EDITORSIAL

Management of autoimmune urticaria
Arun C. Inamadar, Aparna Palit

VIEWPOINT

Cosmetic dermatology versus cosmetology: A misnomer in need of urgent correction
Shyam B. Verma, Zoe D. Draelos

REVIEW ARTICLE

Psoriasiform dermatoses
Virendra N. Sehgal, Sunil Dogra, Govind Srivastava, Ashok K. Aggarwal

ORIGINAL ARTICLES

A study of allergen-specific IgE antibodies in Indian patients of atopic dermatitis
V. K. Somani

Chronic idiopathic urticaria: Comparison of clinical features with positive autologous serum skin test
George Mamatha, C. Balachandran, Prabhu Smitha

Autologous serum therapy in chronic urticaria: Old wine in a new bottle
A. K. Bajaj, Abir Saraswat, Amitabh Upadhyay, Rajetha Damisetty, Sandipan Dhar

Use of patch testing for identifying allergen causing chronic urticaria
Ashimav Deb Sharma

Vitiligoid lichen sclerosus: A reappraisal
Venkat Ratnam Attili, Sasi Kiran Attili
**BRIEF REPORTS**

Activated charcoal and baking soda to reduce odor associated with extensive blistering disorders
Arun Chakravarthi, C. R. Srinivas, Anil C. Mathew ................................................................. 122

Nevus of Ota: A series of 15 cases
Shanmuga Sekar, Maria Kuruvila, Harsha S. Pai ................................................................. 125

Premature ovarian failure due to cyclophosphamide: A report of four cases in dermatology practice
Vikrant A. Saoji .................................................................................................................. 128

**CASE REPORTS**

Hand, foot and mouth disease in Nagpur
Vikrant A. Saoji .................................................................................................................. 133

Non-familial multiple keratoacanthomas in a 70 year-old long-term non-progressor HIV-seropositive man
Hemanta Kumar Kar, Sunil T. Sabhnani, R. K. Gautam, P. K. Sharma, Kalpana Solanki, Meenakshi Bhardwaj ................................................................. 136

Late onset isotretinoin resistant acne conglobata in a patient with acromegaly
Kapil Jain, V. K. Jain, Kamal Aggarwal, Anu Bansal ................................................................. 139

Familial dyskeratotic comedones
M. Sendhil Kumaran, Divya Appachu, Elizabeth Jayaseelan ................................................................. 142
<table>
<thead>
<tr>
<th>Title</th>
<th>Authors</th>
<th>Page</th>
</tr>
</thead>
<tbody>
<tr>
<td>Nasal NK/T cell lymphoma presenting as a lethal midline granuloma</td>
<td>Vandana Mehta, C. Balachandran, Sudha Bhat, V. Geetha, Donald Fernandes</td>
<td>145</td>
</tr>
<tr>
<td>Childood sclerodermatomyositis with generalized morphea</td>
<td>Girishkumar R. Ambade, Rachita S. Dhurat, Nitin Lade, Hemangi R. Jerajani</td>
<td>148</td>
</tr>
<tr>
<td>Subcutaneous panniculitis-like T-cell cutaneous lymphoma</td>
<td>Avninder Singh, Joginder Kumar, Sujala Kapur, V. Ramesh</td>
<td>151</td>
</tr>
<tr>
<td>Letters to Editor</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Using a submersible pump to clean large areas of the body with antiseptics</td>
<td>C. R. Srinivas</td>
<td>154</td>
</tr>
<tr>
<td>Stratum corneum findings as clues to histological diagnosis of pityriasis lichenoides chronica</td>
<td>Rajiv Joshi</td>
<td>156</td>
</tr>
<tr>
<td>Author’s reply</td>
<td>S. Pradeep Nair</td>
<td>157</td>
</tr>
<tr>
<td>Omalizumab in severe chronic urticaria</td>
<td>K. V. Godse</td>
<td>157</td>
</tr>
<tr>
<td>Hypothesis: The potential utility of topical eflornithine against cutaneous leishmaniasis</td>
<td>M. R. Namazi</td>
<td>158</td>
</tr>
<tr>
<td>Nodular melanoma in a skin graft site scar</td>
<td>A. Gnaneshwar Rao, Kamal K. Jhamnani, Chandana Konda</td>
<td>159</td>
</tr>
</tbody>
</table>
Palatal involvement in lepromatous leprosy
A. Gnaneshwar Rao, Chandana Konda, Kamal Jhamnani ................................................................. 161

Unilateral nevoid telangiectasia with no estrogen and progesterone receptors in a pediatric patient
F. Sule Afsar, Ragip Ortac, Gulden Diniz .......................................................................................... 163

Eruptive lichen planus in a child with celiac disease
Dipankar De, Amrinder J. Kanwar ........................................................................................................ 164

Xerosis and pityriasis alba-like changes associated with zonisamide
Feroze Kaliyadan, Jayasree Manoj, S. Venkitakrishnan ........................................................................ 165

Treatment of actinomycetoma with combination of rifampicin and co-trimoxazole
Rajiv Joshi ................................................................................................................................................... 166

Author’s reply

Vitiligo, psoriasis and imiquimod: Fitting all into the same pathway
Bell Raj Eapen ........................................................................................................................................... 169

Author’s reply
Engin Şenel, Deniz Seçkin ....................................................................................................................... 169

Multiple dermatofibromas on face treated with carbon dioxide laser: The importance of laser parameters
Kabir Sardana, Vijay K. Garg .................................................................................................................... 170

Author’s reply

Alopecia areata progressing to totalis/universalis in non-insulin dependent diabetes mellitus (type II): Failure of dexamethasone-cyclophosphamide pulse therapy
Virendra N. Sehgal, Sambit N. Bhattacharya, Sonal Sharma, Govind Srivastava, Ashok K. Aggarwal ........................................................................................................... 171

Subungual exostosis
Kamal Aggarwal, Sanjeev Gupta, Vijay Kumar Jain, Amit Mital, Sunita Gupta ........................................... 173
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Clinicohistopathological correlation of leprosy
Amrish N. Pandya, Hemali J. Tailor ............................................................................................................................ 174

RESIDENT’S PAGE
Dermatographism
Dipti Bhute, Bhavana Doshi, Sushil Pande, Sunanda Mahajan, Vidya Kharkar ........................................................ 177

FOCUS
Mycophenolate mofetil
Amar Surjushe, D. G. Saple ......................................................................................................................................... 180

QUIZ
Multiple papules on the vulva
G. Raghu Rama Rao, R. Radha Rani, A. Amareswar, P. V. Krishnam Raju, P. Raja Kumari, Y. Hari Kishan Kumar .............................................................................................................. 185

EIJDV
Net Study
Oral isotretinoin is as effective as a combination of oral isotretinoin and topical anti-acne agents in nodulocystic acne
Rajeev Dhir, Neetu P. Gehi, Reetu Agarwal, Yuvraj E. More .................................................................................... 187

Net Case
Cutaneous diphtheria masquerading as a sexually transmitted disease
T. P. Vetrichevvel, Gajanan A. Pise, Kishan Kumar Agrawal, Devinder Mohan Thappa ............................................................................................................................................. 187

Net Letters
Patch test in Behcet’s disease
Ülker Gül, Müzeyyen Gönlü, Seray Külcü Çakmak, Arzu Kulç ................................................................................ 187

Cerebriform elephantiasis of the vulva following tuberculous lymphadenitis
Surajit Nayak, Basanti Acharjya, Basanti Devi, Satyadarshi Pattnaik, Manoj Kumar Patra ...................................................................................................................................................... 188

Net Quiz
Vesicles on the tongue
Saurabh Agarwal, Krishna Gopal, Binay Kumar ....................................................................................................... 188
Nevus of Ota: A series of 15 cases

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ABSTRACT

Background: The nevus of Ota is a dermal nevus characterized by bluish pigmentation in the distribution of the first and the second division of the trigeminal nerve. Aim: Our aim was to study the cutaneous and extracutaneous manifestations of the nevus of Ota. Methods: A total of 15 cases were included in our study. A detailed history, clinical examination along with direct ophthalmoscopy and otoscopy were done for all the cases. Results: Most of the patients (60%) had lesions at birth and the majority (86.7%) were females. Five (33.3%) patients belonged to Tanino class II. Combined dermal and ocular involvement was observed in 60% of the cases. Conclusion: Tanino class II was the most common type observed in our studies. A few rarer associations such as nevus of Ito and hemangioma were also noted in our patients.

Key Words: Dermal melanocytosis, Nevus of Ota, Tanino

INTRODUCTION

The term ceruloderma[1] was first introduced in 1980 to describe various clinical syndromes characterized by melanin and nonmelanin pigmentation of the dermis. The nevus of Ota consists of hyperpigmentation of facial skin and mucous membranes in the distribution of the ophthalmic, maxillary and occasionally, the mandibular divisions of the facial nerve.[2] The dermal lesions are bluish, confluent, nonhairy, flat, pigmented macules with poorly defined margins. The melanocytosis also affects the oral cavity, nasal mucosa, external auditory canal, tympanic membrane, orbital fissures, meninges and the brain.[3] Tanino[4] has classified the nevus of Ota into four types:

Type I. IA. Mild orbital type: Distribution over the upper and lower eyelids, periorcular and temple region.

IB. Mild zygomatic type: Pigmentation is found in the infralapalpebral fold, nasolabial fold and the zygomatic region.

IC. Mild forehead type: Involvement of the forehead alone.

ID. Involvement of ala nasi alone.

Type II. Moderate type: Distribution over the upper and lower eyelids, periorcular, zygomatic, cheek and temple regions.

Type III. The lesion involves the scalp, forehead, eyebrow and nose.

Type IV. Bilateral type: Both sides are involved.

We studied the clinical features of 15 cases of nevus of Ota.
RESULTS

Out of the 15 cases, nine (60%) patients presented in their third decade of life followed by three (20%) in their fourth decade. The youngest patient was eight years old and the oldest was 37 years old. The majority of the cases [13 (86.7%)] was female. The female: male ratio were 6.5:1. The lesions were present at birth in nine (60%) cases [Table 1] while in four (26.6%), they appeared in the second decade of life. Only one (6.7%) patient had a history of similar lesions in the family.

Five patients [Table 2] belonged to Tanino class II while three belonged to class III and three to class IV. There were no patients in class ID. Dermal and ocular involvement was observed in nine (60%) cases while dermal involvement alone was observed in six (40%) cases. Of the nine cases with ocular involvement, all had bluish pigmentation in the episclera [Figure 1] while three (33%) had involvement of the palpebral conjunctiva and three of the retina. The alae nasi and hard palate were involved in three (20%) each. None of the patients had any involvement of the tympanic membrane. The nevus of Ito [Figure 2] and a deviated nasal septum were separately observed in two (13.4%) cases. Hemangioma with glaucoma was observed in a patient with bilateral nevus of Ota.

DISCUSSION

Hulkey first described oculodermal melanosis in 1861 and in 1916, Pusey was the first to draw attention to the relationship of a pigmented lesion of facial skin to the pigmentation of ipsilateral sclera in a Chinese student. In 1939, Ota and Tanino described several cases of pigmented nevus of the skin and eye and named them “nevus fuscoceruleus ophthalmomaxillaris of Ota.” Melanocytes move from the neural crest to the skin during early embryonic life. Failure of complete migration into the epidermis before birth with ensuing dermal nesting and melanin production produces characteristic blue patches. Dermal melanin produces blue color because of the Tyndall effect in which all but the blue end of the light spectrum penetrates into the deep dermis and is absorbed by dermal melanin. Sex hormones have been implicated in the pathogenesis of the nevus of Ota.

![Figure 1: Episcleral involvement in nevus of Ota](image1)

![Figure 2: Nevus of Ito in a patient with nevus of Ota](image2)

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<thead>
<tr>
<th>Table 1: Age of onset</th>
</tr>
</thead>
<tbody>
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<td>Age (years)</td>
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<td>Birth</td>
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<th>Table 2: Types of nevus of Ota</th>
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proposed that stress and trauma may be accompanied by increased secretion of propiomelanocortin, a precursor of melanocyte-stimulating hormone. Under these conditions, a small unnoticed cell nest could be stimulated to enlarge in size. One patient gave a history of similar lesions in her grandfather. Very few instances of familial occurrence have been reported. Trese et al.[9] and Teekhasaenee et al.[5] have reported familial incidence in their studies.

In our study, five (33.3%) cases belonged to Tanino class II while three (20%) each belonged to classes III and IV. In a study by Teekhasaenee et al.[5] 27% each belonged to classes II and III. Blue colored lesions were observed in 13 of our patients while two had slate grey lesions. The color of the lesions depends blue in color due to the Tyndall effect whereas the more superficial lesions are slate grey in color. The grey-colored lesions were examined under Wood’s lamp, which caused attenuation in the intensity confirming the dermal origin.

Twelve patients (60%) had unilateral involvement while three (20%) had bilateral involvement. Bilateral involvement is rare and seen only in 4% of all cases.[4] In our study, nine (60%) patients had both ocular and dermal involvement while six (40%) had only dermal involvement. Ocular involvement alone was not seen. In a study by Teekhasaenee et al.[5] 59.3% had ocular and dermal involvement while 35% had dermal involvement. Of the nine cases with ocular involvement, all had episcopal involvement while three (33.3%) each had involvement of the palpebral conjunctiva and the retina. Teekhasaenee et al.[5] reported 100% episcopal involvement, 10% conjunctival involvement and 18% retinal involvement. Glaucoma was observed in a case of bilateral Nevus of Ota. Foulks et al.[9] and Khawly et al.[10] have reported this association. Obstruction to aqueous outflow by accumulated melanocytes is the mechanism which is most likely to be directly associated with the nevus of Ota. Palatal involvement was observed in 20% of the patients. Page et al.[11] and Rathi[12] have reported cases of Nevus of Ota with palatal involvement. None of the patients had any tympanic membrane involvement in our study.

Nevus of Ito was observed in a patient along with bilateral nevus of Ota. This association is very rare. Hidano et al.[13] has described a case of bilateral nevus of Ota with nevus of Ito with pigmentation of lips while Mukhoupahady[14] has reported a case of unilateral nevus of Ota and nevus of Ito in an Indian male. Hemangioma was observed in a patient with bilateral nevus of Ota. Reinke et al.[15] has reported a case of nevus of Ota with hemangioma and Takayasu arteritis. Neural crest cells may be disturbed by an overlying vascular malformation, thus explaining the occurrence of nevus of Ota.

To conclude, a majority of our patients of nevus of Ota presented in the third decade of life and most of them were females. Tanino class II was the most common type observed in our studies. Rare presentations observed in our study were bilateral nevus of Ota associated with Nevus of Ito and bilateral Nevus of Ota with bilateral hemangioma and glaucoma. Hence, a detailed systemic examination along with ophthalmological and oral examination is required in all cases of nevus of Ota. The psychological impact of these lesions is high and early treatment with lasers would considerably reduce the stress later in life. The lesions are usually asymptomatic but lifelong follow-up is required as a few cases of malignant melanoma have been reported in literature.

REFERENCES