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A Rare Case report of Coinfection with Hymenolepis nana and Giardia lamblia in a patient of Pachydermoperiostosis/acromegaly from India

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Abstract

Pachydermoperiostosis is a rare genetic disorder manifesting in early adolescence. Here, we report a rare case of mixed infection of Hymenolepis nana and Giardia lamblia in a teenage acromegalic patient. The patient was on continuous oral steroids leading to immunosupression. The patient was given empirical treatment with a single dose of Praziquantel 25mg/kg and albendazole (400 mg)for 5 days. Complete remission of the parasitic infestation was seen in the patient in about two weeks. This case adds a new perspective to the incidence of parasitic infection and immunosuppressive clinical conditions.

Keywords: Pachydermoperiostosis- Hymenolepis nana- Giardia – Immunosupression-Steroids.

Introduction

Pachydermoperiostosis (PDP) (Touraine–Solente Gole-syndrome) is a rare disorder characterized by clubbing (acropachy) of the fingers and toes thickening of the skin (pachyderma), usually of the face excessive sweating (hyperhidrosis); and new bone formation associated with joint pain [1] . The diagnosis should only be made when at least two of the following are present: positive family history, clubbing, hypertrophic skin changes, and bone pain/radiographic changes. The estimated prevalence is of approximately 0.16%. It usually manifests in adolescence, occurring almost exclusively in males, with a male to female ratio of 7:1.[2]

Hymenolepiasis is the most common intestinal tapeworm infection of humans caused by worm of family cestoda, genus hymenolepis and species nana .[3] H. nana is commonly known as the dwarf tapeworm. Hymenolepis nana has a worldwide distribution with estimates of 50–75 million carriers .[4] Hymenolepis nana infection is observed in households with poor hygiene and overcrowding, and can have a prevalence of 5–25% in

children[5] .Worldwide prevalence ranges from less than 1% in the United States to about 9% in Argentina, with an average worldwide prevalence of 4%.

When eggs are ingested (in contaminated food or water or from hands contaminated with feces), the oncospheres contained in the eggs are released. The oncospheres (hexacanth larvae) penetrate the intestinal villus and develop into cysticercoid larvae. Upon rupture of the villus, the cysticercoids return to the intestinal lumen, evaginate their scolices , attach to the intestinal mucosa and develop into adults that reside in the ileal portion of the small intestine producing gravid proglottids . Eggs are passed in the stool when released from proglottids through its genital atrium or when proglottids disintegrate in the small intestine. The life span of adult worms is 4 to 6 weeks, but internal autoinfection allows the infection to persist for years**. [6]**

Giardia lamblia is a common intestinal protozoan with a higher prevalence rate in tropical countries as compared to the Western world **.[7]** WHO estimates the worldwide prevalence is 280 million cases. This protozoa is frequently found in diarrheal disease throughout the world.[8]The prevalence of giardiasis is 20 to 30% in developing countries. It is a flagellated pear shaped parasite which has two life forms, the active trophozoite stage and the dormant cyst stage which is the infective stage. It is one of the most common causes of waterborne disease outbreaks associated with drinking water. [9,10]Giardiasis is a noninvasive infection that adheres to the duodenal epithelium and in children it is associated with diarrhea, malabsorption and growth retardation**. [11,12]**

Here we describe the first Case report of mixed coinfection of H. nana with Giardia in a patient suffering from Acromegaly with Pachydermoperiostosis.

Case Report

A teenage male presented to our outpatient department with chief complaints of pain and swelling in both hands and feet for 2 years. The pain was insidious in onset, throbbing in nature and not relieved by over-the-counter medications. The patient also complained of profuse sweating, progressive enlargement of hands and feet, and gradual coarsening of facial features. The patient also complained of poor apetite, diarrhea, abdominal pain, dehydration and fetid flatulence since last 1month.

On enquiry the patient revealed that his mother who was a shopkeeper usually prepared food early in the morning and left for work only to return at night. This food was to be consumed at breakfast, lunch, and dinner without being reheated in between. The mother did not wash her hands regularly prior to meal preparation and basic hand hygiene was also not followed by the family members before having their meals. Food was usually kept in the kitchen without any covers and so, could be easily contaminated with flies and insects.

His family history was significant for consanguinity – his grandparents had a consanguineous relationship. There was otherwise no family history of a similar illness, and this was the first time the patient sought medical attention for this issue. There was no history of fever, palpitations, heat intolerance, or tremors. On examination, patient had Clubbing of all his fingers and toes, pronounced folds in the area of forehead, between the eyes, in the nasolabial grooves and on the chin. Patient had symmetrical enlargement of bilateral forearm and legs.

Routine blood investigations, ESR, CRP, Thyroid function tests, Liver and Kidney function tests were normal. Levels of Growth hormone was elevated to almost five times the normal limit and Oral glucose tolerance test showed no suppression of Growth hormone.

Direct stool examination was done on three sequential fresh stool specimens using a simple wet mount with saline and iodine mount. This examination revealed eggs of Hymenolepis nana **(Fig.1, 2)** and cysts of Giardia **(Fig 3)**. On an average, there were around one egg of H.nana or two cysts of Giardia per high-power field (40X) indicating heavy infestation. Stool for occult blood was negative. Kinyoun and Trichrome staining for opportunistic pathogens was negative. Additionally,ELISA for Giardia lamblia antigen (Nova Tec,Immunodiagnostica GMBH,Germany)was also positive on the stool sample.This kit is intended for the qualitative determination of Giardia lamblia antigens in faeces.).

Xray skull lateral view showed thickening of inner table of calvarium ,enlarged frontal sinuses with frontal bossing and prognathism**.(Fig.4)** Radiograph of bilateral ankle showed osteoarthritis of ankle joint with irregular subperiosteal bone formation and cortical thickening of distal tibia, fibula, calcaneum and talus. Radiograph of bilateral wrist joints showed excessively long digits with soft tissue thickening of tufts giving spade like appearance (Acromegalic hands **)(Fig .5)**

The patient was advised a single dose of Praziquantel 25mg/kg and albendazole (400 mg)for 5 days . A repeat direct stool examination after two days of treatment revealed no eggs of H.nana but had cysts of Giardia.Subsequent stool examination revealed no cysts of Giardia after 5 days of treatment and complete remission was seen in the patient.

The patient was also advised selective COX-2 inhibitors (Eltrocoxib 60mg PO, OD), steroids (Prednisolone 5 mg PO, OD), oral retinoids (20 mg morning and 10 mg HS), and retinoid ointment for 2 weeks. The patient subsequently returned for follow-up after 15 days and his joint pain and swelling had improved markedly with treatment. He was asked to continue the same treatment with outpatient follow-up scheduled for 1 and 6 months later, respectively. On 6 th month follow-up ,The patient had benefitted from the medications and had no relapses or complications from the condition or the medications.

Discussion

Our case report emphasizes the possibility of intestinal parasites in immunocompromised humans on oral corticosteroids. H. nana is the most commonly reported cestode of humans, infecting 175 million people worldwide [13]**,** but few cases have been described involving human immunodeficiency virus–infected patients. [14]Literature shows that bizarre disseminated parasitic infection due to Hymenolepis nana may occur in patients with immunosuppression **.**[15] Experimental animal studies have further substantiated the role of immunosuppression caused by T-cell deprivation or by steroid treatment in Hymenolepis nana infection **.**[16] Studies in immunocompromised patients imply that antibody-mediated acquired immune responses and a minimal T cell availability are of major importance for parasite clearance. These mechanisms likely play in concert with natural resistance mechanisms that are present in the intestinal mucosa. In humans no sterile immunity is acquired after infection. Epidemiological studies further suggest passive protection from symptomatic giardiasis in breastfed children. However, there is a fundamental lack of knowledge about the underlying immunological mechanisms of human giardiasis.[17]

The patient in our case is also on regular steroids and will continue on immunosuppression for a long time. Presence of co-infection with H. nana and Giardia in this scenario may be attributed to immunosuppression as well as unhygienic cooking practices and unsafe drinking water.

Giardia is one of the most common causes of waterborne disease outbreaks associated with unsafe drinking water and Hymenolepis nana infection is observed in households with poor hygiene and overcrowding.

Conclusion

Pubmed literature search does not show any such case of mixed infection with intestinal parasites in Acromegaly patients on steroids. Ours is the first case report from North India of a young male with acromegaly and Pachydermoperiostosis showing co-infection with H.nana and Giardia. Innate immunity plays a dominant role in parasite clearance in the humans. Hence, Immunosupression does not always warrant co-infection with intestinal parasites but certainly is seen in such cases. This finding needs to be studied further with more samples and case reports from Acromegaly patients.

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Legend Figure

Figure 1: Unstained stool wet mount showing H.nana egg with hooklets and polar filaments.

Figure 2: Iodine wet mount of stool showing H.nana egg with polar filaments and hooklets.

Figure 3 : Unstained wet mount of stool showing Cysts of Giardia lamblia.

Figure 4 : Xray skull lateral view showed thickening of inner table of calvarium ,enlarged frontal sinuses with frontal bossing and prognathism.

Figure 5: Radiograph of bilateral wrist joints showed excessively long digits with soft tissue thickening of tufts giving spade like appearance (Acromegalic hands).