

Chronic sialoadenitis caused by *Enterobius vermicularis*: case report

Raro caso di scialoadenite cronica da *Enterobius vermicularis*

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Key words

Salivary glands • Chronic sialoadenitis • *Enterobius vermicularis*

Parole chiave

Patologia ghiandole salivari • Scialoadenite cronica • *Enterobius vermicularis*

Summary

Enterobius vermicularis infection, an oro-faecal transmitted parasitosis, is a frequent finding in infant communities. *Enterobius vermicularis* is located predominantly in the caecum, appendix, and proximal areas of the ileum and colon, even if reports of some rare extra-intestinal cases have appeared in the literature. The case is reported here of a 62-year-old male presenting a mass in the right submandibular triangle. Histological examination, following removal of submandibular gland, revealed a granulomatous sialoadenitis due to *Enterobius vermicularis*. A review of the international literature confirms that this is a very rare site and it would appear to be the first report concerning enterobiasis in the salivary glands. Authors advance a hypothesis concerning a possible pathogenic mechanism.

Riassunto

L'enterobiasi è una parasitosi a trasmissione orofecale, molto frequente nelle comunità infantili. La localizzazione specifica dell'*Enterobius vermicularis* è rappresentata dall'intestino cieco, dall'appendice e dalle zone prossimali dell'ileo e del colon, tuttavia la letteratura cita forme rare extraintestinali. Gli Autori riportano il caso di un paziente, maschio di anni 62, affetto da tumefazione a carico della loggia sottomandibolare destra. L'esame istologico, dopo exeresi della ghiandola sottomandibolare, ha evidenziato una scialoadenite granulomatosa da *Enterobius vermicularis*. La revisione della letteratura internazionale conferma la rarità della localizzazione e consente agli Autori di affermare che la loro è la prima segnalazione di enterobiasi a carico delle ghiandole salivari. Gli Autori concludono spiegando il probabile meccanismo patogenetico.

Introduction

Enterobiasis, a parasitosis typical of cosmopolitan communities, particularly those with a high concentration of infants, is found predominantly in the lower social classes with a poor socio-economic and hygiene level¹.

Enterobius vermicularis, in cycles, re-infects not only the host, but also those individuals with whom they live in close contact. The nematode has a life cycle of 37-53 days.

Infection is via the oro-faecal route, occurring in children, due primarily to the hands carrying the eggs from the perianal area to the mouth, and, in adults, due to eating infected vegetables (contamination of the soil with human faeces). Furthermore, the eggs, on account their peculiar adhesive properties of the shell, adhere to underwear and are carried by dust, in the various living areas².

Enterobius vermicularis attaches itself with the lips to the caecal mucosa, to the appendix and proximal areas of the ileum and colon.

The females travel towards the anal opening where, in contact with air, they deposit their eggs.

It appeared worthwhile describing the present case of granulomatous sialoadenitis due to *Enterobius vermicularis* inasmuch as a review of the literature failed to detect any report of the infection localised in the salivary glands.

Case report

RC: a 62-year-old male, came to our attention in October 2001.

For 20 years, the patient had been aware of a mass, about the size of a chick pea, in the right submandibular area. Over the last 3 months, the mass had gradually increased in size. The patient did not feel any pain and did not present any other important symptoms.

At physical examination, a mass, covered with intact skin, with clear margins, of hard parenchymous consistency, mobile in the upper layers and not painful, was found to occupy the right submandibular area.

Routine examinations, in preparation for surgery, were all within normal limits.

In the right submandibular gland, US revealed the



Fig. 1. US: presence of hypoechoic mass, with clear margins, localised in right submandibular gland.

presence of a nodular hypoechoic formation with a smooth outline (Fig. 1).

The patient was submitted to surgery, under general anaesthesia, for removal of the right submandibular gland.

Examination of the tissue removed at surgery revealed a nodular formation, approximately 3 cm in diameter, in a sub-capsulated position, clearly surrounded with a yellowish-white surface, with a rough lobulated appearance and focal cyst-like areas.

Histological examination: chronic flare-up of granulomatous sialoadenitis due to *Enterobius vermicularis* presenting reactive lymphoreticular hyperplasia of the lymph node with histiocytosis of the sinus (Figs. 2-4).

The post-operative period was uneventful and the patient was discharged on the 3rd day.

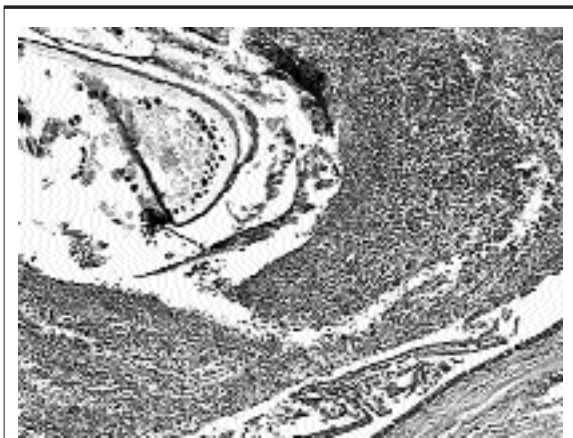


Fig. 2. Excretory duct, filled with *Enterobius vermicularis*, site of marked reactive lymphomonocyte inflammation. (Haematoxylin-eosin, 25X).

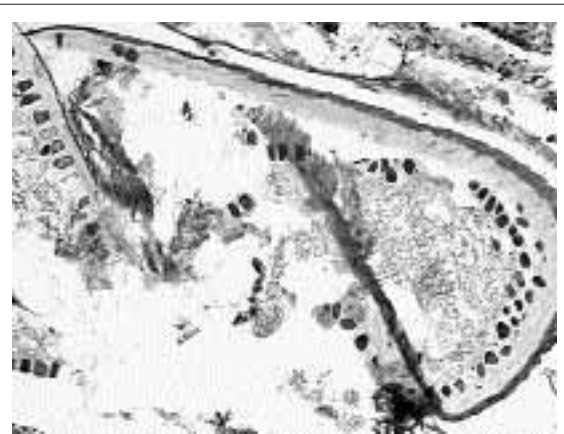


Fig. 3. Detail of *Enterobius vermicularis* located outside the cuticula and muscle (contactile apparatus) and within the uterus containing numerous eggs (pregnant female). (Haematoxylin-eosin, 50X).

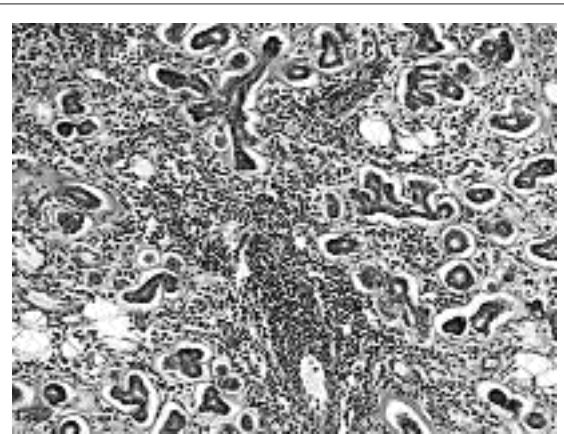


Fig. 4. Salivary gland: site of chronic marked interstitial inflammation damaging epithelium. (Haematoxylin-eosin, 25X).

Given the results of the histological examination, scotch test and culture of faeces were carried out. Results were negative.

Discussion

Enterobius vermicularis is usually localised in the caecum, appendix and proximal areas of the ileum and colon. A review of the literature shows that the most frequent extra-intestinal localisation are the fallopian tubes and ovary (10-20%), due to the close proximity of the female reproductive organs to the

perianal area. Female *Enterobius vermicularis* are, in fact, able to reach the uterus, inducing salpingitis³⁻⁵. *Enterobius vermicularis* has rarely been reported in the peritoneum, liver^{6,7}, lung⁸ or skin⁹.

The mechanism responsible for the presence of the nematode, in these areas, is unknown. Albeit, the nematode is unable to directly invade healthy tissue.

In most cases, ectopic localisation is associated with a concomitant infestation of the bowel.

Localisation in the submandibular gland would appear to be quite unique, inasmuch as no other such cases have been reported in the literature.

From a clinical viewpoint, there are no signs which might be considered useful for diagnosis: the patient merely presented an increase in volume of the submandibular gland resulting from chronic sialoadenitis.

Once the results of the histological examination were obtained, and following more detailed and precise questioning concerning past clinical history, the patient remembered that 20 years earlier, the daughter had had an Oxyuris infection, caught at school.

This finding is, in our opinion, purely casual, since

the parasite is not found in the specific habitat for its survival and, moreover, the time elapsing between infestation and the clinical manifestation is extremely lengthy.

Therefore, the contact had, in our opinion, occurred recently, either due to intake of contaminated foods, or carried by the fingers. In actual fact, carrying eggs to the mouth with the fingers is one of the main mechanisms responsible for infestation; the fingers contaminate the food and objects to which the eggs adhere, due to the adhesive properties of the shell. Furthermore, the eggs resist for about 2 weeks in a humid ambient, at a mild temperature.

Since the saliva in the Warthon duct has an anti-gravitational flow, the fact that the worm travels upwards can only be explained by the existence of a previous chronic sialoadenitis. The chronic inflammation, at gland level, would have determined the dilation of the duct with a concomitant stagnant pool of saliva. This condition would, in our opinion, explain the pathogenetic mechanism responsible for the travelling upwards of the *Enterobius vermicularis* into the Warthon duct, as far as the parenchyma of the gland.

References

- Habbari K, Tifnouti A, Bitton G, Mandil A. *Intestinal parasitosis and environmental pollution: 1343 pediatric cases in Beni-Mellal, Morocco*. Tunis Med 2000;78:109-14.
- Chandler AC, Read CP. *Introduction to parasitology*. New York: John Wiley and Sons; 1961. p. 460-1.
- McMahon JN, Connolly CE, Long SV, Meehan FP. *Enterobius granulomas of the uterus, ovary and pelvic peritoneum. Two case reports*. Br J Obstet Gynaecol 1984;91:289-90.
- Ok UZ, Ertan P, Limoncu E, Ece A, Ozbakkaloglu B. *Relationship between pinworm and urinary tract infections in young girls*. APMIS 1999;107:474-6.
- Vazquez Piloto A, Cruz Robaina JC, Nunez Fernandez F, Sanchez Diaz JM. *Bilateral tubo-ovarian abscess caused by Enterobius vermicularis. Presentation of a case*. Rev Cubana Med Trop 1994;46:65-7.
- Little MD, Cuello CJ, D'Alessandro A. *Granuloma of the liver due to Enterobius vermicularis. Report of a case*. Am J Trop Med Hyg 1973;22:567-9.
- Mondou EN, Gnepp DR. *Hepatic granuloma resulting from Enterobius vermicularis*. Am J Clin Pathol 1989;91:97-100.
- Beaver PC, Kriz JJ, Lau TJ. *Pulmonary nodule caused by Enterobius vermicularis*. Am J Trop Med Hyg 1973;22:711-3.
- Arora VK, Singh N, Chaturvedi S, Bhatia A. *Fine needle aspiration diagnosis of a subcutaneous abscess from Enterobius vermicularis infestation. A case report*. Acta Cytol 1997;41:1845-7.

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