A Rare Manifestation of Cysticercosis Infestation

Sanjeev Gupta[1], Sunita Gupta[2], Amit Mittal[3], Aneet Mahendra[1], Anshu Aggarwal[1], Rohit Batra[4], Nidhi Jindal[1]

[1] Department of Dermatology and STI, MM Institute of Medical Sciences and Research, Mullana Ambala, India.
[2] Department of Medicine, MM Institute of Medical Sciences and Research, Mullana Ambala, India.
[3] Department of Radiodiagnosis, MM Institute of Medical Sciences and Research, Mullana Ambala, India.
[4] Consultant Sir Ganga Ram Hospital, New Delhi, India.

Correspondence mail:
dr. Sanjeev Gupta, H. No. B-2, Near Shiv Mandir, MM Medical College Residential Campus. Mullana Ambala Haryana, India 133207. e-mail: sanjeevguptadr@gmail.com.

ABSTRAK

Ada banyak penyebab urtikaria, berkisar dari infeksi hingga keganasan. Di antara penyebab infeksi, infestasi sistiserkosis (stadium larva cacing pita yang disebut sebagai Taeniasolium) merupakan penyebab yang penting. Laporan kasus ini menyajikan seorang wanita berusia empat puluh empat tahun dengan urtikaria dan pembengkakan wajah. Pembengkakan tersebut kemudian didiagnosis sebagai sistiserkosis dengan pemeriksaan ultrasonografi non-invasif. Urtikaria mereda setelah diberikan pengobatan sistiserkosis. Kami melaporkan kasus ini karena langkanya gejala dan tanda yang tampak.

Kata kunci: *sistiserkosis, urtikaria kronik, Taenia solium, ultrasonografi.*

ABSTRACT

There are many causes of urticaria, which may vary from infections to malignancy. Among the infections, infestations by cysticercosis (larval stage of the tapeworm called Taenia solium) is an important cause. The present report is of forty four years old female who presented with urticaria and swelling on face. The swelling was later diagnosed as cysticercosis by noninvasive ultrasonography. The urticaria subsided after the treatment of cysticercosis. We report this case for rarity of its presentation.

Key words: *cysticercosis, chronic urticaria, Taenia solium, ultrasonography.*

INTRODUCTION

Urticaria is a common health problem that affects 10–30% of the population. Thirty percent of patients with chronic idiopathic urticaria are because of some autoimmune cause. The non-allergic causes of urticaria are auto-immunity, hormonal, stress, exercise and temperature and environmental factors apart from infections[1,2] and infestations. Among infestations, intestinal worms specially cysticercosis is a most important causatives of urticaria. Human cysticercosis is an infection of larval (cysticercus) stage of the tapeworm called *Taenia solium* which can present as subcutaneous/intramuscular swellings.

CASE ILLUSTRATION

A forty four years old female patient presented in dermatology department with history of on and off urticarial lesions of one month duration which were distributed all over the body since one month. On detailed history and examination no relevant cause for urticaria could be established.

She was treated with antihistamines with a partial response to treatment. Her routine investigations were normal. On subsequent follow up, she presented with a swelling over right cheek of approximately fifteen days duration. Patient looked very depressed at that time and was reluctant to give history of swelling as she thought that swelling and skin lesions were not related to each other. On repeated questioning, she told that she had gone to many clinicians for her swelling and was not diagnosed yet. She was suspected of having mumps by otolaryngologist and a surgeon had advised her FNAC (fine needle aspiration cytology) or biopsy. She had also taken lot of antibiotics without any improvement.

Swelling was initially small in size and gradually increased to present 4x5 cm size. She had difficulty in mastication and swallowing due to pain in the swelling. On dermatological examination, urticarial wheals were present over the body. Swelling over the cheek was 4x5 cm in size, firm, mildly tender and not freely mobile. Skin over the swelling was pinchable with no local rise in temp. Provisional differential diagnosis of abscess, hematoma, parotid tumour, muscle hypertrophy was considered. Patient was reinvestigated and all investigations were again within normal limit except for mild eosinophilia.

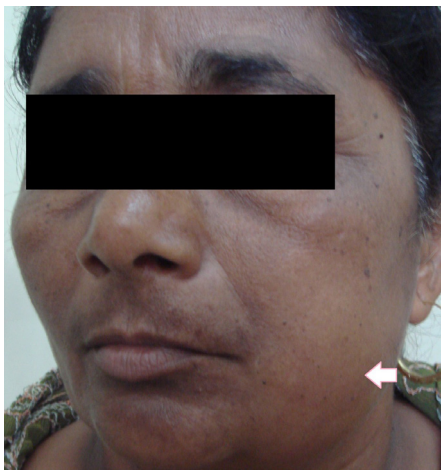

Figure 1. Arrow showing swelling over left cheek

As a routine practice, we subject every patient presenting with cutaneous or subcutaneous swelling for ultrasonography to get some clue to the diagnosis before going any invasive investigation. As the cause of urticaria was not yet established, so, to make the diagnosis of swelling was very important. In vew of that, Ultrasonography was done on Logiq 500 Pro (GE medical systems USA) with high frequency (11MHz) linear transducer which revealed a small cystic lesion with echogenic scolex and surrounding hypo echoic phlegmon in the right masseter muscle. Therefore, on sonography, the diagnosis of intramuscular cysticercosis with surrounding phlegmon was made. On musculoskeletal X-ray examination and magnetic resonance imaging (MRI) of brain, no calcification was seen. The patient was managed conservatively with albendazole 15 mg/kg body weight for one month under initial cover of short tapering course of steroids to reduce inflammatory response. After one month of conservative treatment, there was drastic reduction in the size of the swelling. Interestingly, there was also reduction in the size and number of urticarial lesions during the course

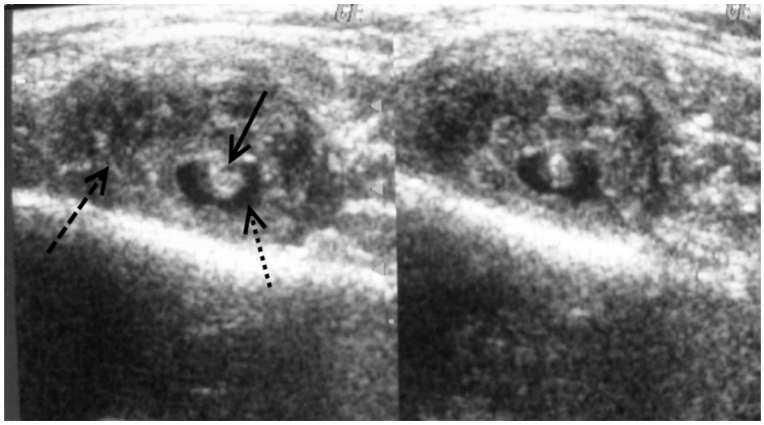

Figure 2. Large inflammatory phlegmon is seen in the masseter muscle surrounding cyst and echogenic nidus of cysticercosis

of treatment. Patient had complete resolution of urticarial lesions after one month of therapy and antihistamines were also withdrawn. On follow up sonography, there was no evidence of active cysticercosis in the muscle. Patient was followed again till three months and there were no complaints.

DISCUSSION

The word urticaria derived from the Latin urtica, nettle (or hives) is a kind of skin rash notable for pale red, raised, itchy bumps. Hives are frequently caused by allergic reactions; however, there are many non-allergic causes. Most of the cases of hives which last for less than six weeks (acute urticaria) are the result of an allergic trigger. Chronic urticaria (hives lasting longer than six weeks) is rarely due to an allergy. The majority of patients with chronic hives have an unknown (idiopathic) cause. Almost 30–40% of patients with chronic idiopathic urticaria will, in fact, have an autoimmune cause. The non-allergic causes of urticaria are auto-immunity, hormonal, stress exercise and temperature and other environmental factors. Infections and infestations can also cause urticaria. Intestinal worms are often said to be associated with urticaria.[3] On extensive literature search, no report was found to have cysticercosis presenting as urticaria. In a study done, to find out the cause of urticaria, a complete blood cell count can be used as a baseline screening investigation in more severe urticaria. Blood eosinophilia should prompt stool examination for parasitic infestations, although this is a rare cause of chronic urticaria in developed countries.[4]

Human cysticercosis is an infection by the larval (cysticercus) stage of the tapeworm *Taenia solium*. Cysticercosis is rare in Europe and North America but not in central and South America, Africa, India and China. Tapeworm infection is common in developing countries where the combination of rural society, crowding and poor sanitation allows greater contact between human and pigs and thus more opportunities for faecal contamination of food and water.[5] Normally humans are the definitive host for *Taenia solium* and other animals may harbour larval forms as intermediate host. The life cycle begins with ingestion of viable larvae in inadequately cooked pork. The cyst wall is destroyed by gastric secretion, releasing scolex that passes into the small intestine where it becomes fixed. The eggs hatch in the small intestine releasing oncospheres that penetrate the bowel mucosa and enter blood stream to reach various tissues.[6] In the tissues these eggs encysts themselves to form cysticercosis cellulosae. The clinical course of cysticercosis depends on number of cysts.[7] When the larvae dies it induces a vigorous granulomatous inflammatory response that produces symptoms, depending on the anatomic location.[8]

The clinical presentation of cysticercosis depends on the number and location of cysticerci in addition to extent of associated inflammatory response or scarring. Cysticercosis often involves central nervous system, eye, subcutaneous tissue, skeletal muscle and heart but the lungs, liver and kidney may be affected. Subcutaneous invasion is the most common form which present in 25 to 65% of patients with cysticercosis. Subcutaneous cysticerci are important in clinical practice due to three reasons. First, they may be mistaken for some other disease presenting with painless swellings like lipoma, neurofibroma, lymphadenopathy or epidermoid cyst. Second, awareness of such lesions leads to early diagnosis and treatment of cysticerci in vital organs. Third, in patients with suspected neurocysticercosis, the presence of subcutaneous cyst provide an important clue to the diagnosis.

Skeletal involvement may cause transient tenderness. In the muscular form, three distinct types of clinical manifestations have been described: the myalgic type; the mass-like, pseudotumour or abscess-like type; and the rare pseudohypertrophic type.

With the advent of high resolution ultrasonography, it can be used widely for diagnosing muscular cysticercosis.[9] There are only a few reports of the ultrasound features of muscular cysticercosis. Sonography shows well defined, elliptical cystic fluid filled lesion with echogenic scolex. Living cysticercosi actively evade immune recognition and do not cause inflammation, However, during the death of larvae, leakage of fluid from the cyst

trigger inflammatory response with formation of inflammatory phlegmon occurs as was also seen in our case. High-resolution ultrasound findings are pathognomonic of cysticercosis and a definitive diagnosis can be made with great confidence. Surgical removal is indicated for localised lesion that causes obvious symptoms. Medical treatment with praziquintal or albendazole have been recommended for neurocysticercosis, subcutaneous and intramuscular cysticercosis. A summary of treatment series has been shown by Garcia et al. Cure rates ranging from 60-85% in usual dosing with most series showed albendazole yielding high cure rates. Albendazole has largely supplanted praziquantel because of slightly greater cure rates, increased efficacy in subarachonoid or ventricular cysts, less cost and increased availability.[10] Preventive measures are important and include the thorough cooking of pork and all vegetables and early detection and complete removal of the worm, including the head. We successfully managed the patient with albendazole and steroids. On extensive literature search, no report was found to have cysticercosis presenting as urticaria. In our case there was complete resolution of urticaria after treatment of cysticercosis. Whether improvement of urticaria was incidental or cysticercosis itself was the cause of urticaria, this question remains unsolved.

CONCLUSION

High-resolution USG, being non-invasive and non-ionizing, plays an important role in establishing the diagnosis in patients with muscular cysticercosis. If lesions with the morphological characteristics described above are encountered on ultra-sound, the diagnosis of cysticercosis can be made with great confidence, and in muscular and subcutaneous cysticercosis no further investigation is required. The advent of newer high resolution USG leads to the greater capability of diagnosing subcutaneous cysticercosis accurately without need of more invasive biopsy or less reliable FNAC.[11] Cysticercosis should be an integral part of the differential diagnosis of all subcutaneous swellings, regardless of the clinical setting.

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