Myositis Ossificans in the Newborn

A Case Report*

BY U. E. PAZZAGLIA, M.D.†, G. BELUFFI, M.D.‡, A. COLOMBO, M.D.†, A. MARCHI, M.D.†, A. COCI, M.D.†, AND L. CECILIANI, M.D.†, PAVIA, ITALY

From the Orthopaedic Clinic, University of Pavia, Pavia

The youngest reported patients with myositis ossificans apparently were two six-year-old boys[6,7]. Therefore, the case we are reporting is the first observation of myositis ossificans in a neonate.

Case Report

A girl born on August 5, 1983, weighed 3550 grams and was apparently normal, but at the age of forty days the mother observed a painful swelling of the lateral aspect of the left leg. No history of trauma to the left leg was recorded.

At the age of fifty-two days the baby was first examined by us. There was a painful swelling on the lateral aspect of the leg, with normal overlying skin and limited plantar flexion of the foot. The following laboratory tests were performed, with results within the normal range: hemogram, sedimentation rate, creatine phosphokinase, acid and alkaline phosphatases, plasma protein, and electrophoresis. The levels of immunoglobulins, transaminases, lactic acid dehydrogenase, cholinesterase, plasma calcium and phosphate, and parathyroid hormone were also within normal limits. Cultures of blood and urine were negative, as was the serology.

Radiographs showed some small radiopaque granules between the tibia and fibula (Fig. 1, *a*), and a bone scintigram showed hypercaptation in the region of the swelling.

A biopsy was done. Histological examination of the specimen showed hypercellular, immature collagenous tissue. There were areas of osteoid with many osteoblasts and foci of chondroid differentiation. Some calcified areas showed resorptive osteoclasts. The specimen had a multifocal pattern with a mixture of foci of poorly organized and partially ossified fibrous tissue, and a zonal arrangement was barely distinguishable.

The swelling increased, as did the volume of the multiple calcified masses (Fig. 1, *b*), and broadening and deformity of bones of the leg became increasingly evident as the masses grew (Fig. 1, *c*). Because an irreducible calcaneus deformity of the foot was developing, the anterolateral compartment of the leg was explored on December 1, 1983. A hard mass that involved the entire tibialis anterior and compressed the extensor hallucis longus and the extensor digitorum longus and the interosseous membrane was excised.

Manipulation and treatment with a plaster cast were started soon after the operation and were maintained until the child began to walk, at the age of fourteen months.

At the eighteen-month follow-up examination, the child walked well. The bones of the leg had undergone extensive remodeling, and radiographs showed almost normal morphology (Fig. 1, *e*). At the time of writing, the tibiocalcaneal angle was normal. Passive dorsiflexion was decreased to about half of normal; active dorsiflexion was present, but was weaker than that of the other leg.

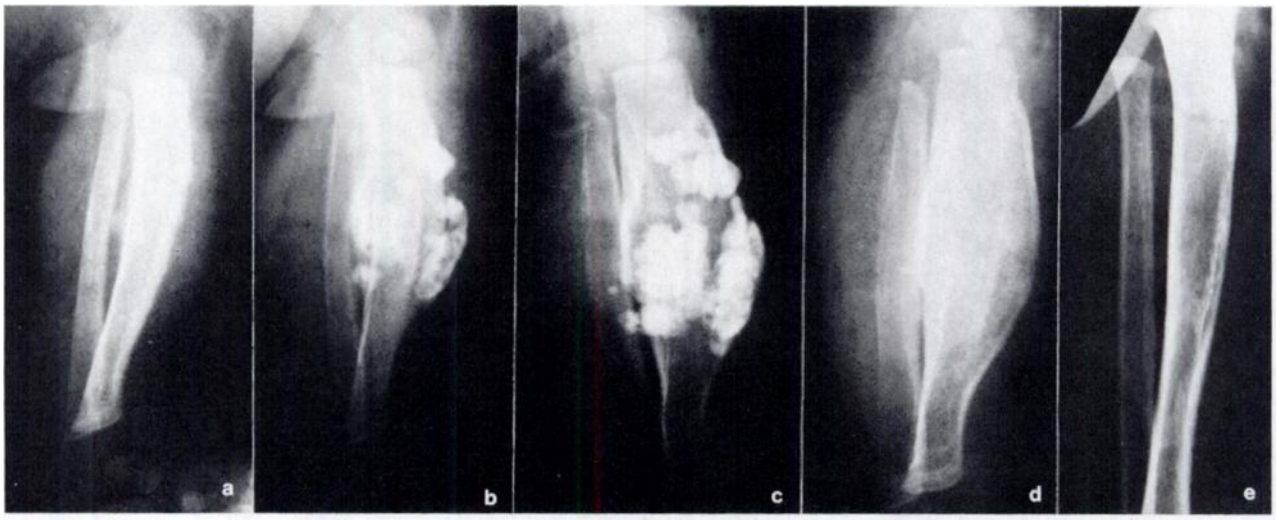

FIG. 1

Radiographs showing evolution of the lesion. *a*: At the age of fifty-two days, small radiopaque granules are observed between the tibia and the fibula. *b*: At the age of seventy-four days, the volume of calcified masses is increased and periosteal apposition is also evident. *c*: At the age of three months and nineteen days, calcified masses are further increased. The tibia and fibula are divaricated. *d*: At the age of three months and twenty-eight days, after the mass was removed, the wide periosteal apposition is evident on both the tibia and the fibula. *e*: At follow-up, when the patient was fifteen months old, remodeling of the leg bones is almost complete.

* This article was accepted for publication prior to July 1, 1985. No conflict-of-interest statement was requested from the authors.

† University of Pavia, Via Taramelli, I-27100 Pavia, Italy. Please address reprint requests to Dr. Pazzaglia, Orthopaedic Clinic.

‡ IRCCS Ospedale San Matteo, Pavia, Italy.

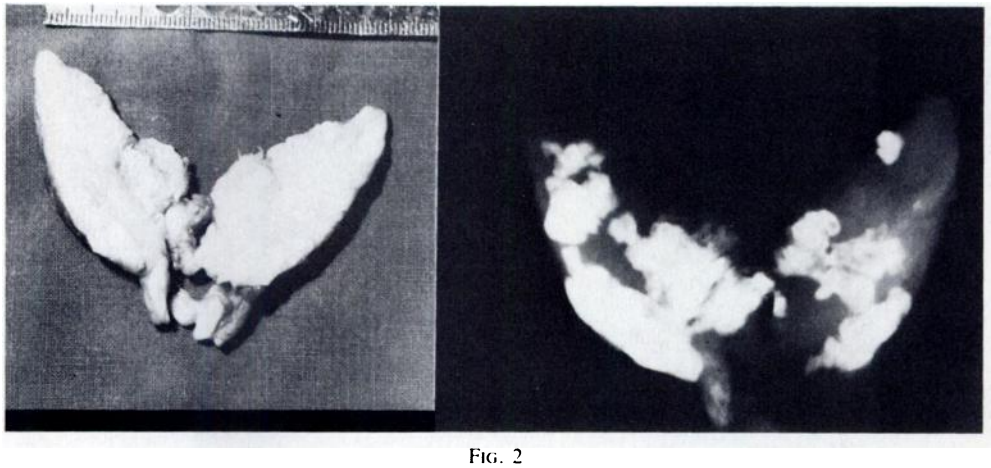

FIG. 2

The excised mass is formed by a fibrotic tibialis anterior.

Histological Findings

The whole tibialis anterior was fibrotic (Fig. 2), and only scattered muscle fibers were present in the mass of fibrous tissue. The zone phenomenon, with definite maturation at the periphery, was observed, although it was not as orderly as has been evident in other patients[1,5,10].

Multiple foci of cellular proliferation and masses in different phases of maturation were seen. The masses were well delimited by the surrounding fibrous tissue only when they had the definitive aspect of an ossicle (Fig. 3, *B*) or when they were on the border of the mature zone. No clear demarcation between the proliferating zone and the adjacent fibrous tissue was observable.

The proliferating zone was characterized by a very cellular tissue with a scanty intercellular matrix. The spindle-shaped cells showed some nuclear pleomorphism and occasional mitoses. Polymorphonuclear giant cells were observed exclusively in this zone. Neither hemosiderin nor inflammatory cells were present. Around the proliferating zone, the same spindle-shaped cells underwent modulation toward osteoblastic or chondroblastic activity. Chondroid tissue was well represented, but it never assumed the features of mature cartilage.

Calcium deposition was observed on both the osteoid and the chondroid tissue of the intermediate zone. Remodeling of the calcified primary bone and chondroid tissue led to mature cancellous bone of the outer zone.

Discussion

The age of the patient and the course of the lesion are unusual, but the pathological findings fit with the diagnosis of myositis ossificans[2-5,8,11,12]. The peculiarities of myositis ossificans in our patient were the onset soon after birth; the multifocal cellular proliferation, leading to multiple masses in different phases of maturation; the rapid proliferation of the mass; the remodeling of the tibia and fibula around the mass; and the complete fibrosis of the tibialis anterior. In our patient, possible trauma cannot be excluded as an etiological mechanism, although there was no history of trouble with delivery, and no signs of hemorrhage (such as iron pigment) were observed in the tissue.

A role for the periosteum has been advanced often in theories of the pathogenesis of myositis ossificans, and much radiographic evidence of periosteal involvement has been presented[1,9,10]. In our patient there was a wide zone of periosteal bone deposition on both the tibia and the fibula that involved the entire diaphysis. The pathological calcified tissue was more radiopaque than the reactive periosteal bone. At no time was there continuity between that bone and the pathological ossification within the mass of the lesion. Reactive bone had grown so extensively around the mass, however, that when it was removed large cavities

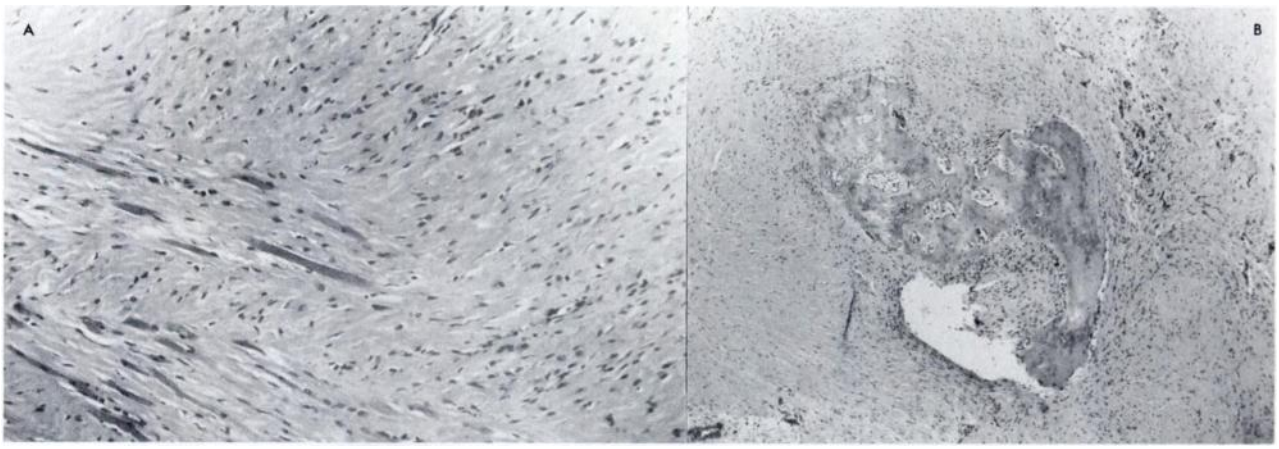

FIG. 3

A, Few muscle fibers are observed inside the fibrotic tibialis anterior (hematoxylin and eosin, × 100). *B*, Remodeling of the ossifying masses led to formation of an ossicle inside the fibrotic muscle (hematoxylin and eosin, × 40).

were left in both the tibia and the fibula.

The rapid remodeling of the bones adjacent to the mass may be accounted for by the extreme youth of the patient, while the extensive fibrosis may have been due to the anatomical location of the lesion in the inextensible anterolateral compartment of the leg. The anatomical site of the lesion is atypical for myositis ossificans, and that localization for this lesion has never been reported before, to our knowledge.

NOTE: The authors thank Dr. P. D. Byers and Prof. J. A. Kirkpatrick for the useful discussion of the case and Dr. P. Fiori and Mr. Cassinari for their help during the follow-up and preparation of the illustrations.

References

1. ACKERMAN, L. V.: Extra-Osseous Localized Non-Neoplastic Bone and Cartilage Formation (So-Called Myositis Ossificans). Clinical and Pathological Confusion with Malignant Neoplasms. J. Bone and Joint Surg., **40-A:** 279-298, April 1959.
2. ANGERVALL, L.; STENER, B.; and ÅHRÉN, C.: Pseudomalignant Osseous Tumour of Soft Tissue. A Clinical, Radiological and Pathological Study of Five Cases. J. Bone and Joint Surg., **51-B(4):** 654-663, 1969.
3. CAULET, T.; ADNET, J. J.; PLUOT, M.; GOUGEON, J.; and HOPFNER, C.: Myosite ossifiante circonscrite. Étude histochimique et ultrastructurale d'une observation. Virchows Arch. pathol. Anat., **348:** 16-35, 1969.
4. FINE, GERALD, and STOUT, A. P.: Osteogenic Sarcoma of Extraskeletal Soft Tissues. Cancer, **9:** 1027-1043, 1956.
5. JEFFREYS, T. E., and STILES, P. J.: Pseudomalignant Osseous Tumour of Soft Tissue. J. Bone and Joint Surg., **48-B(3):** 488-492, 1966.
6. JOHNSON, J. T. H.: Atypical Myositis Ossificans. J. Bone and Joint Surg., **39-A:** 189-194, Jan. 1957.
7. LAGIER, R., and COX, J. N.: Pseudomalignant Myositis Ossificans. A Pathological Study of Eight Cases. Hum. Pathol., **6:** 653-665, 1975.
8. MAINI, P. S., and SINGH, M.: Localized Myositis Ossificans Progressiva. A Case Report. J. Bone and Joint Surg., **49-A:** 955-960, July 1967.
9. NORMAN, ALEX, and DORFMAN, H. D.: Juxtacortical Circumscribed Myositis Ossificans: Evolution and Radiographic Features. Radiology, **96:** 301-306, 1970.
10. OGILVIE-HARRIS, D. J., and FORNASIER, V. L.: Pseudomalignant Myositis Ossificans: Heterotopic New-Bone Formation without a History of Trauma. J. Bone and Joint Surg., **62-A:** 1274-1283, Dec. 1980.
11. PATERSON, D. C.: Myositis Ossificans Circumscripta. Report of Four Cases without History of Injury. J. Bone and Joint Surg., **52-B(2):** 296-301, 1970.
12. VALENTIN, P.; GODENÈCHE, J.-L.; PIOLLET, J.; and DE LAGUILLAUMIE, B.: Tumeur osseuse des parties molles dite "pseudo maligne". Rev. chir. orthop., **55:** 351-357, 1969.