Case Report

Severe Relapsing Polychondritis Occurring After Ear Piercing

JACQUES SERRATRICE, NICOLETA ENÉ, BRIGITTE GRANEL, PATRICK DISDIER, CHRISTINE DE ROUX-SERRATRICE, LAURE SWIADER, GUY MOULIN, ANTOINE GIOVANNI, and PIERRE JEAN WEILLER

ABSTRACT. We describe a case of relapsing polychondritis with laryngo-tracheal involvement, occurring after ear piercing in a 39-year-old woman. Polychondritis was clearly time-related to ear piercing. This association draws attention to the risk of relapsing polychondritis during body art practices with cartilage trauma. (J Rheumatol 2003;30:2716–7)

Key Indexing Terms:
RELAPSING POLYCHONDRITIS CARTILAGE TRAUMA EAR PIERCING

Ear piercing of the upper third of the pinna has become a fashionable practice in body art. We report a single case of severe and generalized relapsing polychondritis (RP) in a 39-year-old woman after a high ear piercing.

CASE REPORT

A 39-year-old woman consulted our service in February 2002 for severe RP. Clinical history started in July 1998, during a first pregnancy, when she underwent a piercing in the upper third of the pinna of the right ear with a stainless steel ring in a jewelry shop. The hole in the cartilage remained painful despite local treatment. In August 1998, she changed the ring for a silver one. She kept her new ring for 4 weeks and then removed it because of persistent inflammation of the ear pinna. Topical antiseptics had no long-lasting results. In April 1999, an ear chondritis with local *Staphylococcus epidermidis* infection was diagnosed. She received pristinamycine with no improvement. At that time she gave birth to a normal boy; however, she complained of rib pain. The chondritis of the right ear was followed by generalized chondritis involving the nose, ribs, and respiratory tract. A diagnosis of RP was finally made. Treatment with dapsone was not effective and long-standing steroid therapy (prednisone 1 mg/kg/day at onset followed by slow tapering) was maintained with a mild effect. She was first admitted to our service in February 2002.

She had had silicone breast implants for augmentation mammoplasty 6 years before, and underwent esthetic surgery for "riding breeches" in 2001. She had not experienced any chondritis or rheumatologic symptoms before the ear piercing. She complained of inspiratory dyspnea with wheezing, hoarseness, and weakness of the voice. She presented with some aspirations. Her nose and ears were inflamed. Cartilage of the auricle of her right ear was destroyed, with collapse of the concha and cauliflower-ear aspect (Figure 1). Purpuric macules were present on the low neckline. Palpation of the ribs was painful and palpation of thyroidal cartilage was followed by coughing. Otherwise, clinical examination was normal. No aphthous lesions were found in oral or genital mucosa.

Laboratory findings were as follows: hemoglobin 10.9 g/dl, white blood cell count 10×10^9/l with 80% neutrophils, erythrocyte sedimentation rate 42 mm/h, C-reactive protein 22 mg/l, and serum proteins immunoelectrophoresis showed a polyclonal increase of IgM and IgG. There was no proteinuria. Serological tests for human immunodeficiency virus and hepatitis B and C virus were negative. Antinuclear antibodies, anti-DNA antibodies, and antineutrophil cytoplasmic antibodies were all negative. Human anti-type II collagen antibodies were positive: (4.05 U/ml by ELISA; normal < 1). HLA phenotyping was A03 A26, B07 B37, DRB1*15, DRB1*.

Laryngotracheal endoscopy showed an inflammatory process of the arytenoidal part of the subglottic larynx. Larynx and tracheal computerized tomography (CT) scan showed chondritis of thyroid and cricoid cartilages and inflammatory involvement of the first tracheal ring. Thoraco-abdomino-pelvic CT scans and echocardiography were otherwise normal.

Faced with this typical RP, which had started near the location of an ear

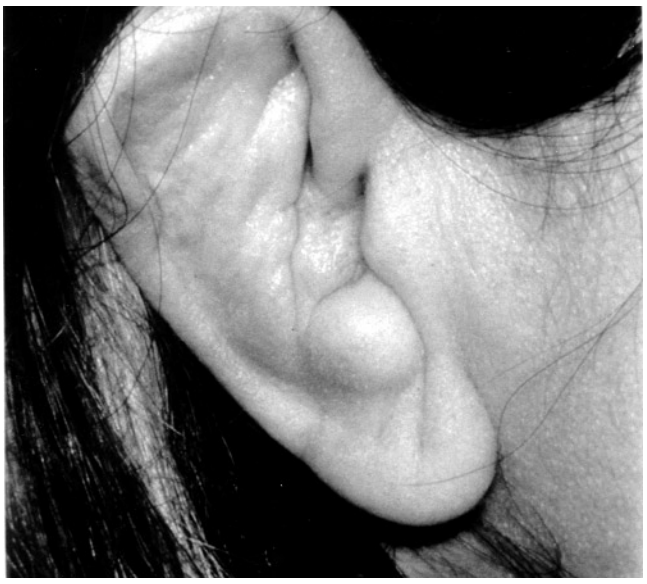

Figure 1. Cauliflower-ear after destruction of the cartilage of the right ear. Collapse of the concha of right auricle.

From the Service de Médecine Interne, the Fédération d'Oto-Rhino-Laryngologie, and the Service de Radiologie, CHU Timone, Marseille, France.

J. Serratrice, MD; N. Ené, MD; B. Granel, MD; P. Disdier, MD; C. deRoux-Serratrice, MD; L. Swiader, MD; G. Moulin, MD; A. Giovanni, MD; P.J. Weiller, MD.

Address reprints requests to Dr. P. Disdier, CHU Timone, 264 rue Saint-Pierre, 13385 Marseille Cedex 5, France. E-mail: patrick.disdier@ap-hm.fr

Submitted January 22, 2003; revision accepted May 28, 2003.

piercing, we decided to initiate steroid therapy (prednisone 1 mg/kg/day) with methotrexate (12 mg/wk). After 12 months of followup, with slow tapering of steroids (prednisone 1/4 mg/kg), inflammation was mildly reduced.

DISCUSSION

According to Michet, *et al*, our patient fulfilled diagnostic criteria for RP[1] and presented autoimmunization against cartilage-specific protein, as attested by the presence of anti-type II collagen antibodies[2-4].

Although we do not exclude a fortuitous association, we feel that in our patient RP was induced by ear piercing for the following reasons. First, ear piercing and RP were strongly time-related, as the patient experienced onset of RP subsequently to piercing with secondary generalization to all other cartilages. A case with very similar clinical history was recently reported by Alissa, *et al*[5]. Interestingly, our patient was pregnant before and during the onset of RP. Pregnancy is usually not considered to modify the course of RP[6]; however, a gravidic form of RP has been reported[7]. According to the above data, we believe that in our patient, hormonal and immunological modifications due to pregnancy might have played an additive role in the onset of RP. Second, an animal model has shown that unilateral implantation of metal ear studs in rats is frequently followed by ipsilateral and then bilateral, pinally-restricted chondritis characterized by multifocal granulomatous chondritis, progressive destruction of cartilaginous plates, and IgG deposits in the cartilaginous matrix[8]. The author concluded that the chondritis observed in this animal model was due to an autoimmune response initiated by a chronic inflammatory process at the insertion site[8].

In our patient, puncture and presence of foreign material in the cartilage could have triggered an autoimmune disorder and facilitated anticollagen antibody expression by exposing unusual epitopes of cartilage matrix protein. As shown by Rogero, *et al*, commercial studs left in contact with body fluids can trigger inflammatory reactions, especially those with low corrosion resistance and high cytotoxicity[9]. According to immunologic theory, metal ions are too small to provoke an immune response, but they may be immunogenic after their conjugation with protein carriers, and the metal part may be an antigen determinant[10]. In our patient, the disease may have been triggered by the ear ring.

Third, in our patient, a staphylococcal infection was documented at the site of insertion, but antibiotics were unable to stop chondritis evolution and extension. Chondritis and perichondritis of infectious origin are the most common complications of ear piercing. This condition is usually induced by incorrect technique as reported by More, *et al*[11]. Medical complications include bleeding, tissue trauma, loco-regional bacterial infection[12], and finally the endstage cauliflower-ear. In our patient, an infectious process may have facilitated the autoimmune reaction, as described in various systemic disorders such as Wegener's granulomatosis[13].

Thus we feel that in our patient, RP was induced by cartilage piercing as a result of initial cartilage trauma and infection, antigen exposure to immune surveillance, and autoimmune response facilitated by presence of foreign material.

REFERENCES

1. Michet CJ Jr, McKenna CH, Luthra HS, et al. Relapsing polychondritis survival and predictive role of early disease manifestations. Ann Intern Med 1986;104:74-8.
2. Matsumoto Y, Imanaga T, Kawajiri T, et al. Measurement of anti-type II collagen antibody diagnosis and follow-up useful in a case of relapsing polychondritis. Nihon Kokyuki Gakkai Zasshi 2002;40:45-9.
3. Vinceneux P, Pouchon J, Piette JC. Polychondrite atrophiante. In: Khan MF, Peltier AP, Meyer O, Piette JC, editors. Maladies et syndromes systémiques. 3rd ed. Paris: Flammarion; 2000:623-49.
4. Hansson AS, Heinegard D, Piette JC, Burkhardt H, Holmdahl R. The occurrence of autoantibodies to matrilin 1 reflects a tissue-specific response to cartilage of the respiratory tract in patients with relapsing polychondritis Arthritis Rheum 2001;44:2402-12.
5. Alissa H, Kadanoff R, Adams E. Does mechanical insult to cartilage trigger relapsing polychondritis? [letter]. Scand J Rheumatol 2001;30:311.
6. Papo T, Wechsler B, Bletry O, Piette AM, Godeau P, Piette JC. Pregnancy in relapsing polychondritis: twenty-five pregnancies in eleven patients. Arthritis Rheum 1997;40:1245-9.
7. Bellamy N, Dewar CL. Relapsing polychondritis in pregnancy. J Rheumatol 1990;17:1525-6.
8. Meingassner JG. Sympathetic auricular chondritis in rats: a model of autoimmune disease? Lab Anim 1991;25:68-78.
9. Rogero SO, Higa OZ, Saiki M, Correa OV, Costa I. Cytotoxicity due to corrosion of ear piercing studs. Toxicol In Vitro 2000;14:497-504.
10. Golub ES, Green DR. The nature of antigens. In: Immunology, a synthesis. 2nd ed. Sunderland, MA: Sinauer; 1992:21-41.
11. More DR, Seidel JS, Bryan PA. Ear-piercing techniques as a cause of auricular chondritis. Pediatr Emerg Care 1999;15:189-92.
12. Mayers LB, Judelson DA, Moriarty BW, Rundell KW. Prevalence of body art (body piercing and tattooing) in university undergraduates and incidence of medical complications. Mayo Clin Proc 2002;77:29-34.
13. Boudewyns A, Verbelen J, Koekelkoren E, Van Offel J, Van de Heyning P. Wegener's granulomatosis triggered by infection? Acta Otorhinolaryngol Belg 2001;55:57-63.