

## FAMOUS MEDICAL CASE REPORTS: CONGENITAL SUBGALEAL CYSTS OVER THE ANTERIOR FONTANELLE IN NIGERIANS (ADELOYE-ODEKU DISEASE)

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### SUMMARY

The “Adeloye-Odeku disease, or congenital dermoid cysts over the anterior fontanelle, is a rare congenital disorder of children initially described at the University College Hospital, Ibadan, Nigeria. The disease is a congenital midline occipital cystic swelling of the head that is operated upon by Neurosurgeons. The case reports by Latunde Odeku and Adelola Adeloye spurred many similar reports from other countries. The report of the disease by the Nigerian Neurosurgeons counts among the world’s most famous case reports.

### Medical Case Reports

A medical case report is a detailed description of symptoms, signs, diagnosis, treatment, and follow-up of an individual patient. Case reports may contain a demographic profile of the patient, but usually describe an unusual or novel occurrence. Some case reports may also contain a literature review of other reported cases. Case reports are professional narratives that provide feedback on clinical practice guidelines and offer a framework for early signals of effectiveness, adverse events, and cost. They can be shared for medical, scientific, or educational purposes. Different types of case reports include an unexpected association between diseases or symptoms, an unexpected event in the course of observing or treating a patient, findings that shed new light on the possible pathogenesis of a disease or an adverse effect, unique or rare features of a disease, unique therapeutic approaches, and a positional or quantitative variation of the anatomical structures. Odeku, Nigeria’s first Neurosurgeon, reported a massive subaponeurotic inclusion cyst in a Nigerian in 1967.<sup>1</sup> In 1971, Adeloye and Odeku reported the abnormality in 18 Nigerians.<sup>2</sup> This condition became known as the Adeloye-Odeku disease.

### Adelola Adeloye and E. Latunde Odeku

Latunde Odeku was born on 29 June, 1927 to the family of Deacon and Mrs Ladipo Odeku. He began his education in 1932 at St Aroloya School, Lagos. From there he gained admission to the prestigious Methodist Boys’ High School, Lagos. A serious minded and brilliant student, he completed his studies in record time. He proceeded to study Medicine at Howard University, North America. In 1955, he returned to Nigeria briefly as a medical officer at the Lagos General Hospital before proceeding to Postgraduate School of Medicine, University of Michigan, to study Neurosurgery. During that period, he worked on the muscle tonus in macaque monkeys. All through his

training and career he displayed extreme competence and expertise. He was by every measure a fine surgeon and a gentleman. In July 1960, as a visiting foreign scientist, he took up Neuropathology at the Armed Forces Institute of Pathology, Walter Reed Army Medical Centre, Washington DC. It was in 1960 that he intensified efforts towards returning to Nigeria to set up a Neurosurgery unit.

In the same year (Nigeria’s historic year of 1960), Adelola Adeloye graduated from the medical school of Nigeria’s premier University, the University of Ibadan. Born to the family of Chief and Mrs. Ebenezer Ajayi Adeloye at Ilesha in Nigeria, Adelola Adeloye started school in 1941 at St. Paul’s CMS Anglican Primary School, Ikole. He later attended Christ’s School, Ado Ekiti and proceeded to the University College, Ibadan to study Medicine. On graduation, he went to the United Kingdom and United States for postgraduate medical studies, becoming a most distinguished Neurosurgeon. On completion of postgraduate training, he returned to Nigeria to join the Neurosurgery Unit of the University of Ibadan and the University College Hospital, Ibadan, headed by Prof Latunde Odeku. Adeloye met Odeku in 1967; the union of the two intellectual giants shaped the terrain of Neurosurgery not only in Nigeria but in the entire continent of Africa. In 1965 and 1967, in case reports published in the Ghana Medical Journal, Odeku described “a certain peculiar solitary cyst not infrequently present in the midline over the anterior fontanel in the infant”.<sup>1,3</sup> This was the beginning of a chain of case reports on curious subgaleal cysts now known as the Adeloye-Odeku disease.

### Adeloye-Odeku Disease

The “Adeloye-Odeku disease” or congenital dermoid cysts over the anterior fontanelle, is a rare congenital disorder of children initially described at the University

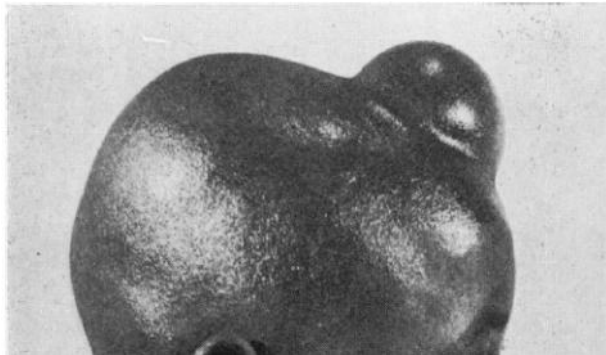


Adelola Adeloye



E Latunde Odeku

College Hospital, Ibadan, Nigeria. The disease was reported by Adeloye and Odeku to be more prevalent in females in a female to male ratio of 2:1; occurring from birth, and resembling an encephalocele. In the 1967 paper, Odeku highlighted the differences between a subgaleal cysts and an encephalocele.<sup>1</sup> He described the cyst as solitary and variable in size, found on the sincipital region of the head over the anterior fontanelle and under the galeal aponeurosis. The cyst was also described as soft, fluctuant, and having a broad sessile base. Between 1959 and 1969, 18 cases were operated upon which constituted the 1971 report.<sup>2</sup> The cysts had no intracranial extension and were easily excised intact. Clinical, radiological, and histological features were similar to congenital inclusion dermoid cysts of the anterior fontanelle.



*The oldest patient (18 years) in the 1971 series<sup>1</sup>*

### **The Differential Diagnosis**

Adeloye and Odeku listed some conditions that may be mistaken for Congenital Subgaleal Cysts. The conditions mentioned were meningocele, encephalocele, dermoid cyst, simple inclusion cyst, sebaceous cyst, lipoma, lymphangioma, haemangioma, cystic lymphoma, and cephalohaematoma.<sup>2</sup>

### **Some subsequent reports of the Adeloye-Odeku Disease**

Following the initial description of the disease in Nigerian by Odeku (1965 and 1967) and Odeku-Adeloye in 1971, other reports were published in

literature by workers from other countries. A search of Google Scholar using the search term 'Congenital Subgaleal Cysts' (done on 24 Dec 2017) returned 1,200 results. The 1971 publication was the first of the results. In 1975 Gellis, Feingold, and Adeloye, reported another case of congenital subgaleal cyst over the anterior fontanel in the *American Journal of Diseases of Children*.<sup>4</sup> In 1982, Chaudhuri and colleagues wrote a paper on congenital inclusion cyst of the subgaleal space.<sup>5</sup> They reviewed published reports on the disease and concluded that although most of the cysts occurred at the anterior fontanel, the cysts could also occur anywhere in the midline, and that the subgaleal space of the anterior fontanel was not an exclusive site. In 1983, a report of a 30-month-old Mexican girl with an inclusion subgaleal cyst located over the anterior fontanel was described by Garza-Mercado and Tamez-Montes.<sup>6</sup> The authors stated that the case was the first diagnosed at their institution and likely the first reported from Mexico. They concluded that the case was another example of such a lesion to be described in an infant of non-African descent. In the same year, a case report of a midline subgaleal epidermoid was published in the journal *Indian Paediatrics*.<sup>7</sup> Over the years the evidence for the uniqueness of the condition reported by Adeloye and Odeku continued to grow.

Another report that drew much global attention to the disease as a unique and new entity was the report of Chaudhuri in 1984.<sup>8</sup> Chaudhuri described the condition in 23 children and stated that the series countered the motion that such cysts were extremely rare in whites and that the anterior fontanel was their only site of location, as reported by various authors from Africa. In 1986, Nicolau and colleagues in a French article wrote on midline subepicranial (subgaleal) dermoid cysts in children. Report of 6 cases and review of the literature.<sup>9</sup> In the same year Wong, Wann, and Lee reported congenital dermoid cysts of the anterior fontanelle in Chinese children. Hubault-Marcade, Hepner-Lavergne, and Pannier reported the disease in a 36 year old adult.<sup>10</sup> The disease has been

reported from different countries in different populations.<sup>11-15</sup> The condition became known as the Adeloeye-Odeku disease.<sup>16-19</sup>

## CONCLUSION

The “Adeloeye-Odeku disease” or congenital dermoid cysts over the anterior fontanelle, is a rare congenital disorder of children initially described in Nigeria. The case reports by Adeloeye and Latunde Odeku count among the most famous case reports globally. Achievements of this magnitude bring prestige to institutions and countries where they were made. Adeloeye and Odeku recognize the uniqueness of the cysts that they encountered among the Nigerian patients they operated on. They had the confidence and courage to report an unknown occurrence at that time. With remarkable powers of examination and skilful documentation, they achieved a feat that only few in the world attain to. Their case report led to global acclaim and more importantly knowledge that has helped thousands of children born with the disorder. Such is the power of a case report. Medical case reports will always be relevant to medical advancement.

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