Human oesophagostomiasis: clinical presentations and subclinical colonic pathology

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CHAPTER 2

Clinical epidemiology and classification of human oesophagostomiasis

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ABSTRACT

The intestinal helminth *Oesophagostomum bifurcum* is highly and focally endemic in northern Ghana and Togo, and its juveniles produce a nodular inflammatory response as they develop in the intestinal wall. This pathology can produce clinical symptoms.

The 156 cases of oesophagostomiasis presenting to Nalerigu hospital in northern Ghana between 1996 - 1998 are reported. The disease accounted for 0.2% of the out patient department new presentations (approximately 1 patient per week), and 1% (16) of the major acute surgical cases. Children between 5 and 9 years were most commonly affected.

Multinodular disease (13% of the cases) results from hundreds of pea sized nodules within the colon wall and other intra-abdominal structures, and presents with general abdominal pain, persistent diarrhoea and weight loss. Dapaong tumour (87%) presents as an abdominal inflammatory mass often associated with fever. The tumour is painful, well-delineated, smooth, spherical, 3 - 6 cm, 'wooden', periumbilical, and adhered to the abdominal wall.

Cases most commonly presented during the late rains and early dry season. Diagnosis by ultrasound has reduced the need for exploratory surgery, and the ability to sonographically evaluate conservative treatment with albendazole has curtailed management by colectomy or incision and drainage.

INTRODUCTION

Oesophagostomum bifurcum is a human intestinal helminth occurring in parts of West Africa. The distribution of the infection is highly focal, with approximately 250,000 people infected - measured by stool cultures - and 1 million people at risk [43;49]. The juvenile stages of the parasite develop within the intestinal wall, eliciting an inflammatory, granulomatous reaction [150]. These nodular lesions can present with clinical disease: 61 cases of oesophagostomiasis have previously been reported from northern Ghana and Togo.[15;56;57]

The importance of oesophagostomiasis within this locality is suggested by the fact that several of the affected people groups have names for the disease in their languages, and instantly recognise the condition when it is described to them. Indigenous healers and medicine men know of 'Turtle in the belly', and use several treatments to alleviate it, including incision and drainage with locally made knives [116].

Despite 53 case reports of the disease from other areas of the world, including Ivory Coast [58;59], Guinea [60], Kenya [61], Uganda [20;62-67], Sudan [68], Ethiopia [69], Zimbabwe [70], Brazil [71], Indonesia [72;73], Brunei [74] and Malaysia [75], nothing is known of the endemic situation in these places, although there have been two reports of adult Oesophagostomum worms being found in the stool of Nigerians [76;77].

Until 1991, human infection with *O. bifurcum* was assumed to be a zoonosis with primates as the reservoir. However, in northern Ghana and Togo, it was discovered that people are the definitive host in whom it can complete its life cycle, and that the monkey population within this area is too small to provide a reservoir for an infection of such prevalence [43]. In 7 days of moist conditions, eggs excreted with stool develop on the ground into infective 3rd stage larvae, which are probably ingested. They develop within the intestinal wall before inhabiting the colonic lumen and laying eggs.

The 114 cases of oesophagostomiasis reported in the literature have mostly been diagnosed from pathology specimens, and clinical details have been sparse. 42 were confirmed by identification of worms removed from resected bowel. Colectomy was the recommended management procedure.

There is little accurate knowledge among the medical profession within northern Ghana of the characteristic symptoms and distribution of oesophagostomiasis [49], and

consequently, the disease is often missed. Despite the multiplicity of cases described, this is the first report to classify oesophagostomiasis into distinctive clinical presentations, and the first to examine the clinical epidemiology of the disease.

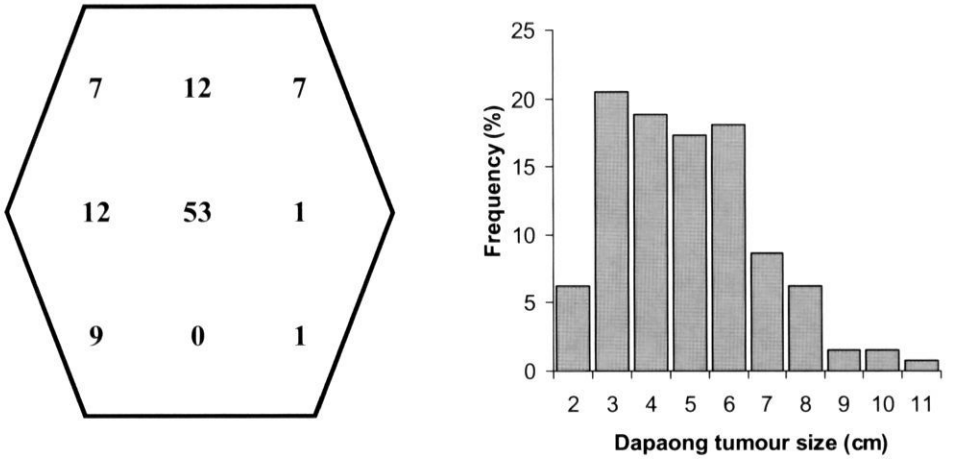
METHODS

From January 1996 until October 1998, weekly oesophagostomiasis clinics were held at the Baptist Medical Centre (BMC) hospital in Nalerigu, in the Northern Region of Ghana some 36 miles from the Togo border. Suspected cases of oesophagostomiasis presenting to the hospital out patient department (OPD) were referred to these clinics by the hospital clinicians, before or after treatment, depending on their seriousness. The patients were reimbursed transport costs to and from the clinic, and all tests and treatment were provided free. The existence of the clinic was advertised to all health posts in the area.

Table 1: Signs and symptoms of multinodular disease and Dapaong tumour. Key: [×] General abdominal pain, [†] Pain localised to the nodule.

Presenting signs and symptoms	Multinodular disease		Dapaong tumour	
	Number of patients	% (n = 20)	Number of patients	% (n = 136)
Abdominal pain	17 [×]	85 [×]	121 [†]	89 [†]
Fever associated with the pain	14	70	79	58
Decreased appetite	12	60	36	26
Vomiting	12	60	32	24
Symptoms of self-resolving ileus	11	55	32	24
Mucus stool	12	60	31	23
Diarrhoea	17	85	29	21
Weight loss	15	75	12	9
Mass adhered to abdominal wall	-	-	112	82
Mass adhered to skin	-	-	27	20
Skin shiny/elephantine	-	-	17	13
Fistula to skin surface	-	-	4	3

Abdominal history and clinical examination were conducted with the help of the same translator using a semi-structured questionnaire comprising closed and open ended questions. Abdominal palpation and sonographical assessment of the abdomen in all sections, including an evaluation of the colon throughout its course, was performed. The decision to treat conservatively or to perform surgery was taken by one of the hospital doctors, and patients were persuaded to continue attending the clinic until they no longer had signs or symptoms of the disease.



Figures 1a & b: Abdominal sector distribution and size of Dapaong tumours (% of 165 tumours from 136 cases).

PATIENTS

156 cases of oesophagostomiasis presented to Nalerigu BMC hospital during the 34 months of the study. These cases represent 0.2% of all new presentations to the hospital OPD, or, approximately one new patient every week, an incidence comparable to gonorrhea, tuberculosis, typhoid fever, and infectious hepatitis at this hospital. 50 (32%) of the cases underwent surgery: 34 had incision and drainage, and 16 had laparotomy. These laparotomies accounted for approximately 1% of the acute major surgical procedures performed at BMC during this period.

Diagnosis of the 73 cases seen during the first 18 months of the clinic was based on

history and clinical examination, or was made during exploratory surgery. The criteria used was based on the previous experience of the BMC doctors. Ultrasound was introduced at the clinics from July 1997 onwards, and was used to assess the 83 patients who presented during the remaining time.

Nine suspected patients referred to the clinic were unable to attend, and 35 patients seen at the clinic were not included in this analysis because the evidence that they had oesophagostomiasis was insufficient.

Presentations

The cases are divided into two distinctive presentations: multinodular disease and Dapaong tumour.

Table 2: Surgical management of Dapaong tumour.

Year (total number of cases)		1996 (n=56)	1997 (n=49)	1998, J-O (n=31)
Number of cases (%)	Incision & drainage	18 (32)	10 (20)	5 (16)
	Laparotomy	4 (7)	4 (8)	2 (7)

Multinodular disease

20 (13%) of the 156 patients were classified as having multinodular disease, typified by hundreds of pea-sized nodules within the wall of the colon, together with gross thickening and oedema of the colon wall. The most common presenting symptoms (Table 1) were general abdominal pain, persistent diarrhoea and weight loss. The intense, short-lived, colicky pain of temporary, self-resolving intestinal obstruction was described by 11 (55%) of the patients. A stool culture [43] was positive for *O. bifurcum* third stage larvae in 13 (65%) of the cases, and the colon was palpable in 10 (50%) of the cases. Eosinophilia (>10%) was found on the differential count in 3 of the 12 cases tested (25%).

Pale nodules between 5 and 15 mm in diameter were always detected within the wall of the entire colon, and a few nodules were noticed on the terminal ileum in 4 out of the 6

laparotomy cases. In one case, nodules were also found studding the liver and pelvic wall. White, 9 - 13 mm long *O. bifurcum* juveniles were always discovered alive in the nodules within thick yellow/green pus. The female worms tended to be larger than the males, and did not contain eggs.

Before the use of ultrasound became a routine at the clinics in July 1997, 2 of the 4 cases of multinodular disease recorded were not diagnosed until during exploratory laparotomy, and 3 of the 4 cases eventually underwent colectomy. One of these patients was treated for 5 days with 10 mg / kg / day albendazole, but continued to deteriorate. During subsequent surgery, several of the nodules were opened to reveal non-moving and degenerated *O. bifurcum* juveniles. (Interestingly however, a live worm was found free in the peritoneal cavity.) After the introduction of ultrasound to the clinics, 16 cases

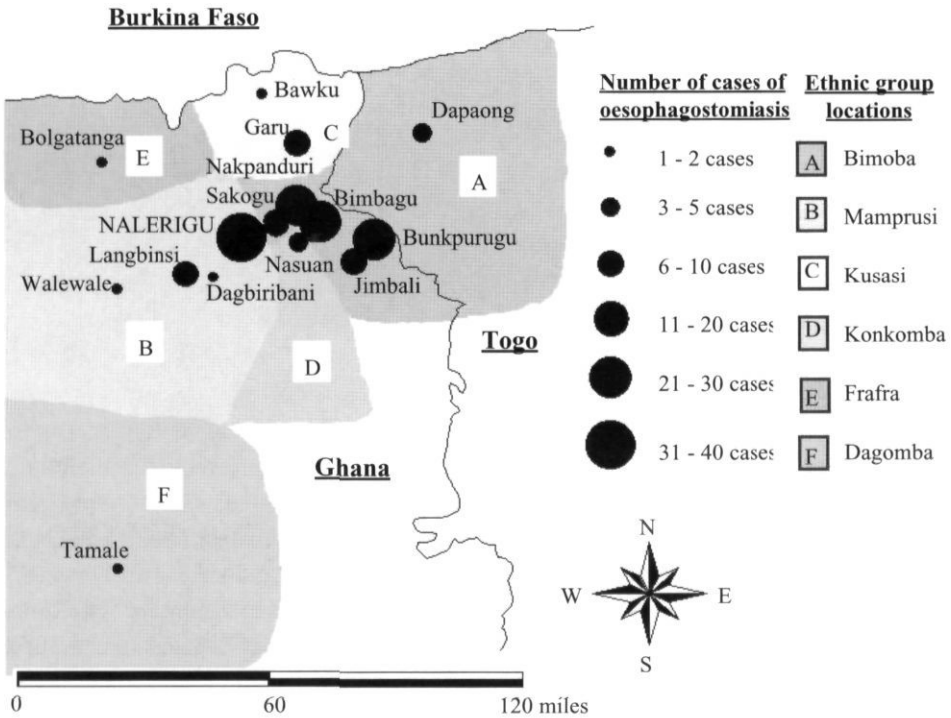


Figure 3: Geographical distribution of oesophagostomiasis cases presenting to Nalerigu hospital.

of multinodular disease were diagnosed, in comparison to the 4 identified in 1996. Conservative chemotherapy with albendazole (10 mg / kg for 5 days) was attempted in all 16 cases, using ultrasound to monitor the efficacy of therapy, and all 16 patients recovered without the need for colectomy. The sonographical appearance of multinodular disease has been confirmed by exploratory laparotomy [151].

Table 3: Ethnicity of 156 cases of oesophagostomiasis in relation to the ethnicity of out patient presentations for all diseases

People group	Oesophagostomiasis		All diseases (%)
	Number of cases	%	
Bimoba	86	55.1	10.3
Mamprusi	38	24.4	28.4
Kusasi	12	7.7	7.7
Frafra	7	4.5	7.4
Busanga	6	3.8	5.4
Konkomba	3	1.9	6
Moshi	2	1.3	5.5
Dagomba	1	0.6	10
Talansi	1	0.6	1.1

Dapaong tumour

During the study period, 136 patients with a Dapaong tumour presented to the clinic. The most common presentation was a painful, well-demarcated, smooth, spherical, 'wooden', 3 - 6 cm peri-umbilical mass adherent to the abdominal wall and associated with fever (Table 1, and Figures 1a & b). Involvement of the abdominal wall was very common, as the inflammatory process infiltrated through the musculature sometimes incorporating the skin, particularly when the tumour was deep to the linea alba. In these cases, the skin became shiny, progressing to a cracked dull elephantine appearance as the condition resolved. Tumours deep in the abdomen were not locally painful. Intense, short-lived, colicky pain and vomiting were described by 24% of the patients. 36 cases (26%) had a stool culture positive for *O. bifurcum* third stage larvae. The median tumour size was 5 cm, with a minimum of 2 cm and maximum of 11 cm. Eosinophilia (>10%)

was found on the differential count in 6 out of the 62 cases tested (10%).

Most (88%) of the individuals with a Dapaong tumour had 1 solitary mass, but occasionally, multiple discrete masses and conglomerations of tumours were found. 5 (7%) of the 69 patients seen before the use of ultrasound were found to have more than 1 tumour by palpation, and 11 (16%) of the 67 cases analysed by ultrasound were found to have multiple tumours.

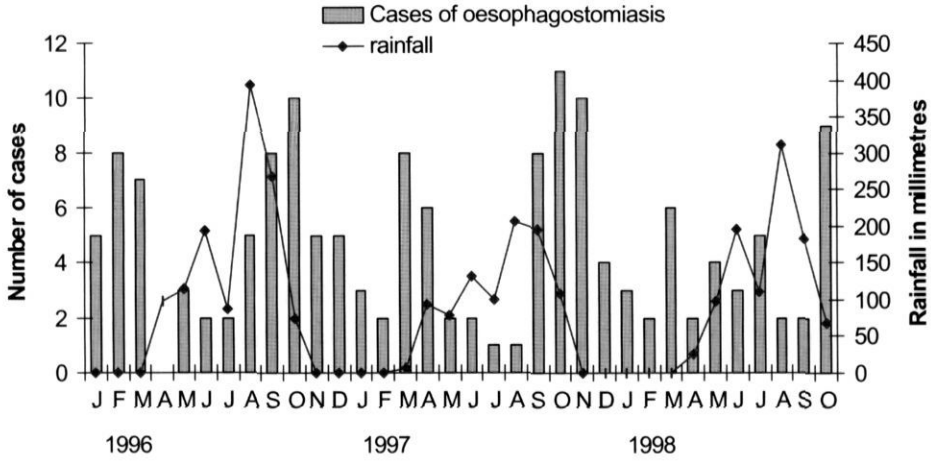


Figure 4. Presentation seasonality of clinical oesophagostomiasis (n = 156).

Surgery was performed in 43 (32%) of the cases: laparotomy in 10 (23%) of these, and incision and drainage (without sonographical assistance) in 33 (77%). The surgical management pattern throughout the 3 years is shown in Table 2, illustrating a shift away from the use of incision and drainage. 10 mg / kg body weight albendazole for 5 days was used instead. The laparotomy cases presented with acute abdomen secondary to tumour rupture and abscess formation (6 cases), and small bowel obstruction secondary to inflammatory adhesion formation around the tumour (4 cases). Thick creamy yellow/green pus was always discovered in the tumours, and worms were located in 16 (37%) of the surgical cases. Tumours contained 1 worm each, which were living, white, and 7 - 13 mm long. The female worms tended to be larger than the males, and did not contain eggs. There was no apparent seasonality of the cases needing surgery.

Gender and age distribution of clinical oesophagostomiasis

The 20 cases of multinodular disease were evenly distributed between males and females, and these patients had a median age of 16 years. 80 males and 56 females presented with Dapaong tumour, and had a median age of 7 years. The proportion of oesophagostomiasis due to multinodular disease rather than Dapaong tumour increased with age (Figure 2). In 1996, 17% of patients presenting to the Nalerigu OPD for all conditions were less than 15 years old, and 47% were males (1996 hospital data). During the 3 years of the clinic, 82% of the oesophagostomiasis patients were less than 15 years old, and 58% were males.

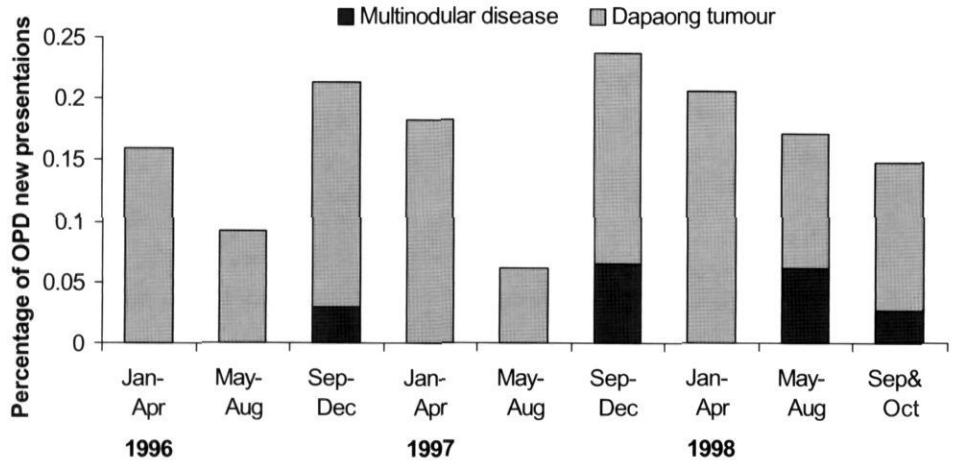


Figure 5: Cases of oesophagostomiasis as a proportion of all OPD presentations (156 cases out of 94,232 OPD presentations)

Ethnicity of clinical oesophagostomiasis

The geographical distribution of cases was centred around Nalerigu hospital. The areas with the highest prevalence of oesophagostomiasis are inhabited by the Bimoba people group (Figure 3). Bimobas account for more than half of the oesophagostomiasis cases, and Mamprusi make up almost one quarter (Table 3). Bimobas are highly over-represented at the OPD for oesophagostomiasis compared to their presentations for all diseases, and Dagombas, the people group inhabiting the Northern Region capital, are highly under-represented for oesophagostomiasis. (Ethnic presentations for all disease

data based on a survey between August and October 1998.) All the patients belonged to people groups from northern Ghana and Togo, despite 5.4% of OPD presentations being from people groups of southern origin. Fulanis and Kusina account for 3.0% and 2.4% of the OPD presentations respectively, but were not found with oesophagostomiasis.

Presentation seasonality of clinical oesophagostomiasis

Presentation of clinical oesophagostomiasis to the OPD in Nalerigu shows a modest degree of seasonal variation, with more cases occurring during the late rains and early dry season (Figure 4). When the cases are given as a percentage of total new OPD presentations, to overcome other factors affecting OPD presentation, this pattern is retained (Figure 5). Multinodular disease was diagnosed in the early rainy season for the first time in 1998 (Figure 5). In 1996 and 1997, it had been found only in the late rainy season. It should be noted that a diagnosis of malaria is given for approximately 40% of OPD presentations.

DISCUSSION

Whilst oesophagostomiasis is rare from a global perspective, the cases presented here represent a significant health problem for northern Ghana. Awareness of the disease within this region would lead to reliable and prompt diagnosis and more effective management. The majority of cases in this study were diagnosed on the basis of characteristic clinical, surgical and sonographical findings. Juvenile *O. bifurcum* worms were found in all surgical cases of multinodular disease, and in 37% of the surgical cases of Dapaong tumour.

Previous descriptions of clinical oesophagostomiasis in the literature correspond with our classification of clinical oesophagostomiasis, but whilst the distinction between multinodular disease and the Dapaong tumour is certainly well-defined, it is not indisputable: 12% of the cases labeled as Dapaong tumour had more than one lesion. These lesions can be present as apparently asymptomatic nodules only visible by ultrasound, or patients may have multiple symptomatic tumours requiring attention.

Use of stool cultures as a diagnostic method for clinical oesophagostomiasis is conceptually inappropriate: the nodular pathology is caused by trapped juvenile (sexually immature) worms within the colon wall. 74% of the cases of Dapaong tumour were stool negative, indicating the unsuitability of stool cultures as a diagnostic test. However, the high proportion of stool positive cases with multinodular disease suggests that stool

cultures are a useful marker of this presentation. Eosinophilia was present, but this finding is not unusual in a rural African community.

Females are found to be more frequently and intensely infected with *O. bifurcum* than males using parasitological methods (stool cultures) of assessment [43] but males more often present with clinical oesophagostomiasis. This pattern may simply reflect regrettable family gender priorities, or it could have an immunological basis. The Dapaong tumour is predominantly a disease of the middle childhood years, and multinodular disease is evenly distributed throughout the age groups.

From parasitological surveys, the areas most highly infected are inhabited by Bimobas followed by Kusasis and Mamprusis (unpublished data). Nalerigu is the principal Mamprusi town, on the border with the Bimoba area and not far from the Kusasi area. A hospital visit usually necessitates an over-night stay, and consequently, people go to Nalerigu only if they have extended family living in the town. Most of the general OPD presentations are by Mamprusis and Bimobas. Kusasis and Frafras have hospitals in their own main towns, and an escarpment just north of Nakpanduri is a natural obstacle to BMC presentation for the Kusasi people. The shift in distribution of cases into the Bimoba area and the high over-representation at the OPD of Bimobas with oesophagostomiasis both suggest that this people group have a propensity for oesophagostomiasis.

Transmission of *O. bifurcum* occurs during the rainy season [51] , and the seasonal distribution of clinical oesophagostomiasis - most common during the late rainy season and early dry season - fits with this observation. However, the presentation to hospital of sub-acute disease is governed by several factors. Families are farming during the rainy season, their food for the following year, and hence do not have time or money to spare for hospital visits. The number of hospital presentations may be an underestimate of the real number of cases of clinical oesophagostomiasis.

The possibility of diagnosis by ultrasound has reduced the need for exploratory laparotomy in multinodular disease. The ability to sonographically evaluate treatment with albendazole has decreased the number of incision and drainage procedures performed on cases of Dapaong tumour, and has curtailed the use of colectomy in cases of multinodular disease. Knowledge of the sonographical appearance of multinodular disease has increased the number of cases identified at BMC.

During the past decade at the Nalerigu hospital, oesophagostomiasis has become recognised as a significant disease entity. However, in this community, where many cannot afford hospital care unless in a life and death situation, the actual impact of the disease remains uncertain.

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