Intraosseous Glomus Tumor in the Ulna

A Case Report

LEO M. ROZMARYN, M.D.,* ARTHUR H. SADLER, M.D.,**
AND HOWARD D. DORFMAN, M.D.†

Glomus tumors are benign vascular tumors composed of round to oval uniform cells associated with vascular structures, usually found in the soft tissue of the nail bend. A 24-year-old woman with a two-year history of progressive right elbow pain and swelling was found to have a histologically characteristic intraosseous glomus tumor in this unusual location. Excision was followed by complete relief of symptoms.

Glomus tumors are benign vascular tumors composed of round to oval uniform cells associated with vascular structures. The cell of origin may be derived from the neuromyoarterial glomus body. Usually a soft tissue lesion, commonly found adjacent to the distal phalanx, it occasionally erodes into bone.[2,8] Glomus tumors rarely arise primarily within bone.[2,10] The following report describes a patient with an intraosseous glomus tumor in a location that is heretofore unreported.

From Montefiore Medical Center and Albert Einstein College of Medicine, Bronx, New York.

* Chief Resident in Orthopaedics at Montefiore Medical Center and Albert Einstein College of Medicine.

** Assistant Professor of Orthopaedic Surgery at Montefiore Medical Center and Albert Einstein College of Medicine; Director of Orthopaedic Surgery, Bronx Municipal Hospital Center, Bronx, New York.

† Professor of Orthopaedic Surgery, Pathology and Radiology at the Albert Einstein College of Medicine; Head, Section of Orthopaedic Pathology, Department of Orthopaedic Surgery, Montefiore Medical Center.

Reprint requests to Howard D. Dorfman, M.D., Section of Orthopaedic Pathology, Department of Orthopaedic Surgery, Montefiore Medical Center, 111 East 210 St., Bronx, NY 10467.

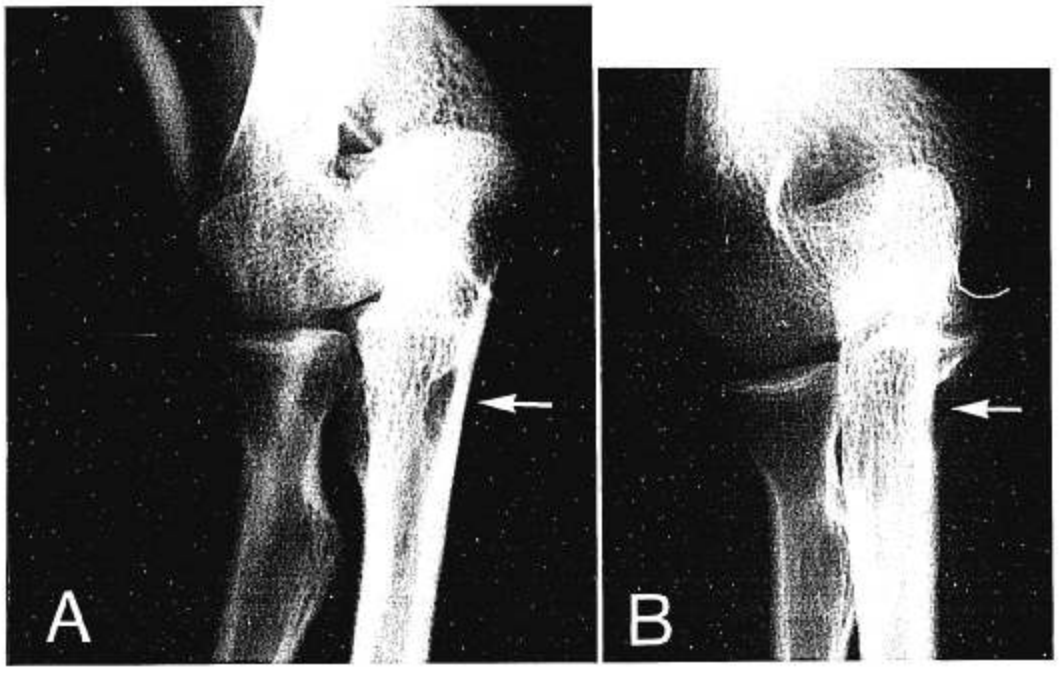
Received: February 21, 1986.

CASE REPORT

A 24-year-old woman had a two-year history of progressive right elbow pain and a soft tissue mass progressively increasing in size along the medial aspect of the proximal forearm. The mass was extremely tender to the touch and sensitive to changes in temperature. The onset of pain was sudden, without any antecedent trauma, and the pain was constant and localized to a small area on the medial aspect of the forearm just distal to the elbow. There was no limitation of movement of her elbow, wrist, or forearm. The pain did not awaken the patient from sleep. There was no swelling or erythema, but the mass became so tender that the patient became preoccupied with the protection of her elbow from any contact. There were no constitutional complaints, such as fever, chills, or weight loss. There were no abnormal laboratory findings, and the serum calcium and phosphorus were within normal limits. The pain did not respond to salicylates or oral narcotic analgesics.

Roentgenographic examination of the right elbow revealed an area of bone destruction in the anteromedial area of the proximal ulna, just distal to the coronoid process (Figs. 1A and 1B). The lesion was radiolucent and measured 1.0 cm in length and 3.0–4.0 mm in width. Its location was within the medial cortex, and there was no sclerotic border. A bone scan with 15 mc Technetium (New England Nuclear, Billerica, MA) revealed mild diffuse uptake in the right elbow. Angiography revealed a fine vascular blush, which ballooned out medially from the medial surface of the ulna.

The patient was surgically treated. A curved incision was made along the medial aspect of the subcutaneous border of the proximal ulna, beginning at the tip of the olecranon and progressing distally for 4 cm. Immediately deep to the flexor carpi ulnaris a mass of tan and focally hemmorrhagic tissue, which was soft and friable, was encountered lying



FIGS. 1A AND 1B. (A) Roentgenogram of right elbow showing lytic defect in the anteromedial portion of ulna distal to the coronoid process. (B) Eccentric radiolucent defect limited by a thin cortical shell in the proximal ulna at the site of maximal point tenderness.

within a cavity in the cortical surface of the ulna, measuring 1.2 × 0.5 × 0.5 cm. The lesion had eroded through a 0.3-cm opening in the bony cortex into the surrounding soft tissues. The lesion was curretted to the cancellous bone. The postoperative course was unremarkable. The patient was entirely without pain and there was no swelling or tenderness about the forearm. At a follow-up evaluation three months later, the wound was fully healed and the patient was asymptomatic.

Light microscopy revealed that the tumor consisted of sheets of uniform polygonal to round cells with distinct cell boundaries. The nuclei were round or oval and vesicular, with finely dispersed chromatin. Nucleoli were not prominent. Some nuclei showed a round eosinophilic intranuclear inclusion almost filling the nucleus. The cytoplasm was finely granular, and some cells showed clear vacuoles. Mitoses were extremely rare. There was a fine reticulin framework throughout the lesion, and in many areas the reticulin fibers surrounded individual cells. A few endothelial-lined vascular channels were found within the tumor (Fig. 2).

Desmin and actin were demonstrated in the tumor cells with the use of immunoperoxidase technique. This observation confirms the close relationship between the tumor cells and smooth muscle cells.

DISCUSSION

The glomus body is a specialized form of arterial venous anastomosis composed of neural, smooth muscle, and vascular elements. It is usually found in the dermal–subdermal junction in the subungual region of the fingers and may be involved in distal temperature regulation.[3] The glomus tumor is a distinctive type of vascular neoplasm whose cell type is a modified smooth muscle cell closely resembling the glomus body from which it is derived.

The glomus tumor was first described in 1924, by Masson.[7] Since then, sporadic reports of glomus tumors of varying types and locations have appeared in the literature. Glomus tumors located within bone are distinctly rare.

The earliest report of a glomus tumor within bone was by De Latorre,[4] who in 1939 re-

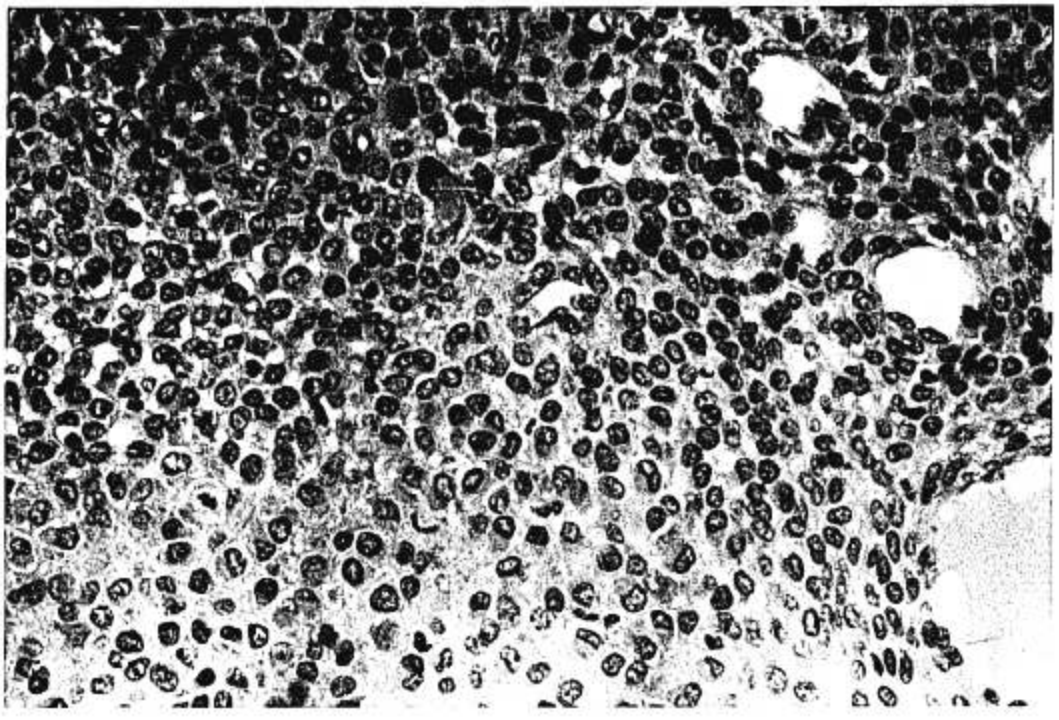


FIG. 2. Photomicrograph of the intraosseous glomus tumor showing sheets of plump spindle cells with endothelial-lined vascular spaces. Note the uniform nuclei and epitheloid appearance of the glomus cells. (Hematoxylin and eosin; original magnification, ×320)

ported a glomus tumor in the distal phalanx of the thumb, which was treated by an interphalangeal disarticulation. In a report of a case of an intraosseous glomus tumor by Lattes and Bull,[6] the possible histogenesis of this tumor was discussed. Tissue culture work by Murray and Stout[9] indicated that glomus tumor cells are modified smooth muscle cells in close contact with endothelial cells and unmyelinated neural fibers. Their work also indicated that these cells arose from multipotential mesenchymal stem cells similar to those that give rise to pericytes of Zimmerman. It is this proximity to the rich neural bed that is believed to be responsible for the high sensitivity to pain and temperature change.[1]

Until 1981, only nine cases of intraosseous glomus tumors had been reported in the literature.[10] Jaffe[5] reported that a normal glomus body exists in the medullary cavity of bone and that a primary glomus tumor of the bone could be expected to occur. All cases described to date, however, have occurred in the terminal phalanges.

Osteoid osteoma may have a long progressive history of pain, tenderness, and swelling with a radiolucent defect in the cortex, and it is usually eccentric. However, osteoid osteoma causes a vigorous sclerotic reaction around the lesion, and the pain is often dramatically relieved with the use of salicyclates.

Characteristically, resection of the mass yields complete relief of pain, tenderness, and temperature sensitivity. Although these lesions may be locally erosive, as in the case presented herein, they are solitary; metastatic spread and

malignant degeneration are unknown. The rare glomangiosarcoma is a *de novo* lesion and requires a histologic diagnosis, because its clinical behavior mirrors the benign glomus tumor.

This case represents the first reported instance of an intraosseous glomus tumor of the ulna. The origin of the tumor in soft tissue with erosion into the bone must be considered but is unlikely in view of the roentgenographic and operative findings.

REFERENCES

1. Chan, C. W.: Intraosseous glomus tumor. A case report. J. Hand Surg. 6:368, 1981.
2. Dorfman, H. D., Steiner, G. C., and Jaffe, H. L.: Vascular tumors of bone. Hum. Pathol. 2:349, 1971.
3. Enzinger, F. M., Weiss, S. W.: Glomus Tumors. Soft Tissue Tumors. St. Louis, C. V. Mosby, 1985, pp. 450–462.
4. De Latorre, I., Gomez Camajo, M., and Palacios, G.: Consideraciones clinicas anatomicas. Cir. Ortop. Traum. 7:11, 1939.
5. Jaffe, H. L.: Tumors and Tumorlike Conditions of Bones and Joints. Philadelphia, Lea and Febiger, 1958, p. 254.
6. Lattes, R., and Bull, D. C.: A case of glomus tumor with primary involvement of bone. Ann. Surg. 127: 187, 1943.
7. Masson, P. L.: Neuromyoarterial des regions Tactiles et ses tumeurs. Lyon Chir. 21:257, 1924.
8. Monsees, N. L., and Murphy, G. A.: Distal phalangeal erosive lesions. Arthritis Rheum. 27:449, 1984.
9. Murray, R. M., and Stout, A. P.: The glomus tumor. Investigations of deformity and behavior. Am. J. Pathol. 18:183, 1944.
10. Sugiura, I.: Intraosseous glomus-tumor. A case report. J. Bone Joint Surg. 58B:245, 1976.