Solitary Milialike Idiopathic Calcinosis Cutis Unassociated with Down’s Syndrome: Two Case Reports

Sir,
In 1978, Sano et al. (1) first described a case of milialike idiopathic calcinosis cutis (MICC), and since then 12 cases have been reported in the literature (2–10). Most cases of MICC develop in children with Down’s syndrome and/or syringoma (1, 3–9), and only 3 cases have been reported in the absence of Down’s syndrome (2, 10). We report here 2 cases of solitary MICC unassociated with Down’s syndrome, one occurring on the right sole of a 2-year-old boy and the other on the lateral side of the left 5th finger of a 55-year-old woman.

CASE REPORTS

The first patient, a 2-year-old healthy boy, presented at our hospital complaining of a small, tender papule, which had been present on his right sole for 1 year. His past medical history and familial history were non-contributory. His physical and mental developments were normal. Examination revealed a firm, whitish, dome-shaped, verrucous papule, 3 mm in diameter, on his right sole (Fig. 1A). The patient’s parents denied any history of previous trauma at the site of lesion. No other cutaneous abnormalities were observed. A diagnosis of plantar wart or epidermal cyst was made and the lesion was treated with shave biopsy followed by electrodessication. Histological examination of the specimen showed marked hyperkeratosis and acanthosis in the epidermis, and closely aggregated deposition of basophilic, amorphous material surrounded by thick collagen fibres and fibroblasts in the upper dermis. The calcific nature of the material was confirmed by staining with Von Kossa. Laboratory findings, including serum calcium and phosphorus levels, were within normal limits. During 6 months of follow-up, there has been no recurrence.

The second patient, a 55-year-old woman was seen because of a tender, whitish papule on the lateral side of the left 5th finger. Her finger had been pricked by a rose thorn 1 month before. The lesion was firm, scaly, whitish and pinhead-sized (Fig. 1B). There was no specific finding in the past and family history. On physical examination, there was no similar lesion elsewhere. Histological examination of the specimen obtained by excision biopsy revealed a roundish calcified deposit surrounded by a thin fibrovascular strip in the papillary dermis (Fig. 2). Laboratory findings, including serum calcium and phosphate levels, were within normal limits. During the follow-up period of 4 months after the operation, there has been no recurrence of the lesion or development of a new lesion.

DISCUSSION

MICC is a peculiar subtype of idiopathic calcinosis cutis, showing the characteristic clinical and histological features (1–10). Clinically, the lesions appear as multiple, round, whitish papules, resembling milia. The most common locations are the hands and feet, although it can occur on other part of the body. Most patients have been reported in association with Down’s syndrome and/or syringoma (1, 3–9). On histological examination, it is characterized by a well-defined, roundish, calcified deposit surrounded by a fibrous rim in the papillary dermis, mimicking a pseudocyst structure, which separates this entity from other forms of calcinosis cutis. The clinical and histological features in the cases described here are somewhat similar to MICC reported previously. In the cases described here the lesion appeared as a solitary papule in otherwise healthy persons who had no evidence of Down’s syndrome. In the literature, there have been only 3 cases of MICC unassociated with Down’s syndrome.
syndrome (2, 10). In 1981, Eng & Mandrea (2) reported 2 cases of MICC occurring on the pubic area and groin of 2 healthy children. Recently, Lee et al. (10) described a case of solitary MICC, developing on the dorsum of the right hand of a healthy 21-year-old woman. The latter case and the ones described here suggest that MICC can be either solitary or multiple in the absence of Down’s syndrome.

The pathogenic mechanism of MICC remains unclear. We believe that a preceding local trauma may have been responsible for the development of the lesion in our patients, as sites such as the sole or fingers are vulnerable to minor injury. In the second case the patient had a history of pricking by a rose thorn before the lesion occurred. In our opinion MICC is merely a term describing a calcinosis cutis resembling milia clinopathologically and its pathogenesis may be different in each case.

REFERENCES

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Dai-Ho Kim, Hoon Kang, Sang-Hyun Cho and Young-Min Park
Our Lady of Mercy Hospital, The Catholic University of Korea, 665 Bupyung-dong, Bupyung-gu, Inchon 403-720, Korea.
ladyskin@soback.kornet21.net

Mohs Micrographic Surgery in Gothenburg, Sweden

Sir,
We very much enjoyed the positive comments of Professor Cunliffe on Mohs micrographic surgery (MMS) in Sweden, but we have to make some corrections based on our extensive experience. The paper commented on (1) is a follow-up report to our initial 5-year experience of the procedure. Since then the method has been used regularly and more frequently.

Since 1983 we have operated on more than 660 skin cancers using this procedure and we currently carry out about 70 cases/year and all patients are followed up continuously for 5–10 years. The procedure is updated at regular visits to other Mohs centres abroad, and since 1994 the single section method (2) has been used routinely when applicable, which speeds up the procedure and simplifies the orientation of the tissue specimen.

In order to decrease the number of Mohs layers at each operation, preoperative demarcation of the tumours using fluorescence has been tested. The results are published in a preliminary communication (3). Here the lateral limitations of the tumour could be outlined in about 50% of the cases when checked by MMS. After further refinement of this technique we hope that it will be a valuable complement to MMS.

REFERENCES

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Bo Stenquist, Ann-Marie Wennberg and Olle Larkö
Mohs and Dermatologic Surgery Unit, Department of Dermatology, Sahlgrenska University Hospital, S-413 45 Göteborg, Sweden.

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