

Hand Involvement in Paraneoplastic Syndrome: A Case Report

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Cutaneous paraneoplastic syndromes are skin and mucous membrane changes that are associated with cancer. We report a previously healthy 76-year-old man who developed marked finger and thumb contracture, pain, and hypersensitivity of both hands who was diagnosed subsequently as having gastric carcinoma with colonic metastasis. After the gastrointestinal tumors were resected the finger and thumb contracture lessened and the pain eased. Both the temporal relationship between the changes in the hand and the neoplasm and the improvement after resection suggest a paraneoplastic syndrome. (*J Hand Surg* 2005;30A:1087–1090. Copyright © 2005 by the American Society for Surgery of the Hand.)

Key words: Paraneoplastic syndrome, fibromatosis, scleroderma, palmar fasciitis.

Cutaneous paraneoplastic syndromes are skin and mucous membrane changes that are associated with cancer.¹ They can be the first manifestation of a tumor so it is very important to recognize them or to suspect malignancy in a patient with unusual rheumatic-like disorders.^{1,2}

Dermatomyositis, palmar hyperkeratosis, digital clubbing and hypertrophic osteoarthropathy, and palmar fasciitis and polyarthritis (PFA) all have been reported as cutaneous paraneoplastic syndromes associated with neoplasm.¹ The criteria used to establish the relationship between dermatosis and neoplasm include an unusual dermatosis, the concurrent occurrence of a

neoplasm, and the parallel clinical course of dermatosis and neoplasm.^{2,3}

A variant of PFA has been described recently. This variant was associated with pancreatic³ and ovarian⁴ carcinoma in 2 female patients. The main feature of these cases was hand involvement but in contrast to PFA the patients did not have arthritis or systemic rheumatologic symptoms.^{3,4}

We present an additional case of this variant: a man with a gastric carcinoma.

Case Report

A previously healthy 76-year-old man complained of increasing pain, hypersensitivity, and rapidly progressive loss of finger and thumb motion in both hands over the previous 7 months. He had pain at rest and it worsened at night, with no neurologic distribution. He had lost the ability to perform everyday activities because of finger contracture. The clinical presentation was asymmetric because the right hand was affected more. He had no history of illness and was taking no medication. On examination he had palmar subcutaneous nodules affecting the thenar and hypothenar eminence and also the dorsal aspect of the fingers and thumbs as in Dupuytren's disease, but he did not have pretendinous bands (Fig. 1). The palmar skin was thick-

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Received for publication July 23, 2004; accepted in revised form April 18, 2005.

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

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0363-5023/05/30A05-0032\$30.00/0
doi:10.1016/j.jhsa.2005.04.010

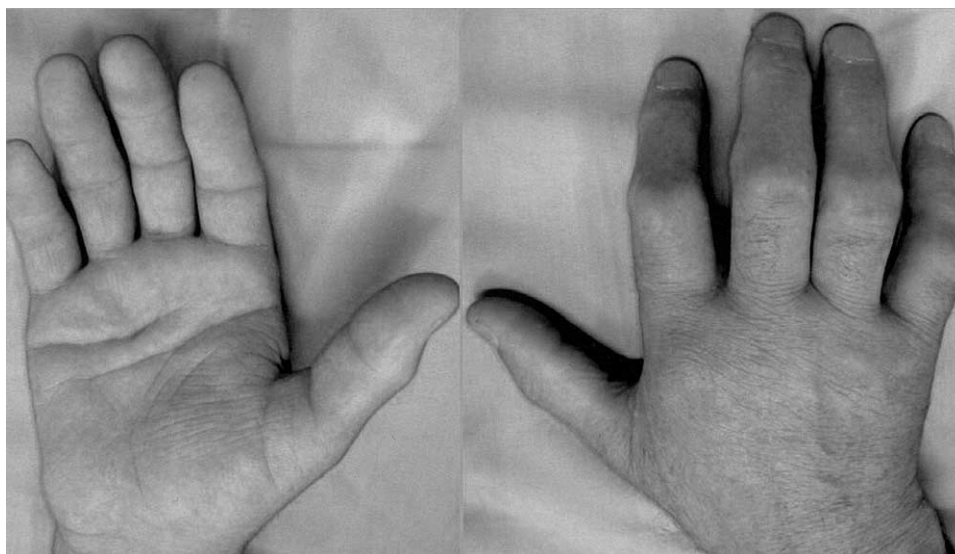


Figure 1. (A) Dorsal and (B) volar aspect of the right hand showing the nodules and the indurated swelling of the fingers and thumb.

ened and the fingers and thumbs were stiff and had induration and swelling, which prevented him from making a fist. The active range of motion of the digits was diminished (Table 1). The metacarpophalangeal joints and proximal interphalangeal joints had flexion contractures. The dorsal aspect of the fingers and thumbs had no skin creases (Fig. 2). The patient's pigmentation was normal and he had no telangiectases. Results of clinical and electrical tests for nerve compression were negative. He had not lost weight and general examination results were otherwise normal. A blood test showed no abnormal results.

Scleroderma and Dupuytren's disease were proposed as the most likely diagnoses.

A magnetic resonance image (MRI) showed nodular images localized in the subcutaneous tissue that did not affect the palmar aponeurosis (Fig. 3). A palmar subcutaneous nodule biopsy examination showed dense fibrous tissue in nodular arrangement with hypercellular areas (Fig. 4).

The clinical features worsened despite administration of nonsteroidal anti-inflammatory drugs and physical therapy. A course of colchicine was administered because of the suspected scleroderma.

Test results for antinuclear and anti-DNA (Scl-70) antibodies were negative. A barium esophagogram showed normal esophageal function. The patient had no history of Raynaud's phenomenon.

Full blood count and erythrocyte sedimentation rate test results were negative. Chest x-rays showed no abnormalities and x-rays of both hands showed mild osteoarthritis in the distal interphalangeal joints.

An atypical image was found in the stomach during the barium esophagogram. An esophagogastroscope showed an ulcerated lesion. Histologic examination showed an intestinal adenocarcinoma. Chest and abdominal tomography scans were negative for metastatic disease.

Nine months after the first consultation a laparotomy was performed to remove the neoplasm. During the

Table 1. Active Range of Motion of the Digits

	Index (deg)		Middle (deg)		Ring (deg)		Small (deg)	
	MCP	PIP	MCP	PIP	MCP	PIP	MCP	PIP
Right hand								
Extension	22	30	17	34	15	32	12	36
Flexion	40	42	43	46	42	43	39	44
Left hand								
Extension	12	8	15	10	14	11	9	12
Flexion	60	52	60	56	62	59	69	63

MCP, metacarpophalangeal joint; PIP, proximal interphalangeal joint.



Figure 2. Dorsal aspect of the fingers showing nodules without skin folds.

procedure another mass was found in the pelvic colon. Histologic examination showed a metastatic tumor from the stomach. Both tumors were removed. No adjuvant treatment was performed.

Two weeks after removal of both abdominal masses the skin changes of the hands stopped progressing. The

patient's condition improved over the next 4 months. Although recovery of the digital motion was incomplete the pain eased markedly. His condition has remained stable.

One year after surgery esophagogastrosocopy and colonoscopy showed no recurrence of gastric carcinoma or colonic metastases. Chest and abdominal computed tomography scans showed neither residual tumor nor metastatic disease. Digital motion improved, principally in the left hand where the symptoms were less severe. The current total range of motion of the digits is shown in Table 2.

Discussion

We report a man with a variant of PFA syndrome associated with gastric cancer. Ovarian carcinoma is the most frequent neoplasm related to PFA syndrome.⁵⁻⁸ Other tumors have, however, been associated with this paraneoplastic manifestation including pancreatic, lung, bladder, and breast carcinoma.^{9,10}

The clinical picture presented by our patient showed features that resemble those of palmar fibromatosis and scleroderma. The noncharacteristic localization of the

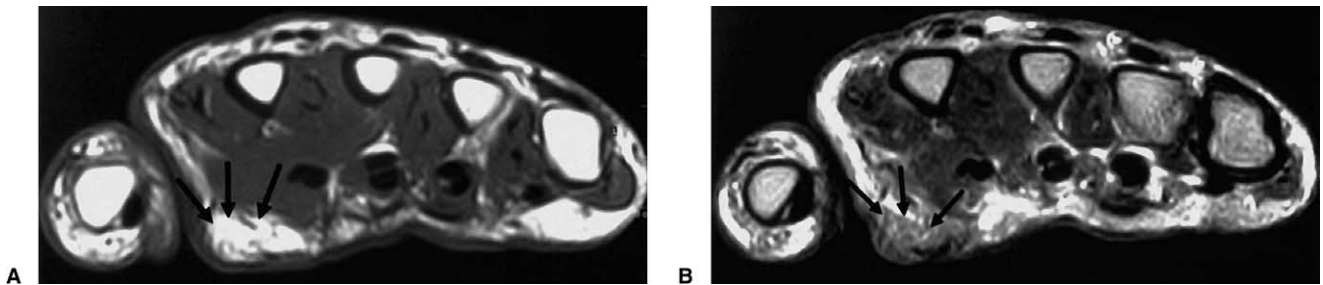


Figure 3. (A) Axial T1-weighted MRI of the right hand showing a nodular image of high signal intensity within the subcutaneous tissue of the palmar aspect (black arrows). (B) Axial fat-suppressed T2-weighted MRI showing the nodular image with low signal intensity (black arrows).

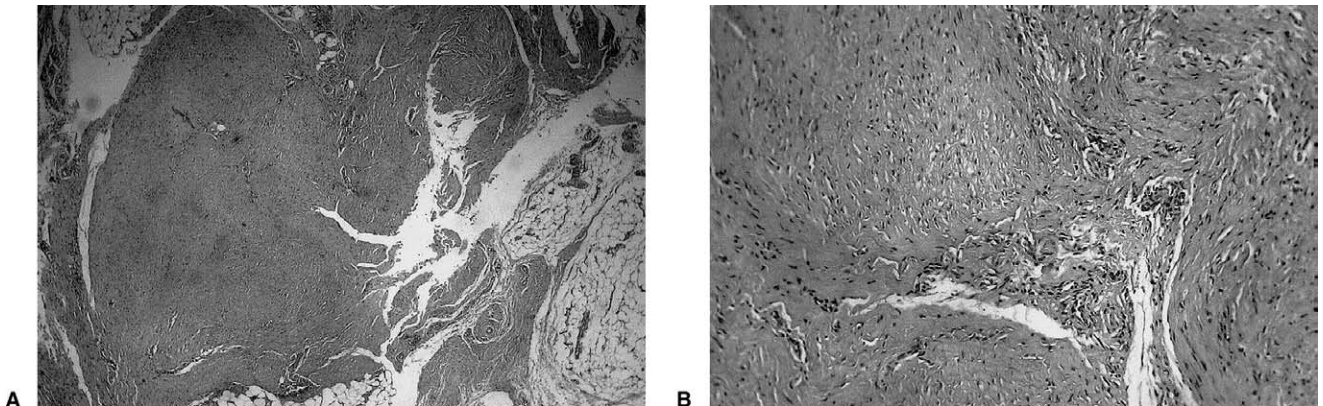


Figure 4. Sections showing hematoxylin- and eosin-stained tissue photomicrographs. (A) Section showing a nodule with uniform spindled cells separated by collagen (original magnification $\times 2.5$). (B) Parallel fascicles of slender cells separated by variable amounts of collagen (original magnification $\times 10$).

Table 2. Current Active Range of Motion of the Digits

	Index (deg)		Middle (deg)		Ring (deg)		Small (deg)	
	MCP	PIP	MCP	PIP	MCP	PIP	MCP	PIP
Right hand								
Extension	13	22	10	20	11	30	10	26
Flexion	72	75	85	71	92	82	90	77
Left hand								
Extension	10	5	7	3	4	2	5	2
Flexion	74	95	85	97	91	100	97	104

MCP, metacarpophalangeal joint; PIP, proximal interphalangeal joint.

nodules, the rapid progression of the clinical features of the hands, the lack of pretendinous bands, the stiffness and indurated swelling of the digits, and the noninvolvement of the palmar aponeurosis made Dupuytren's disease unlikely. Furthermore the absence of Raynaud's phenomenon, the lack of specific antibodies, and the normal esophageal function were atypical features for scleroderma. Conversely this unusual presentation, the lack of response to treatment, and the parallel course with the underlying cancer fulfilled the criteria for establishing the existence of paraneoplastic syndrome.

A variety of musculoskeletal syndromes have been associated with neoplasms.^{1,9} The pathogenesis is not known clearly although several immune and cytokine mediators, hormones, and growth factors may be associated with the tumor.^{2,5}

Palmar fasciitis and polyarthritis syndrome first was described by Medsger et al⁷ as associated with ovarian carcinoma. The clinical features of PFA resemble other conditions associated with contractures of the hands, principally scleroderma and palmar fasciitis (fibromatosis). Palmar fasciitis and polyarthritis, however, causes rapid progressive and disabling flexion contractures of both hands and inflammatory arthritis.^{6,9-11}

A variant of fasciitis-panniculitis syndrome was termed "woody hands" by Cox et al.³ They reported an elderly woman with pancreatic carcinoma, swelling, and deep induration of the hands but without involvement of the legs as is characteristic of panniculitis. In 2003 Alexandroff et al⁴ reported another case of woody hands in a woman with ovarian carcinoma. The main characteristic of these cases was the involvement of the hands without systemic rheumatic symptoms.

We have presented an additional case of a variant of palmar fasciitis without polyarthritis in a patient with gastric carcinoma. Suspicion of this association is important because it can lead to early diagnosis of the underlying cancer and its curative treatment. We rec-

ommend a thorough neoplasm workup including gastric carcinoma in any patient with atypical clinical features of Dupuytren's disease including sudden onset, digital contractures, and pain.

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