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CASE REPORT

A Case of Nevus of Ota Associated with Agminated Blue Nevus

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Abstract: Dermal melanocytosis is characterized by the presence of spindle-shaped melanocytes in the dermis. Common forms include Mongolian spots, nevus of Ota, nevus of Ito, and blue nevus. Nevus of Ota typically presents as bluish-brown patches involving the sclera and the cutaneous regions innervated by the ophthalmic and maxillary branches of the trigeminal nerve unilaterally. Blue nevus usually manifests as a solitary lesion. Agminated blue nevus is a rare variant that often presents as grouped, linear, or blaschkoid-distributed deep blue macules or papules. Only a few cases have reported the co-occurrence of nevus of Ota and agminated blue nevus. We herein report a case of extensive nevus of Ota combined with agminated blue nevus involving all three branches of the trigeminal nerve.

Keywords: nevus of ota, agminated blue nevus, ocular melanocytosis, oral pigmentation

Introduction

Facial pigmentation disorders represent a significant category in clinical dermatology, consistently attracting attention from both clinicians and patients. Among these, nevus of Ota and blue nevus are two distinct entities that can present as pigmented macules on the face. While each condition is individually well-documented, the co-occurrence of unilateral nevus of Ota with agminated blue nevus is exceedingly rare. This unusual clinical presentation poses considerable challenges in terms of diagnosis and therapeutic decision-making. We report the case of a 15-year-old male with a left-sided blue-brown facial macule present since birth. The diagnosis of nevus of Ota combined with agminated blue nevus was confirmed through dermoscopic and histopathological examination, as reported below.

Case Report

A 15-year-old male patient presented with a 15-year history of dark brown to blue-black facial pigmentation. The pigmented macules had been present since infancy, with gradual expansion in area and darkening in color over time, accompanied by the development of new blue-black papules. The patient was otherwise in good health and reported no family history of melanoma or similar dermatologic conditions.

Dermatologic examination revealed extensive, irregular, brownish patches distributed unilaterally on the left side of the face, involving the forehead, periorbital region, temple, zygomatic area, and cheek. Superimposed on these patches were multiple bluish maculopapular lesions, which appeared either scattered or confluent, without surface erosion or ulceration. The pigmentation also involved the left sclera and the oral mucosa on the same side (Figure 1).

Dermoscopic examination revealed multiple, grouped, and confluent brown to blue-black pigment structures against a background of non-structured areas in shades of blue, brown, and gray (Figure 2).

Histopathological assessment showed a largely unremarkable epidermis. The superficial dermis exhibited diffusely distributed bipolar and spindle-shaped melanocytes. Nests of spindle-shaped melanocytes were also observed scattered

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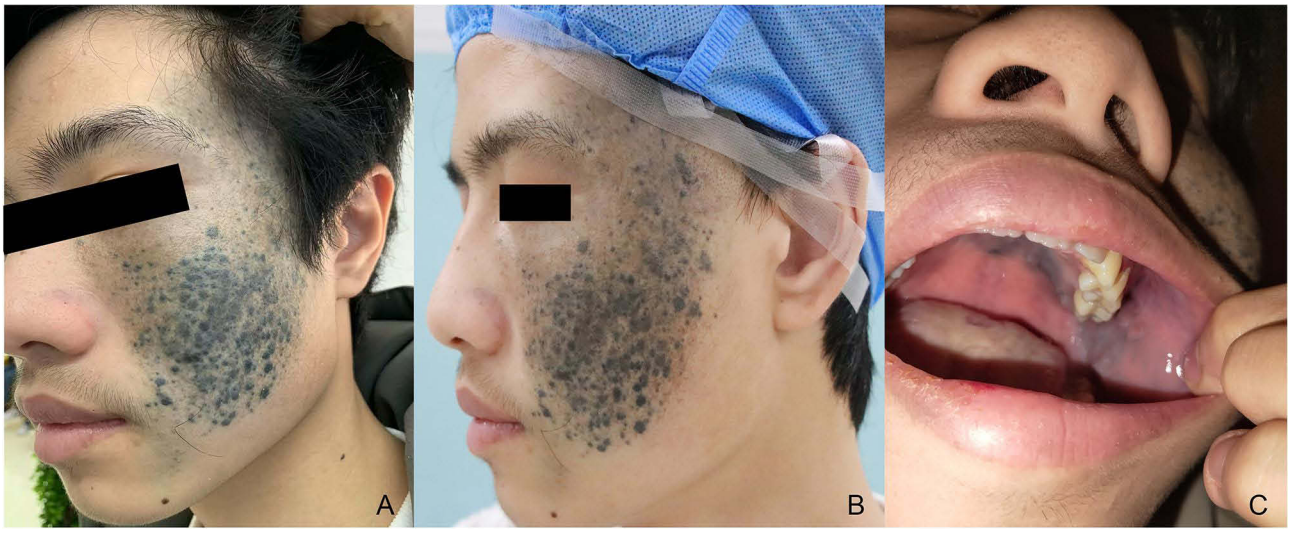

Figure 1 Clinical presentation of the patient. (**A** and **B**) Irregular dark brown to blue-black macules on the left side of the face, with interspersed blue-black papules of varying sizes. (**C**) Pigmentation involving the left sclera and buccal mucosa.

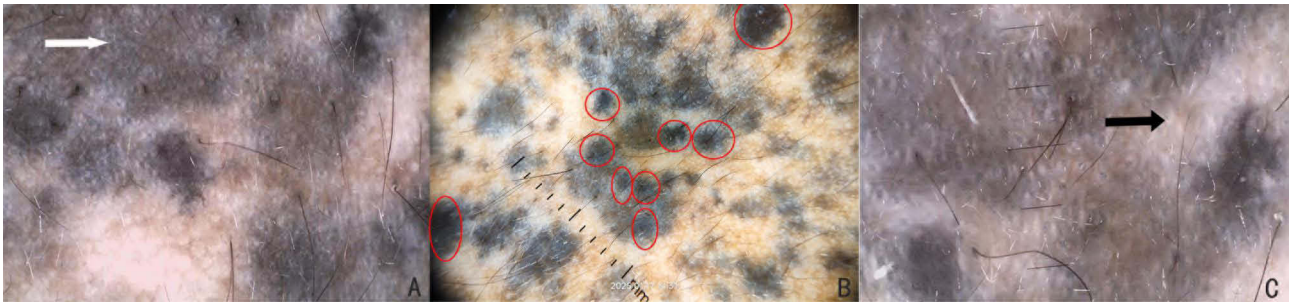

Figure 2 Dermoscopic findings. (**A**) Diffuse brown and gray structureless areas corresponding to nevus of Ota (white arrows). (**B**) Homogeneous blue to blue-black structures with relatively well-defined borders, consistent with blue nevus (red circles). (**C**) Perifollicular hypopigmentation is observed (black arrows).

within the mid-dermis and subcutaneous adipose tissue. Some cells displayed epithelioid morphology with abundant intracytoplasmic pigment granules; no nuclear atypia or mitotic figures were identified (Figure 3). Brain magnetic resonance imaging (MRI) revealed no evidence of an intracranial mass.

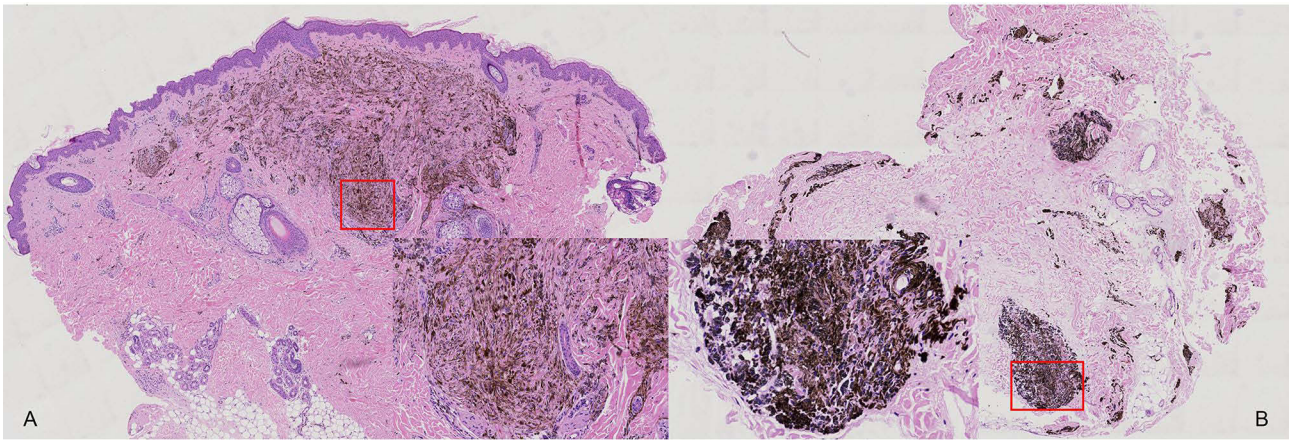

Figure 3 Histopathological examination (**A**) Diffuse infiltration of melanocytes within the superficial dermis (HE×20; inset: red coloured box×200). (**B**) Nests of melanocytes intermingled with collagen bundles, extending into the subcutaneous tissue (HE×20; inset: red coloured box×200).

Discussion

Nevus of Ota typically presents as patchy grayish-brown to gray-blue pigmentation involving the sclera, conjunctiva, cornea, retina, or oral mucosa. Lesions are predominantly unilateral, with bilateral involvement observed in approximately 10% of cases. The distribution commonly follows the first and second branches of the trigeminal nerve, while extension to the third branch is rare.[1] Histopathological examination reveals scattered dendritic melanocytes within the upper dermis, occasionally extending to the papillary dermis and subcutaneous tissue, with melanophages being less frequently observed.[2] In some cases, discrete papules or nodules resembling blue nevus may arise within the nevus of Ota, which can present diagnostic challenges.

Blue nevus typically manifests as solitary, well-circumscribed blue or blue-black macules or papules, usually less than 10 mm in diameter. Agminated blue nevus (ABN) is currently defined as the presence of multiple lesions within an area ≤ 10 cm in diameter. Dermoscopic evaluation of ABN often reveals a predominant blue structureless pattern, frequently accompanied by brown or gray structureless areas and white or skin-colored regions.[3] Under high-frequency ultrasound examination, blue nevus typically presents as regular, oval-shaped, hypoechoic areas with heterogeneous echotexture and ill-defined margins within the dermal layer.[4] Histologically, ABN is characterized by infiltrates of dermal dendritic melanocytes and melanophages, which may extend into the subcutaneous tissue, superficial fascia, and muscle.[5] Despite its distinct clinical and histopathological features, the co-occurrence of ABN within a nevus of Ota, as observed in the present case, underscores the importance of comprehensive evaluation to avoid misdiagnosis.

Clinically, blue nevus presents as blue-to-black or brown papules, plaques, or nodules, distinguishing it from the brown-to-blue patches characteristic of nevus of Ota.[1] Dermoscopy is valuable for the differential diagnosis of both entities. Furthermore, high-frequency ultrasound may serve as an auxiliary tool, providing useful information regarding lesion depth, internal architecture, and the distinction between benign and malignant features. Nevertheless, histopathological examination remains the diagnostic gold standard.[6,7] Histologically, both nevus of Ota and blue nevus exhibit dendritic melanocytes; however, cellular density is typically higher in blue nevus and more sparse in nevus of Ota.[8] Although the coexistence of nevus of Ota and blue nevus is not uncommon, the presentation of agminated blue nevus with extensive overlap remains extremely rare.[1,9] Emerging evidence suggests that the pathogenesis of such combined lesions may involve novel molecular events, such as GRM1 gene fusions, rather than traditional GNAQ mutations, and must be differentiated from melanoma, pigmented basal cell carcinoma, and malignant blue nevus.[10]

Notably, nevus of Ota occurring in combination with blue nevus carries a potential risk of malignant transformation. Several cases of melanoma arising within such combined lesions have been documented in the literature. These reports suggest that blue nevus-like components may serve as an intermediate or concurrent manifestation in the process of malignant transformation.[11,12]

Regarding management, the primary therapeutic option for nevus of Ota is laser therapy, most commonly utilizing Q-switched or picosecond lasers. Multiple treatment sessions typically lead to satisfactory clearance.[8,13] In contrast, blue nevus demonstrates poor responsiveness to laser modalities, including the 1064 nm Q-switched Nd:YAG and 755 nm picosecond lasers, as reported in the literature. Surgical excision is therefore considered the first-line treatment for blue nevus.[8] In the present case, due to the extensive and clustered distribution of the blue nevus component, along with its limited laser responsiveness and potential for malignancy, surgical excision following plastic surgery consultation was considered. However, given the large involved area and potential cosmetic impact, along with the absence of current malignant features, the patient opted for regular follow-up. Therefore, for such extensive and structurally complex lesions, patients should be thoroughly informed of the malignancy risk. Brain MRI is recommended to exclude intracranial involvement, and long-term regular follow-up—supported by dermoscopic monitoring—is essential. Any new or rapidly changing nodules, particularly those with ulceration, should prompt prompt pathological biopsy for definitive diagnosis.

We present a case of extensive nevus of Ota combined with agminated blue nevus. Nonetheless, this report has several limitations. First, it describes a single case. Second, the follow-up period was limited to one year; therefore, the long-term stability and prognosis of this combined entity warrant further observation.

Informed Consent

Informed consent was obtained from the patient and his guardian for the publication of clinical information and related images. The Ethics Committee of Guangzhou Dermatology Hospital has approved the publication of the case details (gzsp202565).

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Disclosure

Xinyi Xie and Jiaoquan Chen are co-first authors for this study. No potential conflict of interest was reported by the authors.

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