Isolated Leprosy of Bilateral Ear Lobes, A Rare Presentation of Leprosy

Paudel V,[1] Chudal D[2]

[1]Department of Dermatology and Venereology,
National Medical College,
Birgunj, Nepal.

[2]Nepal Police Hospital,
Kathmandu, Nepal.

Corresponding Author
Vikash Paudel,
Department of Dermatology and Venereology,
National Medical College,
Birgunj, Nepal.
E-mail: vikashpaudel@gmail.com

Citation
Paudel V, Chudal D. Isolated Leprosy of Bilateral Ear Lobes, A Rare Presentation of Leprosy. *Kathmandu Univ Med J.* 2021;74(2):268-70.

ABSTRACT

Hansen disease is a chronic infectious disease caused by Mycobacterium leprae. It can present with a variety of clinical manifestations depending on the immune status. Ear lobes are commonly involved in leprosy. However, isolated earlobe involvement of both ear lobes is rare. We present a case of a 25-year-old man who presented with isolated swelling of both pinna, which on slit skin smear turned to be multi-bacillary leprosy. The bilateral pinna swelling started as asymptomatic, multiple skin colored shiny papules which progressively became lobular over a period of a year. Isolated involvement of the pinna in a patient without lesions in other body parts is an unusual initial presentation of leprosy. However, leprosy should be kept as a rare differential diagnosis of isolated lesions on the ear in patients not responding to conventional treatment.

KEY WORDS

Hansen's disease, Leprosy, Mycobacterium leprae

INTRODUCTION

Leprosy (Hansen's disease) is a chronic infectious disease caused by *Mycobacterium leprae* which is still endemic in many developing countries, with the majority of cases detected in countries like India, Nepal, Sri-Lanka and Brazil and some of the African countries.[1,2] The cardinal signs of leprosy are hypopigmented or erythematous, hypoaesthetic or anesthetic skin lesions, and impaired nerve function with or without thickened peripheral nerves. It primarily affects the cooler areas of the body like peripheral nerves, skin, external ears, and nasal mucosa, but the clinical spectrum is governed by the immune status of the host, thus varying in the tubercular and the lepromatous pole.[3] Our patient had isolated bilateral multiple lesions over both pinna for which he had multiple consultations with dermatologists and otorhinolaryngology before visiting us. The final diagnosis was made which was supported by slit skin smear and thus treatment was started. Isolated ear lobe involvement in leprosy is a rare occurrence, hence we are reporting the case.

CASE REPORT

A 25-year-old male patient residing in Parsa, central Nepal, presented in our clinic with a complaint of gradually progressive painless swelling of both earlobes of six months duration. He had no other skin lesions elsewhere. Besides, he denied a history of atopy, tuberculosis, diabetes mellitus, rheumatic diseases, and was not under any topical or systemic medications. He had no known history of contact with tuberculosis, leprosy, or other infectious illness as well. He was afebrile at the time of presentation with average built and normal vital parameters. The cutaneous examination revealed erythematous and skin-colored papules and nodules, discrete to coalescing of size ranging from 0.4 to 1.5 cm involving his pinna and covering the helix, anti-helix and lobules of both ears (fig. 1, 2). The lesions were asymptomatic, i.e. non-pruritic and nontender. There were neither areas of paresthesia, hypoesthesia over any of his body parts, nor systemic symptoms. There was no palpable nerve thickening. Systemic examination was to normal limits. With all these clinical details, differentials

Figure 1. Patient's right ear lobe with multiple nodules involving pinna, helix, antihelix, and lobule.

Figure 2. Patient's left ear lobe with multiple nodules involving pinna, helix, antihelix, and lobule.

were made which included Hansen's disease, Turkey Ear, or other granulomatous skin diseases like lupus pernio, lupoid leishmaniasis, deep-seated mycosis, cutaneous lymphoma and granuloma annulare.[4] For that, we proceeded with slit skin smear (SSS) test with modified Ziehl-Nelson staining of the pinna which revealed acid-fast bacilli (AFB) of *Mycobacterium leprae* with high bacteriological index showing globi (Bacillary index of +6) (fig. 3). As the findings were suggestive of multi-bacillary mycobacterial disease, possibly multibacillary Hansen's disease, thus other non-mycobacterial differentials were ruled out. Turkey ear, a form of cutaneous tuberculosis could have been a near differential diagnosis as it is caused by acid fast bacilli, Mycobacterium tuberculosis. But, its diagnosis remains questionable because detecting mycobacteria in skin lesions using conventional laboratory examination remains difficult and even cultures are often negative.[5] Biopsies are usually needed to definitively confirm a diagnosis and specify the spectrum of disease. However, skin biopsy was not done in the background of positive SSS as the patient denied it, which is our greatest limitation.

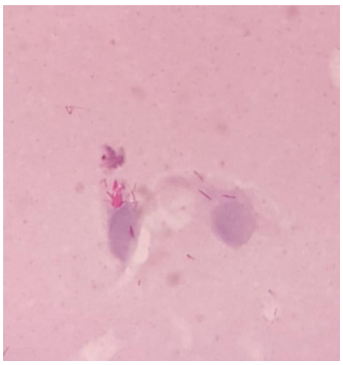

Figure 3. Slit skin smear examination with globi in ZNS (Magnification 100X in oil immersion microscope

DISCUSSION

Leprosy is, unfortunately, still a public health concern in Nepal and globally. Although the national data highlights the elimination of leprosy from Nepal since 2010, local and regional data in Nepal directs in the other way with the prevalence rate more than one per ten thousand.[2] A disease, once thought to be on the verge of becoming history following the introduction of multidrug therapy with a gradual resurgence of incidence in this part of the world, is a cause for real concern.[1]

Leprosy has a wide spectrum of disease presentation based on the immune status of the patient with *M leprae*. The WHO classifies leprosy into paucibacillary and multibacillary based on the number of lesions, nerves and bacillary index. Ridley and Jopling into six classes, ranging from the tuberculoid pole with high cell-mediated immunity on one pole to the lepromatous pole on the other.[3]

Ear lobe is the common site for SSS as it's the easily approachable cooler area of skin harboring *M leprae*. The involvement of earlobe in leprosy ranges from discrete nodules, ulcers, to perichondritis which is a common finding. 'Buddha ear' or megalobule deformity of the pinna has been reported, but isolated involvement of the pinna without nerve involvement and systemic manifestation is supposed to be rare. Ear lobes can be involved in all forms of leprosy and even in reactions including erythema nodosum leprosum (ENL). Skin biopsy with Fite-Faraco staining of the lesions would have helped us to classify concretely the pole of leprosy.[6-9]

Our patient had atypical isolated lesions in both ear lobes without any neurological deficit. Nerve involvement in the tuberculoid pole is early whereas the lepromatous pole is late.[3] Differential diagnoses of pinna swelling include conditions like Turkey ears, lupus pernio, lupoid leishmaniasis, deep mycosis, cutaneous lymphoma and granuloma annulare relapsing polychondritis, otophyma or other forms of granulomatous infiltration of ear lobule.[4]

Limitation of this case study is that we were unable to confirm the diagnosis with biopsy and histopathology, but the findings of SSS were suggestive of multi-bacillary Hansen's diseases with the possibility of lepromatous leprosy.

Here, we highlight the atypical presentation of common infectious diseases which highlights the need for a careful physical examination and histopathological confirmation. In conclusion, isolated involvement of the pinna in a patient without lesions in other body parts could be an unusual initial presentation of multi-bacillary leprosy.

REFERENCES

1. Government of Nepal, Ministry of Health, Department of Health Services, Leprosy Control Division [Internet]. Nepal; Leprosy control division, Teku, Kathmandu;2017 [cited December 2020]. Available from https://lcd.gov.np
2. Leprosy [Internet]; World health organization; 10th September 2019 [Updated 2019 March 2019; cited 21 December 2020]: Available from https://www.who.int/news-room/fact-sheets/detail/leprosy
3. Lastória JC, Abreu MA. Leprosy: review of the epidemiological, clinical, and etiopathogenic aspects - part 1. *An Bras Dermatol*. 2014 Mar-Apr;89(2):205-18. doi: 10.1590/abd1806-4841.20142450. PMID: 24770495; PMCID: PMC4008049.
4. Küçükünal A, Ekmekçi TR, Sakız D. "Turkey ear" as a cutaneous maniestation of tuberculosis. *Indian J Dermatol.* 2012 Nov;57(6):504. doi: 10.4103/0019-5154.103088. PMID: 23248385; PMCID: PMC3519274.
5. Okazaki M, Sakurai A. Lupus vulgaris of the earlobe. *Ann Plast Surg*. 1997 Dec;39(6):643-6. doi: 10.1097/00000637-199712000-00016. PMID: 9418927.
6. Mansfield RE, Storkan MA, Cliff IS. Evaluation of the earlobe in leprosy. A clinical and histopathological study. *Arch Dermatol.* 1969 Oct;100(4):407-12. doi:10.1001/archderm.1969.01610280025003 PMID: 4902083.
7. Paudel D, Sah BP, Bhandary S, Shilpakar SL, Chettri ST, Mishra S. Isolated leprosy of pinna masquerading perichondritis: a rarest entity. *American Journal of Medical Case Reports.* 2014;2(4):87-9. doi: 10.12691/ajmcr-2-4-5
8. Pruthi P, Munganda H, Bangia A, Rani U, Budhiraja R, Brajpuriya S. Leprosy with Atypical Skin Lesions Masquerading as Relapsing Polychondritis. *Case Rep Infect Dis.* 2016;2016:7802423. doi:10.1155/2016/7802423 Epub 2016 Dec 26. PMID: 28116186; PMCID: PMC5220437.
9. Bubna AK. Localized Glistening Nodules of the Right Ear: A Rare De novo Occurrence of Histoid Leprosy. *Indian journal of dermatology.* 2016;61(6):193701-4. doi: 10.4103/0019-5154.193703 PMID: 27904201 PMCID: PMC5122298.