# Palmar fasciitis and polyarthritis as a paraneoplastic syndrome associated with tubal carcinoma: a case report

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73 year old white woman was admitted to our hospital in May 2003 complaining of lower abdominal pain for 1 month. On admission, the gynaecological examination disclosed a large, non-tender lower abdominal mass of 7 cm in diameter, which was highly suspicious of ovarian cancer with peritoneal infiltration in the computed tomography scan. In accordance with these findings, serum levels of CA 12-5 were about 3000 U/ml (normal range <35 U/ml).

Therefore, we performed an exploratory laparotomy. The frozen section of the adnexal mass showed a serous papillary carcinoma of fallopian tube origin. Consecutively, a complete staging with hysterectomy, bilateral salpingo-ophorectomy, omentectomy, and resection of the descending part of the colon due to tumour infiltration was performed.

Recovery was complicated owing to prolonged ileus-like symptoms. First line chemotherapy with topotecan in combination with carboplatin under study conditions (prospective, open label, phase II study) was started 3 weeks after the operation.

Before the diagnosis of carcinoma of the fallopian tube, the patient reported a progressive painful swelling and stiffness of both hands, especially the palms, which were noted by the patient to have thickened progressively since December 2002, with no improvement after initial corticosteroid treatment. Apart from generalised arthrosis, she denied any previous joint diseases, Raynaud's phenomenon, skin tightening, or trauma.

Physical examination on admission was remarkable for symmetrical swollen hands and fingers, painful on active and passive movement, and palmar fascial thickening with erythema (fig 1). The fingers of the patient showed flexion contractures; making a fist was impossible. No evidence of skin sclerosis or arthritis in other locations was found.

The patient's symptoms were orally treated with a cyclooxygenase(COX)-2 inhibitor, a transdermal delivery system for buprenorphine, and local ointments with diclofenac.

Beside an increased acute phase reaction (postoperatively), the immunological laboratory investigation failed to show any specific abnormalities (rheumatoid factor, antinuclear antibodies, antistreptolysin titre, antineutrophil cytoplasmic antibodies, complement CH<sub>50</sub> and C3d). Radiography of both hands showed a mild arthrosis in the joints, but no signs of acute arthritis.

After two cycles of antineoplastic treatment the patient's paraneoplastic symptoms currently show a good response: the fasciitis and acute arthritis have gradually improved, but the contractures in both hands persist despite extensive physiotherapy.

# DISCUSSION

Paraneoplastic syndromes affect a variety of organ systems and are often an initial sign of occult malignancies. About 30 cases have been reported describing palmar fasciitis and polyarthritis (PFPA)-like changes of the hands in association with malignant tumours. PFPA has been reported to be



Figure 1 Both hands of the patient with fingers in maximal extension, showing contractures and nodular palmar fascial thickening.

associated with different neoplasms—for example, ovarian cancer,<sup>1</sup> carcinoma of the breast,<sup>2</sup> carcinoma of the prostate,<sup>3</sup> and gastric tumours<sup>4 5</sup> preceding or accompanying the diagnosis of malignancy.

In our case we present a patient demonstrating a severe progressive deforming rheumatic disease affecting the palms and fingers of both hands, with the onset of the corresponding symptoms 5 months before the diagnosis of a malignant neoplasm was made.

To our knowledge this is the first report of the association of PFPA as a paraneoplastic syndrome with a carcinoma of the fallopian tube.

The underlying immunological mechanisms have not been defined so far and may result from activation of certain factors with profibrotic activities—for example, transforming growth factor  $\beta$  or connective tissue growth factor.<sup>6</sup> As long as the pathomechanisms of PFPA are unclear, the only effective therapy is cancer treatment, which may lead to arrest or even complete resolution of the rheumatic disease.<sup>7</sup>

The characteristic hand deformities of PFPA, recently described with the illustrative term of woody hands,<sup>7</sup> should alert the clinician to search for an underlying malignant disease.

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# Mycobacterium chelonae finger infection associated with Raynaud's phenomenon

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Maximum chelonae infections are uncommon and have not been reported as a complication of Raynaud's phenomenon previously. We describe a patient with this problem who responded well to treatment.

## **CASE REPORT**

A 21 year old female telephonist was referred with a 1 week history of swollen right index and middle fingers. There was no history of trauma but she had had Raynaud's phenomenon since the age of 12. There was no history of recent travel abroad or of unusual hobbies.

On examination she was apyrexial, she had diffuse swelling but no discolouration of the right index and middle fingers. She had no blisters or ulceration and the fingers were not tender to touch. Nine days later, despite flucloxacillin orally and a non-steroidal anti-inflammatory agent, all her fingers were cold and bluish, and the right index and right middle fingers remained diffusely swollen. She was admitted for intravenous prostacyclin because of worsening of her Raynaud's phenomenon.

Her baseline investigations were normal, including erythrocyte sedimentation rate and C reactive protein. A hand xray examination showed no bony abnormality. Blood cultures were negative. Her immunoglobulin levels were normal, and cryoglobulins were not detected. Her autoantibody profile was negative except for raised IgG cardiolipin antibodies at 46 GPLU (reference range 0–13).

As she finished the 72 hour intravenous prostacyclin infusion, a pustule appeared on the pulp of the right index finger, and intravenous ceftriaxone was started. Two days later a second pustule appeared on the other finger (fig 1). Microscopy of a needle aspiration showed acid fast bacilli. Antibiotics were changed to clarithromycin 500 mg twice daily and ciprofloxacin 750 mg twice daily. Both lesions were incised and drained, and histology of the material showed an abscess wall with no granulomata.

*M chelonae* was cultured from pus and was susceptible in vitro to azithromycin and clarithromycin, but resistant to other antituberculous agents, as well as to ciprofloxacin. As her lesions were healing at the time these results became available (14 days later) her antibiotics were not changed.



Figure 1 Pustules on the index and middle fingers of the right hand.

She continued the combination for a total of 5 weeks and her fingers healed completely.

#### DISCUSSION

*M* chelonae is associated with a variety of infections. Cutaneous lesions occur secondary to wound infections after surgery, accidental trauma, or needle injections.<sup>1–2</sup>*M* chelonae isolates are susceptible in vitro to clarithromycin, but generally resistant to ciprofloxacin. The use of at least two drugs (one of which should be clarithromycin) is recommended for treatment of *M* chelonae infection, as the emergence of resistance is a risk associated with monotherapy.<sup>3–4</sup> Treatment is usually given for 4–6 months, but our patient responded to combination therapy within weeks. A case has been reported of postoperative infection at a donor vein graft site that healed after 2 months of clarithromycin treatment, combined with heat treatment.<sup>5</sup> Thus shorter courses of treatment may be appropriate in non-immuno-suppressed patients.

Isolates of *M* chelonae have optimal growth at  $28-30^{\circ}$ C,<sup>6</sup> unlike the standard 35°C for most organisms. Our patient had severe Raynaud's disease before the infection, so the coldness of her hands may have promoted the growth of this unusual organism. We are not aware of any previous reports of