

OUT OF THE FRYING PAN INTO THE FIRE?: GUINEA WORM DISEASE (DRACUNCULIASIS) AND CHRONIC FLUOROSIS IN THE TRIBAL PEOPLE OF THE SCHEDULED AREA OF RAJASTHAN, INDIA

ABSTRACT: The campaign to eradicate Guinea worm disease (dracunculiasis) in the tribal people of the scheduled area of Rajasthan, India, including the provision of borehole water, has been complicated by the emergence, over the past 40 years, of an arguably more serious new disease for the area, chronic fluorosis. This needs to be addressed to satisfactorily complete the mission of dracunculiasis eradication.

Keywords: Dracunculiasis; Guinea worm disease; India; Scheduled area of Rajasthan.

A review in the current issue of *Fluoride* by Shanti Lal Choubisa draws attention to the development in the past 40 years of chronic fluorosis in the tribal people of the scheduled area in Rajasthan, India.¹ The factors that have led to this have been industrial fluoride pollution and a change from using surface water, dug wells, and step wells for drinking and cooking to using groundwater. The use of groundwater, obtained from boreholes, tube-wells, and hand pumps, has been part of a campaign to rid the area of Guinea worm disease (dracunculiasis) which is usually spread by drinking water containing copepods (water fleas) infected with larvae of the parasitic nematode worm *Dracunculus medinensis* L. Although the groundwater has been safe from *Dracunculus medinensis* contamination and helped to almost eradicate dracunculiasis from the tribal area of Rajasthan, it has been at the price of introducing a new health problem, chronic fluorosis, which causes an arguably greater level of morbidity. To complete the mission of ridding the area of dracunculiasis, it is necessary to ensure that water sources are provided which are safe from not only Guinea worm disease but also from chronic fluorosis.

Dracunculiasis, Latin for “affliction with little dragons,” (-iasis from Latin or Greek -asis is added to nouns to form nouns of state or process to form the names of diseases) is a plague so ancient that it has been found in Egyptian mummies and it has been proposed by some to have been the “fiery serpent” described in the Old Testament as torturing the Israelites in the desert.² The name of the parasitic nematode worm which causes Guinea worm disease, *Dracunculus medinensis* (“little dragon from Medina,” dracunculus is the diminutive of draco, the Latin word for dragon, from the Greek word drakon for dragon), derives from the one-time high incidence of the disease in Medina, now Al Madinah Al Munawwarah of Saudi Arabia, and the common name Guinea worm is due to a similar past high incidence along the Guinea coast of West Africa.³ The disease is no longer endemic in either location. It was suggested by Velschius in 1674 that the Rod of Asclepius, of a snake entwined around a stick, a common medical symbol, depicts a recently extracted Guinea worm.³

Dracunculiasis is transmitted to humans through drinking water contaminated with microscopic copepods (water fleas) that are infected with larvae of the worm. About a year after a person has become infected, adult female worms emerge from the skin (usually 1 to 3 emerge simultaneously, but as many as 40 have been documented to emerge from a given person in a season).² If the emerging worms make contact with water, they expel larvae into the water, the larvae are ingested by copepods, and the cycle begins again.² The emergence of the worms, which can be more than 0.6 m long, is painful and often incapacitates people for 2 to 3 months.² The migration site for the adult worm is usually a lower limb and initially there is an intensely painful blister on the skin.³ When an infected person submerges the wound in water to ease the pain, the blister bursts open and the worm spews her larvae into the water. The worm slowly crawls out of the wound over the course of 3–10 weeks and the wound remains painful during this time, disabling the person.³ As the worm migrates to its final site, some people have allergic reactions including hives, fever, dizziness, nausea, vomiting, and diarrhoea.³ As the worm emerges, the open blister often becomes infected with bacteria resulting in redness and swelling, the formation of

abscesses, and rarely, in severe cases, gangrene, sepsis or tetanus (lockjaw). When the secondary infection is near a joint, typically the ankle, arthritis may occur with joint stiffness and contractures.³

The mainstay of treatment is the careful wrapping of the emerging worm around a small stick to encourage its exit.³ Each day a few more cm of the worm emerge and the stick is wound to maintain a gentle tension.³ With too much tension, the worm can break and die in the wound causing severe pain and swelling at the ulcer site.³ There is no effective anthelmintic agent or vaccine.² Infection can be prevented by filtering drinking water through finely woven cloth, which removes the copepods; by killing copepods and larvae with temephos applied to open ponds; by educating villagers about not entering sources of drinking water while infectious; and by providing clean drinking water from safe sources such as borehole wells or hand-dug wells.² In areas where Guinea worm is endemic, the parasite often predominantly infects women, who tend to do most of the washing and the gathering of water for households. During the planting or harvest season, dracunculiasis has sometimes been reported in more than half the population of a given village.²

Until recently, humans and water fleas were regarded as the only animals the parasite could infect but it is now known that baboons, cats, dogs, frogs, and catfish (*Synodontis*) can also be infected naturally and ferrets have been infected experimentally.⁴ Frogs can act as a paratenic host which is a secondary or subsequent intermediate host for a parasite with a complex life cycle that usually involves two or more species.⁵ The parasite does not undergo developmental stages in the paratenic host but exists in a quiescent or encysted state until it has the opportunity to move to a more definitive host species.⁵ Raw fresh water fish may be paratenic hosts in which the immature parasites ingested in water fleas survive but do not develop.⁶ Rodents can be infected experimentally and could act as a paratenic host for carnivores.⁶

The predatory species of copepods which can ingest the *Dracunculus medinensis* larvae were previously included in the single genus *Cyclops* but this has now been subdivided into the genera *Mesocyclops* (*M. aquatorialis* and *M. kieferi*), *Metacyclops* (*M. margaretae*), and *Thermocyclops* (*T. crassus*, *T. incisus*, *T. inopinus*, and *T. oblongatus*).⁶

Because *Dracunculus* larvae need a period of 12 to 14 days to develop in the *Cyclops* to become infective, dracunculiasis is not normally caught from flowing water sources such as rivers and streams.⁶ Deep wells are rarely involved as few *Cyclops* are found in them, probably because the lack of light at the bottom constrains the population of zooplankton which are the *Cyclops* natural diet. Thus ponds, and sometimes shallow or step wells, are the main sources of the disease and the epidemiology of dracunculiasis is chiefly determined by the use of such sources for drinking water.⁶ Ponds, mostly human made, have been found to be the main source of transmission in Nigeria, Ghana, Burkino Faso, Togo, Uganda, Pakistan, India, and what is now Uzbekistan.⁶

In 1957, Rajasthan was the state in India worst affected with dracunculiasis with over 2 million of the 5 million people at risk being in that state. In Rajasthan, the disease was confined mainly to the north western and southern regions. In western desert region, shallow rain water ponds were a habitat for the Guinea worm vector *Cyclops*. In the southern region, including Dungarpur, Banswara, and Udaipur, which are now part of the scheduled area of Rajasthan, the usual water sources were open wells and rectangular, masonry-lined step-wells.⁶⁻⁸

Guinea worm disease was seen to be a promising candidate for successful eradication because *Cyclops* was not a mobile vector like a mosquito, the carrier state in both the *Cyclops* and human hosts was of limited duration, diagnosis was easy and unambiguous, the geographical distribution was limited and even within this area found only in certain communities of endemicity, it was seasonal in distribution, and cheap and effective measures were available to prevent transmission.⁶ India was the

first country to initiate an eradication campaign, with its national programme starting in 1982. By 1990, India was followed by Pakistan, Ghana, Nigeria, and Cameroon.⁶ In the following 5 years, all other known countries of endemicity also established eradication programs.⁶

The global Dracunculiasis Eradication Program was spearheaded by former USA President Jimmy Carter and the Carter Center. The programme involved grassroots public health initiatives involving thousands of village volunteers.² Following a two-decade campaign against Guinea worm disease, including the provision of safe water with borehole and hand-dug wells, the global incidence fell from an estimated 3.5 million cases in 1986 to 25,217 in 2006.² A slight increase in the reported incidence during 2006 was attributable to improved detection in newly accessible areas of southern Sudan.² The eradication programme reduced the number of countries with endemic dracunculiasis from 20 in 1986 to 9 in 2006 (with 5 of the 9 having reported fewer than 30 cases each).² By 2007, the World Health Organization (WHO) had certified 180 countries as free of Guinea worm disease.²

After a decade with no reported cases, Chad reported 10 indigenous cases in humans in 2010 and Guinea worm infections in domestic dogs were reported for the first time in 2012, mostly from communities along the Chari river in Chad.⁸ During January 2020–June 2021, Chad reported 98% of the world's remaining *Dracunculus medinensis* infections, with 94% of these being in dogs.⁹ Stopping transmission in Chadian dogs was seen to be the biggest challenge in the eradication programme.⁹ Fish were seen to be able to serve as transport hosts and frogs as paratenic hosts. *Dracunculus* larvae were recovered from multiple wild frogs in Chad and the eradication programme gave attention to increased active surveillance, proactive containment by tethering of dogs for the 4 months of peak dracunculiasis incidence, temephos application, cooking fish well, and preventing animals from eating fish entrails by burying them.⁹ In 2020, Mali reported its first case in a human in over 4 years and Ethiopia its first cases in humans in over 2 years.⁹ Some continued endemic transmission has occurred in a few dogs and cats in Mali and baboons in Ethiopia in some limited areas.⁹

In 2020, only 27 human cases were reported with only 6 countries still having the disease: Angola, Chad, Ethiopia, Mali, South Sudan, and importations into Cameroon.⁹ In 2021, 15 human cases were reported: Chad=8, South Sudan=4, Mali=2, and one in Ethiopia.¹⁰ By 2022, 199 countries had been certified to be free of Guinea worm with only 7 still to be certified: Chad, South Sudan, Mali, Ethiopia, Angola, Cameroon, and the Democratic Republic of the Congo.¹⁰ In 2021 there were 885 animal cases: Chad: 855 (dogs=790, domestic cats=65), Cameroon: 10 (all in dogs), Ethiopia: 3 (dogs=2, cat=1, baboons=0 compared to 4 in 2020), and Mali: 17 (dogs=16, cat=1).¹⁰ In January-April 2022, only Chad reported animal cases (n=109) and the only two human cases reported were both in Chad.¹¹

In the design of programmes to combat infectious diseases, field experience has been found to be more reliable than theory or basic biology and successful programmes were often based on remarkably little systematic evidence from the field.⁶ Mistakes were made along the way in the eradication programmes and the importance of learning from experience noted.⁶ Given the transmission cycle of the parasite and the absence of an effective vaccine, a number of *a priori* interventions were considered: (i) provision of a safe water supply; (ii) filtration of one's drinking water to remove *Cyclops*; (iii) searching for patients who are active cases and properly managing them; (iv) ensuring that patients avoid contact with ponds; and (v) killing or removing *Cyclops* in ponds.⁶

Safe water supply: In the early discussion of eradication strategies, the provision of a safe water supply was generally seen to be the intervention of choice.⁶ India's rural water supply programme gave priority to villages of endemicity and, by the time that the national eradication programme was concluded, had provided a supply to every

village of endemicity in the country.⁶ This was an important contribution to India's successful elimination of the disease in 1997.⁶ However, some important limitations were found as to the effectiveness of water supply as a possible intervention.⁶

(a) MAINTENANCE: Water supplies cannot function without maintenance. In the Ivory Coast between 1973 and 1985, 12,500 new boreholes were installed in rural areas at a cost of many millions of dollars and the number of cases fell from 67,123 in 1966 to 1,889 in 1985.⁶ However, in 1991 it was estimated at 12,690 and a survey of hand pumps in three sub-prefectures found more than half of them were out of order.⁶

(b) DIFFICULTY SUPPLYING EVERY VILLAGE AND HAMLET: In the Ivory Coast, villages of endemicity with fewer than 150 inhabitants had case rates 4 times greater than the largest villages because they were excluded from the borehole programme as their populations were considered to be too small to justify the cost of drilling and maintaining a hand pump.⁶

(c) FAILURE TO USE THE HANDPUMP: A functioning safe water supply will still be ineffective if it is not used as may occur if it was not close enough to people's homes. In the dry season in the Sahel region of Burkina Faso, a hand pump may be the only safe water source for miles around and during the rains, when Guinea worm transmission peaks, people are often infected from the many ephemeral ponds within a few hundred yards of their houses. For more than 10% of the population in southern Sanmatenga province, Burkina Faso, the pump is more than 1 km away.⁶

(d) SEASONAL MIGRATION: Much of the population of rural Sahel migrates to a number of small and seasonally occupied hamlets to sow crops or tend those where the rainfall has been plentiful and supplying a borehole at \$10,000 for every such hamlet is not cost-effective.⁶

(e) USE OF UNPROTECTED SOURCES: Water supplies alone cannot eliminate dracunculiasis if they are not used exclusively and infection is often acquired through the casual use of unprotected sources when away from home such as when working in fields.⁶

(f) HIGH COST OF WATER SUPPLIES: The high cost of providing water supplies and maintenance is a limiting factor.⁶

Filtration: Filtration of water through cloth can be effective as the adult cyclopoid is over 1 mm long. However, cloth may not last for more than a year. An effective use of a monofilament nylon cloth has been to fix it over the end of a piece of 10–20 mm diameter plastic pipe, 100–200 mm long, which can be worn around the neck on a string and used as a "straw" filter to drink through from ponds when away from home on journeys or in the fields.⁶

Case management: Surgical extraction of the worm before emergence requires great skill to avoid breaking the worm which is sometimes caught around joints or tendons but avoids the pain and suffering caused by the worm emerging and contains the case by preventing contamination of water sources. When practiced by BL Sharma, from 1985 to 1993, in the Banswara, Dungarpur, and Udaipar districts of Rajasthan, it led to a more rapid decline in Guinea worm cases compared to the other districts.² However, maintaining quality standards is difficult in the rough conditions of the field and it was only performed by two people in India. More practical may be applying an occlusive bandage to the lesion which discourages the patient from immersing the affected part in a pond and transforming a neat bandage into a soggy mass of wet cotton.⁶

Preventing patients' contact with ponds: Although in the early stage of the eradication campaign this was seen to be impractical in a rural context where it might be difficult to detect a case within a month of emergence of the worm, later it became clear that people sometimes respond so well to this message that it can have a significant effect on transmission in the complete absence of filter cloth. For example, in an area of 160 villages in Adior district in Bahr el Ghazat province, Sudan, not enough cloth filters were available so that only 44% of households had

one. Nevertheless, a reduction of 88% in the number of cases was achieved, with only 515 cases reported in the first 10 months of 2000 compared with 4,177 in the same period in 1999.⁶

Killing or removing Cyclops: Killing cyclopid vectors with a cyclopicide played a prominent role in the eradication programmes launched in the 1980s in India, Pakistan, and Cameroon. These programmes successfully achieved elimination but it took many years. In more recent years, temephos, an organophosphate insecticide safe for use in drinking water sources has been used in many countries but it is harder to calculate the dose needed for an irregular pond than a rectangular Indian step well. Treatment of ponds can consume substantial resources, particularly in terms of trained staff.⁶

Case containment: By the middle 1990s, as case numbers began to come down, there was increased enthusiasm to step up the level of intervention and move towards “case containment” with the protection of ponds and the community at large from contamination by infected people. The impact of the case containment strategy has been hard to discern in the national surveillance data.⁶

The intervention that had the greatest impact was health education, initially to promote the use of cloth filters to remove *Cyclops* from drinking water and later to prevent the contamination of ponds by patients. Water supply and vector control have proved more expensive in financial or human resources and have been most effective in specific settings.

Although *dracunculiasis* has been considered to have been an eradicated disease in Rajasthan state, India, since 1997,⁸ and in India since 1999,^{12,13} with the WHO declaration that India had eradicated dracunculiasis being made in 2000,¹⁴ some sporadic cases have continued to be reported. Following the declaration that Rajasthan was dracunculiasis free in 1997, three cases were detected in the Dungarpur district, Rajasthan, in July 1997,^{8,15} in 1999,¹⁵ and in 2006.⁸ In the 1997 case, a Guinea worm began to emerge from a swelling on the left breast of a 28-year-old woman and it was then surgically extracted and several motile first stage larvae were identified as *Dracunculus* larvae. In the 2006 case a gravid Guinea worm began to emerge from a painful shallow foot ulcer, which began as a blister, on the left foot of a 40-year-old man. The worm was later extracted surgically.

In 2012, two cases were reported from Maharashtra, a state in the western peninsular region.¹³ One case was of an 8-year-old girl with a 4 year history of recurrent leg pain and ulceration.¹³ An X-ray showed a long linear chain mail type calcification typical of calcified Guinea worm which was removed surgically and confirmed as being a long linear calcified worm. The second case was of a 25-year-old woman with acute inflammation of an abdominal nodule that had been present for 5–7 years.¹³ The lesion was excised and histopathology showed a calcified worm.

Also in 2012, Sheikh reported that two further cases of dracunculiasis were reported, from villages in Rajasthan, in the newspaper *Rajasthan Patrika* on 6 September 2011¹⁶ and asked if dracunculiasis had really been eradicated. The author commented that *Dracunculus* appeared to have succeeded in maintaining its gene pool despite so many efforts of the humans to eradicate it or rather cause its extinction.¹⁶

Similarly, a case presenting as a diabetic foot ulcer was diagnosed in 2013¹⁷ from Kerala state, on the Malabar Coast in the southwestern coastal region. The patient, a 57-year-old man with diabetes mellitus, presented with a swelling of redness and pain of 7 days duration on the left leg near the ankle just above the lateral malleolus which developed into an abscess, measuring 3×3 cm, with a mild cellulitis around it. Azithromycin, chymotrypsin, and ichthammol glycerin were prescribed. A week later the patient reported by phone the emergence of a whitish, thin worm-like structure from the abscess. It was assumed to be a Guinea worm and he was advised to extract the worm by the age-old method using a stick to roll the worm on and to

pull it out slowly. The extracted worm, 25 cm long and 1.5 mm wide was identified as a Guinea worm.

In 2021, two cases were reported from the states of Punjab, in northwestern India, and Maharashtra, in the western peninsular region. One involved a dead worm, identified by high-frequency ultrasonography with 7–15 MHz multifrequency probes with colour and spectral Doppler, near the posteromedial aspect of the right ankle presenting with inflammation of the skin and subcutaneous tissue. In the other, a live worm presented with a erythematous, oedematous, inflamed, non-indurated periorbital swelling with a rope-like structure near the lateral canthus. The worm was extracted intact surgically with forceps and identified as *Dracunculus medinensis*.¹²

Although the provision of safe drinking water for the tribal people in the scheduled area of Rajasthan, India, has played an important part in the almost complete eradication of dracunculiasis, it has come at the cost of introducing high fluoride groundwater to the population resulting in many being affected by chronic fluorosis. A similar situation has occurred in other areas where the provision of water from a borehole has resulted in the occurrence of chronic fluorosis, including dental, skeletal, and non-skeletal fluorosis. For example, in the village of Ban Sankayom, population 1,043, 28 km south of Chiang Mai, northern Thailand, a deep well borehole was dug at the school but the resulting water had an excessively high fluoride content of 6.7 mg/L.¹⁸ To make the water safe for use, seven bone char defluoridators, produced in the village by a local water consultant, were installed.¹⁹ There were four public defluoridators, three at the school and one at the temple which used groundwater from the same borehole, and three were installed in private homes.¹⁸

Both dracunculiasis and chronic fluorosis can cause considerable morbidity but it could be argued that chronic fluorosis can be more debilitating and produce a greater health burden than dracunculiasis. Dracunculiasis is rarely fatal, with Indian studies suggesting a case fatality rate of 0.1% or less.⁶ The permanent disablement rate is less than 1%.⁶ Temporary disability occurs during and after the period during which the worm emerges.⁶ Studies from Nigeria found 58–76% of patients were unable to leave their beds for approximately a month during and after the emergence of the worm.⁶ More severe and protracted disability occurs with secondary infection of the lesion which occurs in approximately 50% of cases.⁶ Exposure to high fluoride drinking water during pregnancy and early childhood may produce a considerable burden with impairment of neurodevelopment and a shift in the mean IQ for a population.¹⁹ Dental fluorosis is a permanent disability. Skeletal fluorosis may cause ongoing pain and disability which will not subside while fluoride exposure continues. Skeletal deformities and severe complications such as paraplegia are permanent.

There is a strong argument for completing the admirable task of eradicating dracunculiasis from the tribal people in the scheduled area of Rajasthan, India. The mission will not be accomplished satisfactorily until the problems associated with high fluoride drinking water from boreholes is also addressed with an appropriate strategy. Considerable experience from India is available on how to prevent chronic fluorosis through a variety of measures including, as appropriate, (i) identifying safe water sources, (ii) dietary counselling, (iii) dietary editing, and (iv) defluoridation of high fluoride water when other options are not feasible.¹⁹ The plight of the tribal people in the scheduled area of Rajasthan, India, merits further attention.

Bruce Spittle, Editor-in-Chief, *Fluoride*, Dunedin, New Zealand

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