First Incidence of Human Gongylonematosis in Hungary

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Gongylonematosis is a rare human helminthosis; only a total of 32 cases have been reported so far, i.e., 7 cases from the Soviet Union, 4 from Bulgaria, 3 from Italy, one both from Yugoslavia and Austria, 10 from the United States, 3 from China, 2 from Morocco and 1 from Ceylon.

Case history

Miss T.K., aged 20, brought on the 24th February, 1972, an about 5 cm long roundworm to the laboratory of the SANEPID Station. She told to have extracted the parasite from her upper lip the night before. About three months earlier, she had similarly removed a "thing" from her mouth, but had thrown it away. Complaints: for about the last three months she had occasionally felt a circular movement and prick in her mouth and had simultaneously felt a slight projection with her tongue which, according to her own words, "migrated". She often vomited after meals, her abdomen puffed up, she felt nervous and irritable, slept little and restlessly, had terrible dreams and felt dizzy at times.

Except for the Giardia lamblia parasitic infection could not been demonstrated by the examination of stool samples.
The worm recovered from the patient's mouth was identified as *Gonçylonema pulchrum* in the Helminthological Research Laboratory, University of Veterinary Medicine.

The helminth, a female, was 54 mm long and its maximum width was 0.25 mm at the vulva. The papilla-like cuticular structures, 30-50 μ in diameter, localized within 1.3 mm distance from the anterior end /Fig. 1/. The vulvar opening was surrounded by a conspicuous projection /Fig. 2/. The oviduct was full of eggs, measuring 52.5 by 25 μ. Mature eggs, 60 x 32.5 μ in size, each comprised a fully developed larva and bore a distinct operculum on both poles /Fig. 3/.

One week after the first consultation, another helminth appeared in the patient's mouth, in the upper right oral fold; its zigzag shape and white colour could easily be distinguished against the slightly hyperaemic surroundings.

The parasite, a not fully mature female, 28 mm long by 0.2 mm wide, was removed in the Stomatology Clinic, University Medical School of Debrecen, by incision of the mucous membrane after a local infiltration anaesthesia with Lidocain. Again one week later, the patient felt the appearance of new worms in her mouth; in this instance they localized sublingually, again in a zigzag pattern.

The main complaints of the patient were on the one hand gastrointestinal symptoms /frequent vomiting, bloating/, on the other nervous signs /excitation, irritability, insomnia/. Since the parasites reappeared regularly, their surgical removal, as suggested in the literature, was only a symptomatic therapy, not necessarily resulting in sanitation. Attempt was therefore made to cure the patient by drug therapy, in order to destroy migrating larvae and adult worms.

Three 150 mg doses of levamisole + /2,3,5,6-tetrahydro-6-phenyl- Decaris tablet, Chemical Works of Gedeon Richter Ltd., Budapest
imidazo-/2,1-b/ thiazole hydrochloricum/ were given at one-week intervals.

Laboratory tests prior to the treatment showed no abnormality. Blood picture: erythrocyte count 3.7 million, leukocyte count 5800; haemoglobin 13 g%; ESR 2 mm/h. Differential count: segmented immature 2 %, segmented mature 71 %, lymphocytes 25 %, monocytes 3 %, eosinophiles 0, basophiles 0. Serum total protein 6.4 % /albumin 58 %, alpha₁ globulin 2 %, alpha₂ globulin 4 %, beta-globulin 13 %, gamma-globulin 17 %. For the 6 months elapsed since the therapy, the patient did not notice new worms and both vomiting and bloating ameliorated considerably.

Discussion

The causative agent of gongylonematosis had first been described by LEIDY in 1850 /GEFTER, 1965/. The roundworm Gongylonema pulchrum Molin, 1857 /synonyms: Filaria labialis, Pane, 1864; Spiroptera scutata, Müller, 1969; G. scutatum, Leuckart; G. ran-somi, Chapin, 1922; G. hominia, Stiles, 1921/, is a nonspecific parasite of a variety of hosts. It parasitizes chiefly domesticated or wild, small and large ruminants and swine, less often ungulates, monkeys, bears, hedgehogs and rodents, inhabiting their gastric and oesophageal mucosa and tongue /swine/. Textbooks of veterinary parasitology /BOCH and SUPPERER, 1971; KOT-LÁN, 1960; SOULSBY, 1965/ have usually referred to G. pulchrum infection simply as a helminthosis, without describing its clinical signs and gross lesions. Apart from G. pulchrum, other Gongylonema species have frequently been encountered in the first stomach of ruminants and in the proventriculus of poultry as well. A single highly pathogenic species, G. neoplasticum, is known to cause a neoplasm-like proliferation in rat stomach.

In the reported cases of human gongylonematosis, G. pulchrum worms arranging in a spiral pattern, were found in the area of
the lips, gingiva, hard and soft palate, mouth corners and tonsils. The oral localization, which differs from the sites of preference in animals can probably be explained by the implication that the best final hosts are animals, above all ruminants, rather than man /FAUST, 1957/.

According to the literary data, the *G. pulchrum* male is 30-62 mm long and 0.3 mm wide at the most, the female is 80-150 by 0.5 mm, but parasites from man and swine are much smaller /FAUST, 1957/. At the anterior end, the cuticle carries characteristic thickenings, arranged in 2-3 longitudinal rows. The recurved tail end of the male bears a total of 5-7 pre- and postanal papillae and a pair of unequal spicules. The vulva of the female localizes posteriorly, 2-7 mm distant from the tail end. The eggs are elliptic, 57-59 by 30-34 μ in size, smooth, transparent, with a small operculum on both ends; at oviposition, they contain first-stage larvae which require an intermediate host for further development.

About 50 species of coprophagous insects /sarcophagous beetles, cockroaches, etc./, chiefly Coleoptera and Orthoptera, belonging to several genera /Aphodius, Blaps, Ontophagus, Blattella, etc./ of 3 families are known as possible intermediate hosts of *G. pulchrum*. The larvae undergo two molts in the intermediate host, developing to encapsulated, infective third-stages in about one month /ALICATA, 1935/. The final host, whether a human or an animal, becomes infected by ingestion of the intermediate host containing third-stage larva, or its parts. The larvae hatch in the stomach and invade its wall, then migrate into the oesophagus or oral cavity and establish themselves in the superficial layers of the mucosa until they reach sexual maturity in about 2 months. Within the mucous membrane, these nematodes are bound to an almost uninterrupted migration, on the one hand for search of nutriments, on the other to avoid extrusion by the continuous epithelial regeneration. The life cycle of the parasite in the final host was studied by ALICATA /1935/ on experimentally infected guinea-pigs.
As it has already been mentioned, the gongylonemas which invade humans establish themselves above all in different areas of the oral cavity, burrowed in the oral mucosa and submucosa, in which they can move freely. After maturation, one end of the worm is extended into the oral cavity, so that the patients are able to remove it themselves. Most patients consulted the doctor for the inconvenience caused by the movements of the parasite, or after the extraction of the mature worm from the mouth.

Most reported cases of human gongylonematosis were oral and occurred among children or young adults living in villages. Apart from the symptoms outlined in the foregoing, pruritus, toothache and salivation are frequent accompanying signs /GEFTER, 1965/; in a single case, even pharyngitis and stomatitis occurred /FAUST, 1955/, and WARD /1916; cit. GEFTER, 1965/ observed additionally a periodic temperature elevation. In most cases, the parasites reappeared several times, causing nervousness, insomnia and excitation /GAUD and CHABAUD, 1951, cit. GEFTER, 1965/.

As it can be seen from the literary data, nearly all cases of human infection took place accidentally through the ingestion of food or water /shallow brook water/ contaminated by cockroaches or other intermediate hosts. Incidence has, however, also been reported in an entirely cockroach-free environment/ENGELH-STEIN, 1965/. In the overwhelming majority of the cases, the infected persons derived their drinking water supply from cisterns which might have been contaminated either with infected intermediate hosts, or with third-stage larvae spontaneously emerged from such carriers. In the case reported here, the source of infection could not be precisely identified. The patient is a town inhabitant, who has lived in a hygienic environment free of cockroaches.

The prognosis of gongylonematosis is good; unlike the G. pulchrum infection of rats, there is no evidence of malignity in humans.
Prevention of the infection may be achieved by destruction of cockroaches and other insect vectors living in the environment of man and by prohibition of drinking from brooks, springs and cisterns, because the infective larvae remain viable for a long time after death and disintegration of the insects (CHEBOTAREV et al., 1961, cit.: GEFTER et al., 1965) and may spontaneously escape from the body of the intermediate host in the water.

Summary

A case of human gongylonematosis, a parasitic infection not previously observed in Hungary, is reported. Three specimens of Gongylonema pulchrum, a frequent helminth parasite of ruminants developing through intermediate hosts (coprophagous beetles, cockroaches, etc.), were isolated from the oral mucous membrane (upper lip, viz. upper right oral fold) of a 20-year-old female patient within one month.

The clinical picture was dominated by gastro-intestinal disorders and nervous complaints.

Since the surgical removal of the parasites from the oral mucosa did not prevent their reappearance, the patient was treated with levamisole (Decaris, Chemical Works of Gedeon Richter Ltd. Budapest), of which 150 mg doses were given on 3 occasions at one-week intervals.

Explanation of the source of infection is only hypothetical.

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Humán gongylonematosis első esete Magyarországon

A szerzők Magyarországon ez ideig nem észlelt humán gongylonematosis első esetét írják le. A közúti gázda (coprophag rovarok, svábbugyak, csótányok) utján fejlődő és a kérődző állatokban gyakori fonálféregnek — Gongylonema pulchrum — 3 példányát izo-
lálták néhány hónap alatt egy 20 éves nőbeteg szajnyálkahártyá- jából /felső ajkából, illetve az áthajlási redőből/.

A betegnél a gyomor-bélpanaszok és az idegrendszeri zavarok u- ralták a klinikai képet.

A férgek folyamatos megjelenése miatt a beteg levamisolt /Deca- ris, Kőbányai Gyógyszerárugyár/ kapott, 3x150 mg pro dosi, 1-1 hetes intervallummal.

A szerzőknek a leírt féregfertőzés forrására vonatkozóan csak feltételezésük van.

References

Plate I.

Fig. 1. Anterior, 0.5 mm long part of body of a *Gongylonema pulchrum* /female/

Fig. 2. Posterior, 4 mm long part of worm body

V = vulvar opening
A = anus

Fig. 3. Eggs of *Gongylonema pulchrum*

→ mature eggs
⇒ immature eggs collected from the uterus

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