olguda daha önce bildirilmemiş olan, lezyon ile sagital sinüs arasında fibröz bir bantın varlığıydı. Bu sebeple, bu tür lezyonlar her ne kadar subaponörotik yerleşimli olsalar da, ameliyatı planlamadan önce olguların nöroradyolojik çalışmaları detaylı olarak yapılmalıdır.

**ANAHTAR SÖZCÜKLER:** Bregma, Anterior fontanel, Dermoid kist, Nöro-görüntüleme, Adeloye-Odeku hastalığı

### INTRODUCTION

Dermoid cyst was first described by Cruveilhier in 1829; demonstrated in cranial cavity by Logue and Till (1952), in spinal cavity by Naffziger and Jones (1935) (2). Midline occurrence of dermoids has been known since 1897 (2). They constitute 0.1%-0.5% of all intracranial tumors and 0.1%-0.2% of all skull tumors (3-4). Dermoid cysts over the anterior fontanelle (AF) have a special name as Adeloye-Odeku disease. Adeloye and Odeku are the first authors who reported this entity and described it in detail in 1971 (2). It has had many different names in the literature since then, such as bregmatic dermoid cyst (BDC).

Four Turkish cases were added to the literature by Aslan et al. (4). They were all male patients (4). We present an 11-year-old girl who is the first female BDC patient in Turkish population where proper diagnostic and management tools were used and discuss the current knowledge on BDC.

### CASE REPORT

An 11-year-old girl presented to another clinic with a history of an anterior scalp swelling since birth, progressively increasing in size. She was referred to us because encephalocele had been suspected. Her growth and developmental milestones were normal. There was no neurological deficit. On physical examination, the mass was soft, fluctuant, and was negative for transillumination. Radiological examination revealed a 5 × 5 × 4 cm swelling localized over the AF. Computed tomography (CT) and magnetic resonance imaging (MRI) scans revealed a closed AF with an erosion under the cyst structure (Figure 1A-C, 2A-D). The cyst fluid was isointense to cerebrospinal fluid (CSF) on both T2W and FLAIRW images. The cyst content was isointense to cerebrum on diffusion image. Surgical total excision was managed through a 10 cm elliptical incision (Figure 3A-E). There was a thin band adhering the cyst to the superior sagittal sinus through a little cranial defect on the AF (Figure 3C-black star). The redundant skin was also excised. The contents were yellow and pultaceous. Hair follicles were observed in the cyst (Figure 3E). Histopathology confirmed the diagnosis of a dermoid cyst.

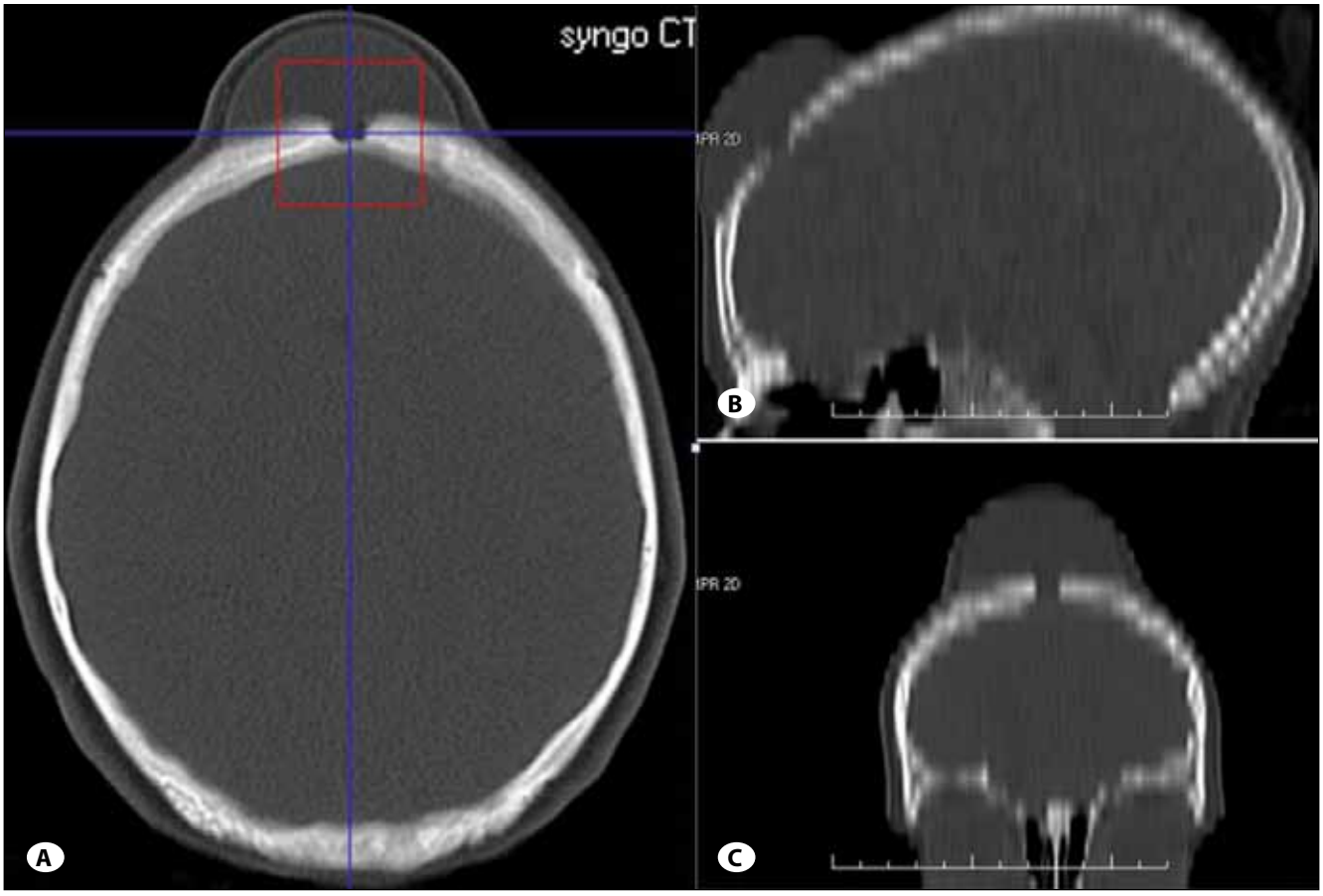


**Figure 1:** Computed tomography image on bone density. The BDC is visible as hypodense mass. There is a bony defect under the mass seen on all planes. **A)** Axial image, **B)** Sagittal reconstructed image, **C)** Coronal reconstructed image.

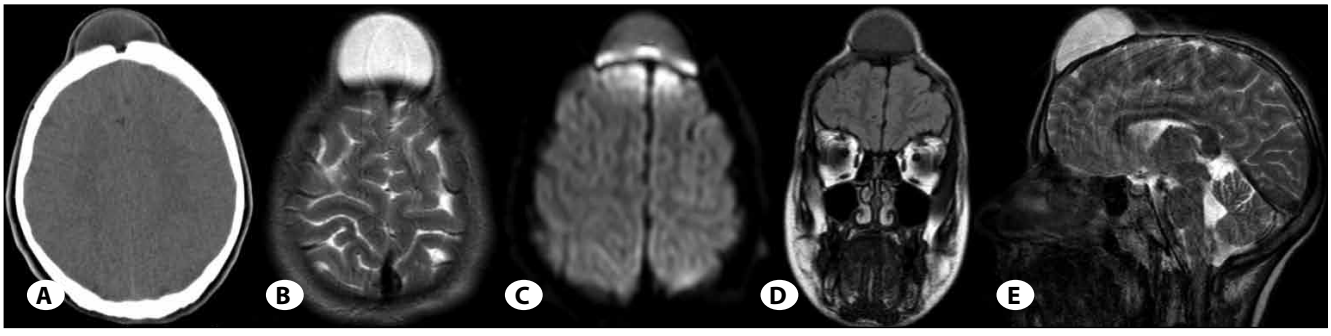


**Figure 2:** The BDC is isodense with the cerebrum on CT **(A)**. It is hyperintense on T2W and FLAIRW MRI **(B, E, D)**. On diffusion weighted-scan, the BDC has an isointense appearance **(C)**. **A)** Axial CT image on cerebrum density, **B)** Axial T2W MRI, **C)** Diffusion-weighted axial MRI, **D)** Coronal FLAIRW MRI, **E)** Sagittal T2W MRI.

## DISCUSSION

Dermoid cysts have 3 types depending on pathogenesis: congenital teratoma type, acquired implantation type, and congenital inclusion type. The congenital inclusion type is composed of sequestered ectodermal cells during embryogenesis (1, 4). They can appear at any site on the skull, but more often over the AF (20%) (4-5). The explanation for this is the affinity of inclusion of skin tissue in suture lines and AF is the largest suture area (5). Dermoid cysts in this region do not have a uniform nomenclature. Due to the first description by Adeloye-Odeku, the entity was named after them.

A literature review clearly shows that there is a predilection for African children, but there are also reported cases from other ethnicities (1, 5). There is no clarity about gender disposition in the literature. Some articles mentioned male dominance and others depicted females as the predominant gender for the

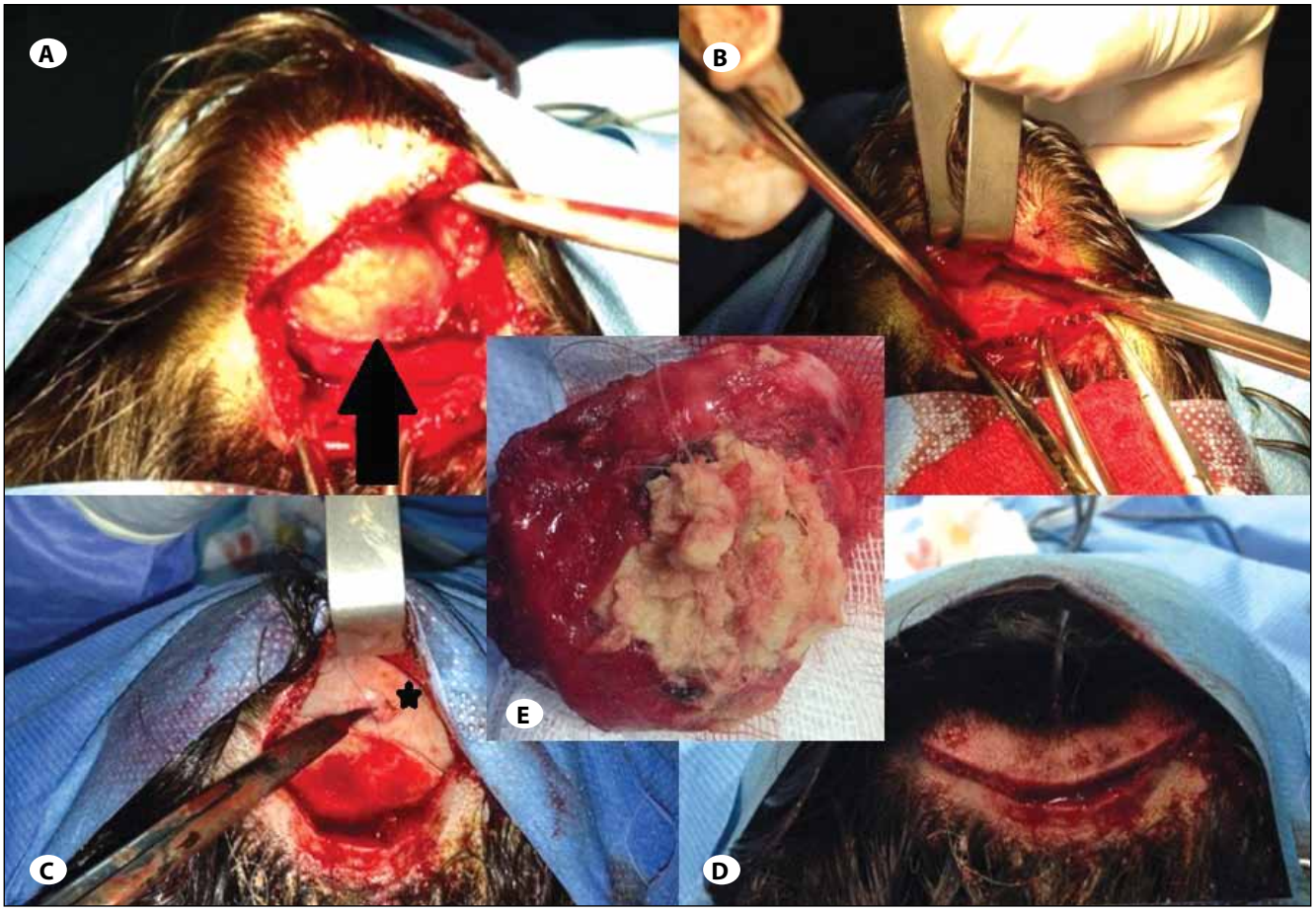


**Figure 3:** Intraoperative images. The yellowish BDC mass is seen under the scalp incision (**A**-black arrow). After retracting the cyst **(B)**, an erosive area is seen beneath it (**C**-black star). The incision type is elliptic **(D)**. Hair follicles in the mass are clearly seen **(E)**.

disease (1-2, 5). Diagnosis is made generally in childhood; but there are some rare adult cases (4). In some reports, the size of the cyst has been related with the age of patient (4-5). The size also enlarges tentatively as the patient strains such as crying (2, 5). Patients apply to clinics with a soft, non-tender mass (4). The mass lesion may present at birth or appear in a couple of weeks after birth (1, 4). There are some other disease entities in the differential diagnosis such as meningoencephalocele, encephalocele, epidermoid, hamartoma, hemangioma, lipoma, lymphangioma, sebaceous cyst, sinus pericranii, cystic hygroma and pilonidal cyst (1-5). Two-thirds of lesions over the AF are dermoid cysts (1).

Magnetic resonance imaging and CT scans are necessary for proper diagnosis and planning the surgery. Bregmatic dermoid cysts are benign lesions but erosion of calvarium may be present (5) such as in our case (Figure 1A-C, 3C). Our case was unique because the dermoid cyst was extending and adhering to the superior sagittal sinus via a fibrous band. This has not been reported for dermoid cysts in AF region, before. Although we did not use it, CT venography could have added some more useful information to our surgical approach. On MRI, the cysts are hypointense on T1W and hyperintense on T2W images. They have low intensity on diffusion-weighted images (1), but our case had an isointense appearance with the cerebrum (Figure 2C). Transillumination is usually positive (2-3, 5); instead it was negative in our case.

Lesions are sub-aponeurotic, so blunt dissection easily results in total excision. Bicoronal or elliptical incision modalities can be applied (2, 4-5). Due to cosmetic reasons, we preferred an elliptical incision model (Figure 3D). Dural tear or leptomeningeal cyst formation may occur during or after surgery (5); but none of these complications occurred in our case. With a bigger cyst, the pressure inside the cyst can erode the lining of the cyst and produce a similar appearance to the rarer epidermoid cysts in this location (5). Because of their different pathogenesis and origin, epidermoid cysts are not expected at this location, especially in the midline (4).

## CONCLUSION

Although our case is the fifth in general; and the first in female gender in the Turkish population, we suspect that many cases have been undiagnosed because of the simplicity of the lesion. Careful preoperative planning is necessary because of the proximity to the superior sagittal sinus and cerebrum.

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