KELOIDAL LESIONS DURING D-PENICILLAMINE THERAPY FOR SYSTEMIC SCLEROSIS
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Previous reports on cases of systemic sclerosis (PSS) with keloidal lesions are few; only about a dozen as far as we know. Although several studies have suggested that D-penicillamine (DPC) is effective in the treatment of PSS, morphea-like reaction and PSS-like lesions during DPC therapy have been described. We report here a case of PSS who developed keloidal lesions during DPC therapy.

REPORT OF A CASE
A 36-year-old woman was first seen in our clinic in January, 1980, complaining of Raynaud's phenomenon and edematous sclerosis of the fingers which had been present for one year and arthralgia of the right wrist. Physical examination revealed also the presence of a finger tip microulcer and chest X-p showed a slight increase in interstitial markings. Histological examination of the right forearm revealed a mild increase in dermal collagen fibers, and administration of elastase was started under a diagnosis of PSS. At the age of 37 oral vitamine E was added, while lung fibrosis became rather prominent and the skin sclerosis rapidly progressed to the upper arms, neck, and chest, which was confirmed histologically as well. At the age of 38, in August, 1981, DPC 200 mg/day was begun, then 400 mg/day was given for about 2 years, and since January, 1984 200mg/day has been continued. Gold sodium thiomalate, up to 600 mg altogether, was occasionally injected for joint pain. The skin sclerosis has improved, while pulmonary fibrosis and esophageal dysfunction have persisted without significant improvement. At the age of 44, several keloidal eruptions with itching appeared on the upper chest, one of which was biopsied in January, 1989. Histological examination of the keloidal nodule revealed a relatively narrow zone of fibrosis in the middermis, which consisted of fibroblastic cells and collagen fibers mostly parallel to the skin surface, containing abundant mucin and a decreased quantity of elastic fibers. Desmin was negative, but vimentin was weakly positive in these cells. These clinical and histological findings on the keloidal lesions were close to, but different from, those of keloids or hypertrophic scars.
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DISCUSSION

Since these keloidal lesions developed during DPC therapy, it is possible to regard them as a side effect of DPC. However, it seems more likely that the fibrotic activity in these lesions was too strong to be suppressed by DPC, because skin sclerosis has been otherwise improved. We believe that this case is nodular (keloidal) scleroderma, which is assumed to be extremely rare.

REFERENCES


Fig. 1. Clinical course

Fig. 2. Keloid like lesions on the upper chest.

Fig. 3. The keloidal nodule revealed a relatively narrow zone of fibrosis in the middermis (H-E, x6.6).