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Scleredema of Buschke Following Streptococcal Infection

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Dear Editor:

Scleredema is characterized by non-pitting hardening of the skin that typically begins on the neck and subsequently spreads to the shoulders, back, and face. Histopathologically, it is characterized by excessive mucin deposition between collagen bundles. It is clinically classified into 3 types:postinfectious scleredema (type I), paraproteinemia-associated scleredema (type II), and scleredema diabeticorum (type III)[1-4]. Here, we report a case of scleredema of Buschke following a streptococcal infection.

An 8-year-old boy presented with a 1-week history of skin firmness of the whole body. He experienced a febrile upper-respiratory infection 2 months prior with abrupt onset of skin hardening starting from his posterior neck and rapidly extending to his face, trunk, arms, and legs. On physical examination, whole-body and non-pitting induration of the skin were noted, especially on the posterior neck and upper back (Fig. 1). A biopsy from the posterior neck revealed dermal thickening with a square-like appearance, and a high-magnification view revealed slight widening of collagen bundles with normal fibroblast number (Fig. 2A, B). Mucin deposition was detected between collagen bundles (Fig. 2C). No other laboratory abnormalities except an elevated antistreptolysin O titer (476 U/L [normal range: 0 ~160 U/L]) were noted. Therefore, he was diagnosed with scleredema of Buschke following a streptococcal infection.

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Scleredema is also termed "scleredema adultorum of Buschke," "scleredema adultorum," or "scleredema of Buschke." The term "scleredema adultorum" was suggested by Buschke in 1902 to distinguish this entity from sclerema neonatorum[2,3]. It is usually classified into 3 types; however, the nomenclature of each subgroup has not been established. Moreover, classification systems differ among sources. Type I, known as the classic type, is characterized by a sudden onset of skin hardening after an acute febrile illness in children and adolescents. The preceding infection is usually bacterial or viral in origin, most commonly a streptococcal upper-respiratory infection. Thus, we suggest referring to this type "postinfectious scleredema,"[1] to distinguish it from other types of scleredema or restricting the use of the term "scleredema of Buschke" to describe type I scleredema as it was first described by Buschke himself. Furthermore, the term "scleredema adultorum" should be avoided because it may cause confusion among physicians. Meanwhile, type II scleredema has an insidious onset and is usually associated with paraproteinemia, hypergammaglobulinemia, or multiple myeloma. Finally, type III scleredema, also called scleredema diabeticorum, is related to poorly controlled type 2 diabetes. However, there are other cases associated with insulinoma, secondary hyperparathyroidism, rheumatoid arthritis, or HIV infection that do not meet the criteria of any of the above mentioned type[4]. Type I scleredema is usually resolved within 2 years and does not require treatment. Thus, our patient has not been treated and has shown a resolution of symptoms over the past 5 months. If symptoms persist, phototherapy including psoralen plus ultraviolet A, ultraviolet A-1, and narrow-band ultraviolet B as well as radiotherapy and intravenous immunoglobulins should be considered. In conclusion, a classification system for scleredema must be established. In addition, type I scleredema should be named "postinfectious scleredema"

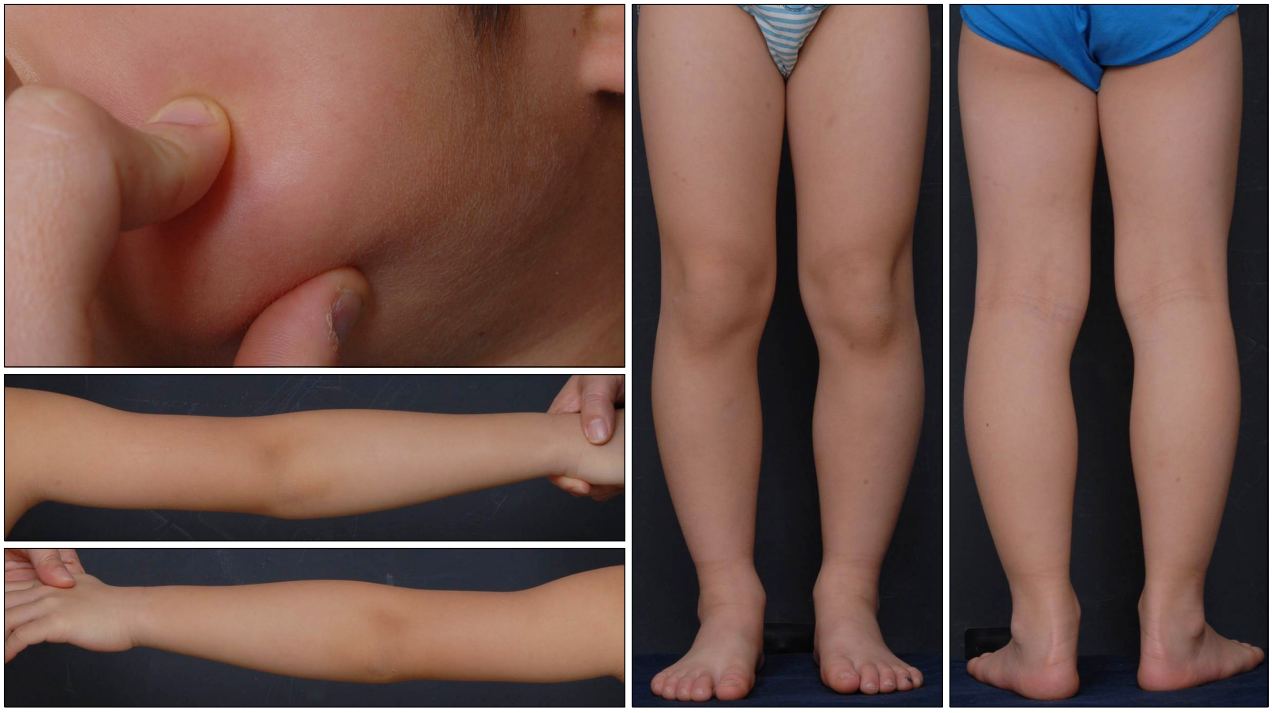

Fig. 1. Whole-body and non-pitting induration of the skin was noted including face. On physical examination, it was hard to pinch his check.

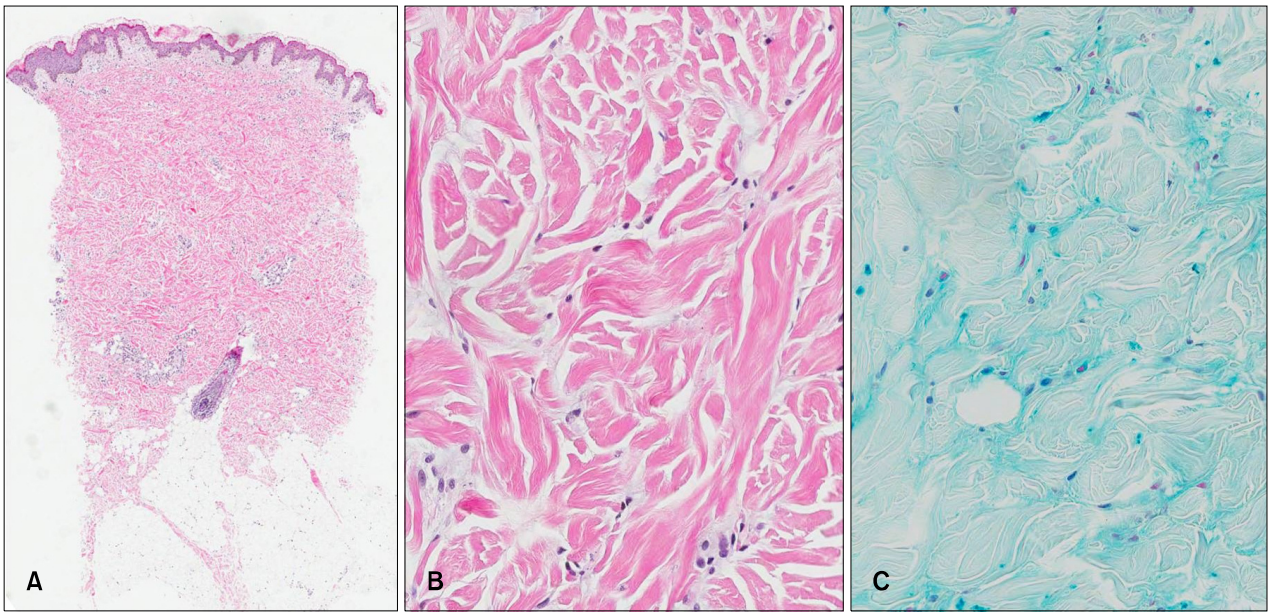

Fig. 2. (A) Skin biopsy specimen revealed a non-tapered appearance on a low-power field (H&E, ×40). (B) Fibroblasts were normal in number, and the spaces between collagen bundles were widened (H&E, ×400). (C) Mucin deposits were detected between collagen bundles in the middermis (alcian blue staining, ×400).

or "scleredema of Buschke."

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Acral Angioosteoma Cutis on the Great Toe Mimicking Pyogenic Granuloma

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Dear Editor:

Acral angioosteoma cutis (AAOC) is a rare benign exophytic tumor characterized by bony and vascular proliferation. Clinically, it occurs on acral skin and resembles pyogenic granuloma (PG). Here, we report an unusual case of AAOC that developed on the periungual area of the great toe.

A 12-year-old girl presented with a solitary tender crusted nodule on her left great toe. She had been suffering from an ingrown nail and the lesion had first appeared two months earlier after a partial nail extraction. The lesion increased in size over time. The patient was otherwise healthy. Physical examination revealed an erythematous to blackish, somewhat firm, crusted periungual nodule, measuring 5 mm in diameter, with paronychia on her left great toe (Fig. 1A). Clinically, the lesion was suspicious for PG or granulation tissue. The lesion was removed by curettage and a histopathological examination revealed a well-circumscribed polypoid tumor containing multiple bony trabeculae and small vessel proliferation in the stroma (Fig. 1B, C). Well-formed dilated capillaries that showed neither a lobular pattern nor endothelial atypia were distributed diffusely among the bony trabeculae. These findings were consistent with a diagnosis of AAOC. We followed the lesion for 1 year and observed no recurrence.

In 2006, Googe et al.[1]. first described AAOC as a benign tumor of unknown histogenesis that is distinctive from other cutaneous lesions showing calcification or ossification. Clinically, AAOC can be easily misdiagnosed as one of the benign acral neoplasms[2-5]. In other similar conditions, the subungual exostosis differs from that in AAOC in that a fibrocartilaginous cap surrounds the lesion without vascular proliferation. In addition, ectopic bone formation protruding from the skeleton is typically found on radiography. Both osteoma cutis and fibro-osseous pseudotumor of the digit show ossification without vascular proliferation. Osteochondroma also has no vascular channels and is characterized by bony trabeculae covered with hyaline cartilage. PG showing metaplastic ossification is the most difficult entity to differentiate from AAOC[4,5], but can be differentiated by its typical lobular vascular proli-

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