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Case Report

Cheek onchocercoma: A case report

Salwan Yousif Hanna Bede^{a,*}, Bashar Hamid Abdullah^b^a Department of Oral and Maxillofacial Surgery, College of Dentistry/University of Baghdad, Bab-Almoadham, P.O. Box 1417, Baghdad, Iraq^b Department of Oral Diagnosis, College of Dentistry, University of Baghdad, Baghdad, Iraq

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ABSTRACT

Onchocerciasis is an infection with cutaneous, ocular and systemic manifestations caused by the filarial nematode *Onchocerca volvulus*, which is transmitted by the bite of various species of the anthropophilic blood-sucking *Simulium* vectors (black flies). Onchocerciasis is endemic to the savannahs and rainforests of subequatorial Africa and in some countries of the Arabian Peninsula, notably Yemen and Oman, and in Central America, and the Amazon basin of South America. Onchocercomas, which can be defined as subcutaneous fibrous nodules containing adult worms, are among the variable clinical manifestations of this infestation; they are either superficial or deep and usually located over bony prominences. In this paper we report a case of an onchocercoma of the cheek affecting an Iraqi child that was surgically excised. To the best of our knowledge, this is the first case of onchocerciasis in general and onchocercoma in particular that is reported in Iraq with an unusual site, since most of the reported cases occur over bony prominences, and head nodules are reported to occur mostly in the scalp region.

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1. Introduction

Onchocerciasis is an infection with cutaneous, ocular and systemic manifestations caused by the filarial nematode *Onchocerca volvulus*, which is transmitted by the bite of various species of the anthropophilic blood-sucking *Simulium* vectors (black flies). Onchocerciasis is endemic to the savannahs and rainforests of subequatorial Africa and in some countries of the Arabian Peninsula, notably Yemen and Oman, and in Central America, and the Amazon basin of South America [1–7]. It is estimated that about 17–40 million people are infected worldwide, and that about 300,000 people are blind and about 500,000 people have visual impairment due to this infection [2–6,8–10].

It was first described in Africa in 1893 and in Latin America in 1917, but it was not until 1927 that the *Simulium* species were discovered to be the vectors by Blacklock [4,7,11].

The incubation period between the inoculation of the infective larvae and the first appearance of symptom producing microfilariae

in the skin is usually 9–18 months and the life span of the adult worm is about 15 years [3,12,13].

The clinical manifestations include nodules, also called onchocercomas, and acute and chronic skin lesions. Ocular lesions manifest in patients with heavy microfilariae infection or with nodules on the head which can provide a source of microfilariae that can readily reach the eye and invade all tissues of the eye except the lens and lead to loss of sight (hence the name river blindness) [1,12,14,15]. Oral presentation of filariasis in general is uncommon and few cases have been reported in the buccal mucosa in asymptomatic patients so there is a paucity of information in the dental literature with respect to these infections [16]. To our knowledge, there are no registered cases of onchocerciasis in Iraq. In this paper, we report a case of onchocercoma of the cheek affecting an Iraqi child.

2. Case report

A 4-year-old patient was admitted to the consult clinic of the Oral and Maxillofacial unit at Al-Yarmouk teaching hospital in Baghdad, Iraq. His parents noticed the presence of a mass in his left cheek about 4 months earlier that was enlarging in size; otherwise the child had no other symptoms. Clinical examination revealed a mass of about 2 cm in greatest dimension which was rubbery in consistency, freely movable and not tethered to the overlying skin or the surrounding tissues; the mass was not pulsatile, and the overlying skin was normal in color and texture (Fig. 1).

* Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

* Corresponding author. Tel.: +964 7906210006.

E-mail address: salwan.bede@gmail.com (S.Y.H. Bede).



Fig. 1. A clinical view showing a left cheek nodule [arrow].

An ultrasonography showed a well-defined heterogeneous mass measuring 22 mm × 17 mm in the buccal space with cystic changes and associated with bilateral enlarged cervical lymph nodes. Fine needle aspiration cytology suggested a non-specific inflammatory process with presence of fibrous tissue and occasional epithelial cells; no malignant cells were seen in the examined aspirate; the results were inconclusive and excisional biopsy was recommended.

Under general anesthesia, the mass was excised through an intraoral approach via a horizontal incision in the buccal mucosa below the parotid papilla; the mass was removed along with a margin of fatty tissue of the buccal pad of fat (Fig. 2). The patient



Fig. 2. Intraoperative view showing the excised nodule with a margin of adipose tissue from the buccal pad of fat.

was discharged the next day and no complications were reported. Gross examination of the specimen showed a nodular mass of 20 mm × 20 mm × 15 mm; on sectioning there was a fibrous wall surrounding a cystic cavity containing tissue debris and a convoluted worm like structure of about 0.5 mm in diameter (Fig. 3). The histopathological examination revealed a dense fibrous connective tissue wall with heavy infiltrate of admixed inflammatory cells composed of histiocytes, lymphocytes and eosinophils with focal foreign body giant cell granuloma formation surrounding multiple cross-sections of coiled parasite with corrugated cuticle; the central part of the parasite showed gravid uteri with microfilariae and a thin layer of striated muscle; microfilariae were also seen in the connective tissue wall (Figs. 4 and 5). This histopathological picture is consistent with filarial infestation of *O. volvulus*. Dermatological and ophthalmological examinations were carried out to exclude the presence of other lesions. There were no other lesions and the patient was asymptomatic. The decision was to keep the patient on a regular follow-up schedule.

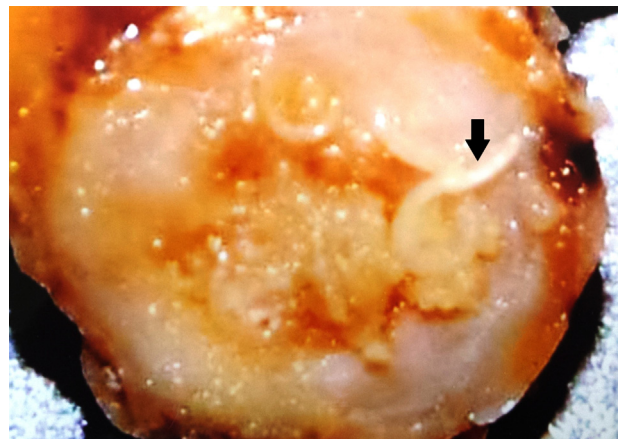


Fig. 3. Sectioned specimen revealing the presence of a worm inside the nodule (arrow).

3. Discussion

The oral helminthic infestations are mainly caused by four roundworm and three tapeworm infections [17]. Different members of the *Simulium* vectors are responsible for the transmission of *O. volvulus*, which occurs near watercourses and fast-running rivers and streams where these vectors breed [1–3,10,12,13]. In our case, the species of *Simulium* vector could not be verified but the parents of the patient confirmed that they live near a watercourse in a suburban area of poor sanitary conditions.

The clinical manifestations are variable and studies have shown that 30% of the cases show dermatitis of which 50% of the patients report itching. The prevalence was shown to be the lowest in individuals ranging between 0 and 10 years of age, while it was the highest in patients 20–30 years of age [4,6].

The clinical picture in our case is consistent with onchocercoma which is defined as a subcutaneous fibrous nodule containing adult worms. It can be detected anywhere in the body, but it is usually located over bony prominences like the iliac crest, ribs or joints [4,6,10]. Onchocercomas of the head usually appear in the scalp [5] and they are generally described as asymptomatic [7] which was also the case in our patient as his parents detected the cheek mass accidentally.

Finding an adult worm in a nodule provides a firm diagnosis of the infection; other methods of diagnosis include observing *O. volvulus* microfilariae in skin snips or in the eye, a diagnostic test known as Mazzotti test, enzyme-linked immunosorbent assay

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