Department of medical history

Goundou: a historical form of yaws

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Like endemic syphilis and pinta, yaws (also known as framboesia tropica and pian) is a form of non-venereal treponematoses. The causative organism *Treponema pallidum* subsp *pertenue* is morphologically and serologically identical to *T pallidum* ssp *pallidum* (syphilis). These microorganisms could have evolved from a common ancestor, and minor genomic differences have been shown between them.[1]

Yaws mainly affects populations living in humid subtropical countries with poor health care. In the 1920s and 1930s, treatment campaigns that used multiple-dose injections of arsenical preparations and bismuth were launched by mobile health teams in Africa.[2] Until the 1950s, yaws was still a major public health problem in Africa and southern Asia. Since then, mass treatment campaigns with longacting penicillin, supported by WHO, have eradicated the disease in several parts of Asia, and have drastically reduced its prevalence in Africa. Nowadays, yaws has virtually disappeared.[3]

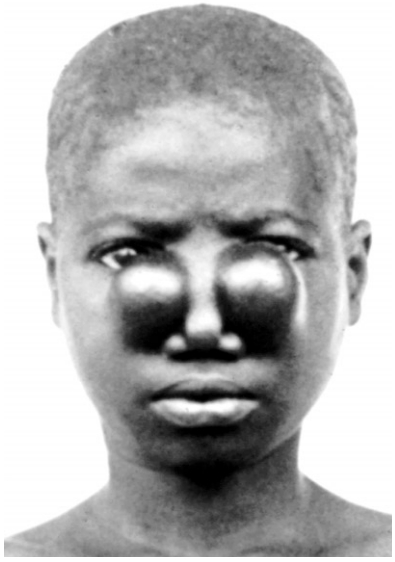

Figure 1: **Typical bilateral goundou in a young boy**
Ivory Coast, 1916 (reproduced by permission of Institut de Médecine Tropicale du Service de Santé des Armées, Le Pharo, Marseille, France).

Figure 2: **Unilateral goundou in a young woman**
Ivory Coast, 1917 (reproduced by permission of Institut de Médecine Tropicale du Service de Santé des Armées, Le Pharo, Marseille, France).

Yaws has three stages. Primary stage (early) yaws usually begins in childhood as a spontaneously healing granulomatous or macular lesion (mother yaws) on a leg. Secondary stage (recent) yaws is characterised by various diffuse skin lesions, and some patients develop osteoperiostitis. Third stage (late) yaws features severe skin ulcers and sometimes bone destruction causing severe disabling deformities and disfiguration. Late lesions include mutilating facial ulcer (gangosa) and gummatous skin lesions.

Goundou is a manifestation of recent yaws, with proliferative exostoses. This disease was rare even when yaws was hyperendemic in the early 1900s, and has now disappeared. Because of its highly stigmatising effect on the patient's appearance, goundou posed a nosological and clinical problem for doctors at the beginning of the 20th century.

In most cases, goundou presented as paranasal swelling, characterised by two large hard tumours that did not adhere to the superficial skin layer. Tumours were round or oval, with an oblique long axis running from the base outward (figures 1 and 2). Pus formation and associated adenopathies were never seen. The tumours could be painful to touch and could develop over several years, beginning with the appearance of diffuse thickening or deposits on the ascending branches of the maxilla. Large lesions could cause blindness by reducing the visual field, sometimes in association with severe infection of the eyeball, and with nasal obstruction.

Involvement of other bones of the face was generally associated with goundou and, in rare cases, was exclusive. In these forms of the disease, hypertrophy pushed the palate into the buccal cavity and enlargement of the alveolar ridge caused wide spacing between teeth. Sometimes tumour growth was so extensive that the whole rhinomaxillary region was pushed forward, resulting in impairment of both breathing and swallowing (figure 3).

Horned men

Although yaws occurred throughout the intertropical zone, goundou was reported mainly in sub-Saharan Africa. The first case was described in 1882. In the 19th century, when Europeans regarded Africa as a mysterious continent, goundou became a source of concern and controversy for doctors. When, in 1883, Alexander MacAlister presented the first picture of a horned man taken by one of his correspondents in the Kingdom of Dahomey (Republic of Benin, western Africa), the Royal Academy of Ireland proposed an explanation that was based on racial atavism.[4] However, the pathological origin of the findings was soon recognised, and more than 50 cases were described before the turn of the century. Goundou was reported in populations of black people not

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only in regions of sub-Saharan Africa—such as the Gold Coast, Ivory Coast, Angola, Zanzibar, and Central Africa—but also in New World countries, such as Jamaica, Brazil, Trinidad, and the southern USA. A few cases were even reported in Asia.[5-16]

The populations that were affected by goundou called the disease by various names. The most widely used terms cited by western physicians in Africa were *Anàkhré* and *Henpuye* (dog-nose). More limited use of the names *Toupakié* in the Baoule language and *Noukrou* in the Bambara language was also reported. The term goundou comes from the dialect used by the Agni people in Ivory Coast, and is the only name that is still used in medical publications.

Disfigurement, especially in severe cases of goundou, had serious social repercussions. Intercultural differences apart, anyone can perhaps understand why patients frequently became outcasts. In 1896, the Agni people in the southern part of Ivory Coast, thought goundou was the work of a personal fetish of King Bettie, called Jero or Zore, who was symbolised by a statuette with two balls on his face.[8,9] The fetish was believed to be a chosen and loyal subject of the king, but people began to think that his powers could turn against the user when the king's son also developed goundou. King Bettie's descendants officially denounced this superstition in 1935. No other folk tales surrounding goundou have been reported.

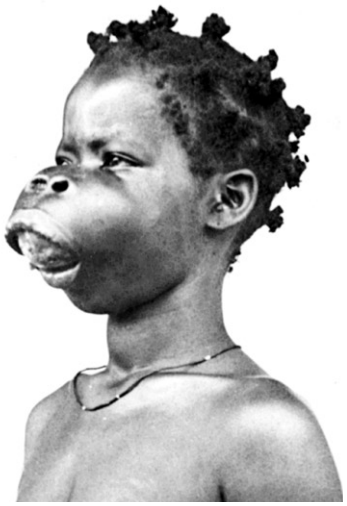

Figure 3: **Diffuse maxillofacial goundou with protrusion of all the maxillae and palate**
Ivory Coast, 1917 (reproduced by permission of Institut de Médecine Tropicale du Service de Santé des Armées, Le Pharo, Marseille, France).

Aetiological debate

After ruling out other causes, such as race, heredity, and trauma (sequelae of tribal scarring rituals), the medical world began to debate the aetiology underlying the features seen in these horned men. Several explanations were proposed, including infection induced by insect larvae penetrating into the nasal cavities, and a disorder resembling Paget's disease.[7] In his insightful analysis published in *The Lancet* in 1900, A J Chalmers[7] mentioned the connection between goundou and the primary lesion of yaws. Some ethnic groups in the Ivory Coast had already noted this connection, and designated mother yaws by the same names—ie, yaws and goundou.[16]

From 1912 to 1917, Paul Botreau-Roussel of the French colonial army medical corps served as a surgeon on the railroad ambulance in the Ivory Coast, which was a hyperendemic region for yaws at that time. On the basis of a personal series of 130 cases, he provided the most thorough epidemiological, clinical, and pathophysiological description of what he called pianic osteitis/goundou.[17,18] These data confirmed the link between goundou and yaws that Chalmers described. The geographic zones of goundou and yaws coincided. Goundou consistently occurred as a concurrent (44%), short-term (49%), or long-term (7%) complication of yaws. In most cases, lesions were associated with diffuse osteoperiostitis,

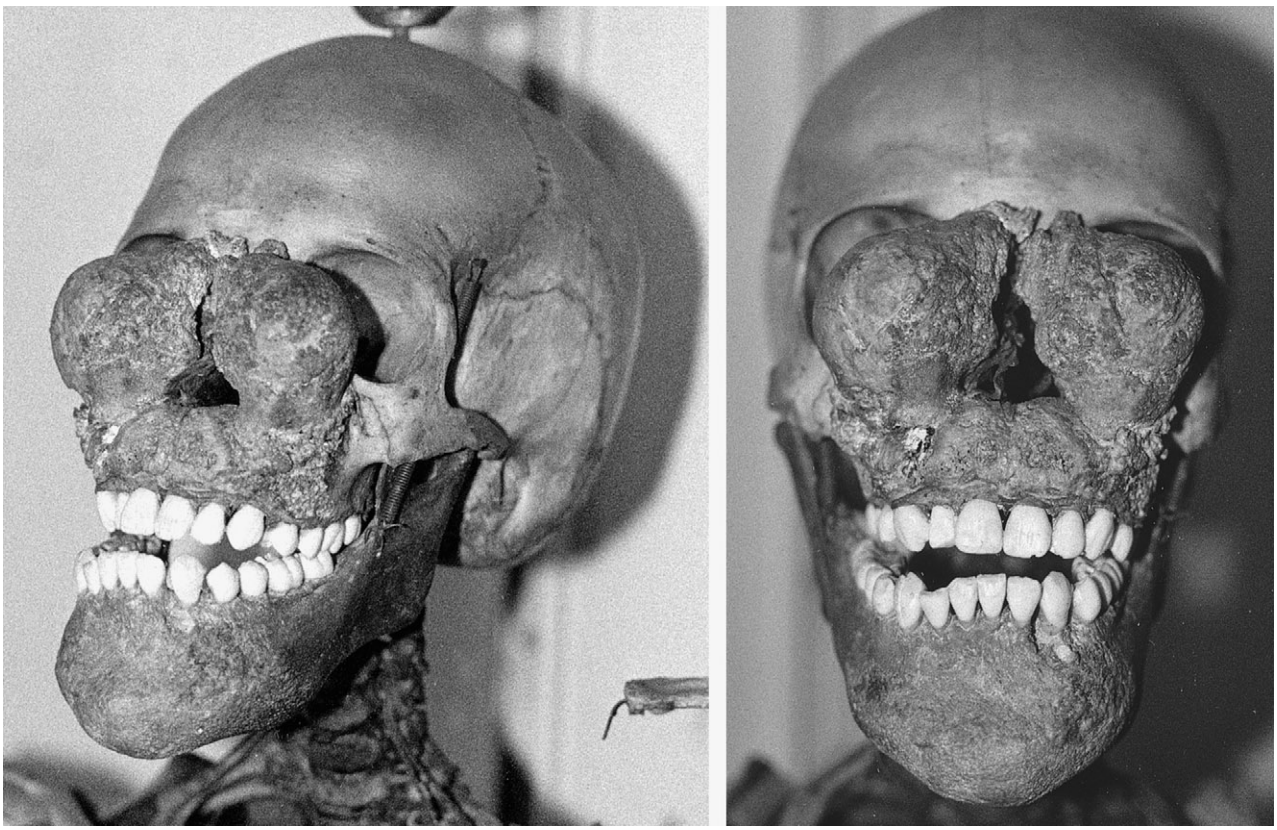

Figure 4: **Bilateral goundou on a skeleton of an adolescent**
Left, the upper jaw and mandible are deformed by an osteoperiostitis but the rest of the skull is intact. Right, the round excrescences caused a major reduction of the visual field and nasal aperture.

mainly on the tibia, which resulted in a swordblade-like appearance. The bones of the forearms, fingers, hands, and feet were also affected. Postcranial lesions, which were often painful, could be associated with skin lesions at the recent and late stages. One remaining source of controversy was histological specimens that confirmed similarity between goundou and postcranial yaw lesions, but were devoid of treponemas and showed no microbial activity in conjunctival and bone tissue.[17] Despite this compelling evidence, the aetiological debate continued. In 1963, Hackett questioned the link with yaws.[19] His main objection was based on the low frequency of goundou in endemic areas for yaws in Africa.

Examination of the skeleton of a teenager who died during an operation by Botreau-Roussel provides a unique opportunity to understand goundou. The body was donated to science by the boy's family and is stored at the Dupuytren Musée de Médecine in Paris, France. Photographs of the patient in life show an undernourished young man with typical features of goundou, including major deformation of both tibias. We examined the skeleton (figure 4) and noted two round excrescences that greatly obstructed the visual field (75%) and nasal aperture. The upper jaw and mandible were deformed because of osteoperiostitis, but the skull was intact. Deposits were also seen on the long bones of the arms, fingers, and especially on the tibias, which have the typical swordblade appearance of pian. These findings confirm clinical observations and the classification of goundou as a form of yaws.

There is general agreement among the WHO and all clinicians with the hypothesis set forth by Chalmers and Botreau-Roussel. Goundou has been classified as an osteoperiostitis of recent yaws.

Eradication of goundou

Surgical resection was the mainstay of treatment for goundou. In patients with limited forms of the disease with two paranasal excrescences the operation was simple, because there was an easily identifiable subperiosteal cleavage plane. Such operations were done by H Strachan in Jamaica in 1894,[6] A J Chalmers in Africa in 1900,[7] and P Mendes in Brazil in 1901.[13] In the Ivory Coast in 1912, Botreau-Roussel operated successfully on a patient with goundou, with the assistance of a Canadian surgeon named J N Roy who was visiting Abidjan from Montreal. In 1916 Botreau-Roussel treated two other patients who had learned about this initial success. As the news spread, 130 patients were brought in free of charge by the colonial government for treatment by the so-called bump remover. A total of 115 patients underwent surgery, with only one death. The other 15 patients were contraindicated because of diffuse lesions of the palate, which were beyond the reach of these skilful surgeons with few limited technical resources.

Surgery was not the only weapon against suspected yaws. At the beginning of the 20th century, early forms of the disease could be treated with treponemicidal drugs containing arsenic derivatives, and later antibiotics were used. However, even without treponemicidal treatment, goundou did not recur after surgical excision. In patients who presented with inoperable late forms, treatment with drugs caused the disease to stabilise, mucosal inflammation to disappear, and facial deformity to regress, but new bone growth persisted.

Mass treatment campaigns in endemic areas have now eradicated goundou. Only three minor, nosologically debatable cases have been reported in the past 30 years.[20–22] Even in hyperendemic areas at the beginning of the century, the incidence of goundou was difficult to estimate in the absence of accurate epidemiological data. In 1917, the goundou-to-yaws ratio was estimated at one in 150 cases in the Ivory Coast, and one in 600 in Central Africa.

Several questions remain unanswered. Why does yaws seem to be the only treponematoses associated with inflammatory hypertrophy? What are the underlying physiopathological mechanisms? Why did these tumours develop on the internal branches of one or both maxillary bones in some patients, and over the whole face in others? Why was the incidence higher in western Africa than in other tropical areas? A possible reason for this high incidence is that the socioeconomic and climatic conditions in those regions were especially unfavourable at that time.

The endemic treponematoses are not yet eradicated, but thanks to the courage of early pioneers such as Chalmers and Botreau-Roussel, goundou belongs to the history of tropical diseases.

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Conflict of interest statement
None declared.

References

1 Zimmer C. Can genes solve the syphilis mystery? *Science* 2001; **292:** 1093.
2 Meheuse A, Antal G M. The endemic treponematoses: not yet eradicated. *Rapp Trimest Statist Sanit Mond* 1992; **45:** 228–37.
3 WHO/World Health Organization. The world health report 1998: executive summary, life in the 21th century : a vision for all. http://www.who.int/whr/2001/archives/1998/exsum98e.htm (accessed Sept 23, 2002).
4 MacAlister A. Further evidence as to the existence of horned men in Africa. *Royal Irish Academy* 1883; **2:** 771–73.
5 Llamprey JJ. Horned men in Africa: further particulars of their existence. *BMJ* 1887; **2:** 1273–74.
6 Strachan H. Bony overgrowths or exostoses in the West Indian Negro. *BMJ* 1894; **1:** 189-90.
7 Chalmers AJ. Report on "Henpuye" in the Gold Coast colony. *Lancet* 1900; **1:** 20–23.
8 Maclaud. Notes sur une affection désignée dans la boucle du Niger et le pays de Kong sous les noms de Goundou et Anàkhré (gros nez). *Arch de Méd Navale* 1895; **63:** 25–33.
9 Cannac. Note sur le Goundou ou Anakré. *Arch de Méd Navale* 1904; **31:** 89.
10 Wellman FC. Goundou in southern Africa. *J Trop Med* 1905; **7:** 286.
11 Friedrichsen. Goundou. *J Trop Med* 1903; **6:** 62.
12 Shircore JO, Edin MB. Goundou. *BMJ* 1910; February 26: 503.
13 Mendes P. A propos d'un cas de goundou ou anakré. *Rev Chir* 1901; **21:** 445.
14 Seheult R. Note on a case of goundou. *Lancet* 1915; **1:** 72.
15 Branch CW. Case of goundou in the West Indies. *J Trop Med* 1909; **5:** 63.
16 Braddon W. Leonard. Note on the occurrence of "goundou" or gros-nez in the Malay peninsula. *J Trop Med* 1901; **4:** 170.
17 Botreau-Roussel P. Ostéites pianiques "Goundou". Paris: Masson, 1925.
18 Botreau-Roussel P. Clinique chirurgicale des pays chauds. Paris: Masson, 1938.
19 Hackett CJ. On the origins of the human treponematoses (Pinta, Yaws, Endemic syphilis and venereal Syphilis). *Bull World Health Organ* 1963; **29:** 7–41.
20 Trenouth M J. Goundou-tertiary yaws in the maxilla. *Br J Oral Surg* 1975; **13:** 166–71.
21 Draws J, Blake P, Draf W. Frambösie: ein seltener fall eines invasiv wachsenden tumors der kieferhöhle. *HNO* 1988; **36:** 426–28.
22 Elango S, Palaniappan S P. Nasal manifestation of yaws. *Ear Nose Throat J* 1989; **68:** 873–75.