Proximal Interphalangeal Joint Fibromatosis After Pyrocarbon Implant Insertion: A Case Report

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Abstract

Background: Pyrocarbon implants represent an increasingly popular method to treat proximal interphalangeal joint dysfunction. To this point, no association has been shown between pyrocarbon biomaterials and fibromatosis. We present a potentially serious and destructive complication associated with pyrocarbon arthroplasty. **Methods:** We demonstrate a clinical case involving pyrocarbon arthroplasty and subsequent fibromatosis development in an otherwise healthy 23-year-old female. To present this association, we illustrate the diagnostic workup involved in a rapidly expanding soft tissue mass of the hand and explain the appropriate treatment. **Results:** Pyrocarbon arthroplasty was associated with development of locally destructive fibromatosis confirmed by histopathological examination. Treatment involved wide resection with preservation of local structures. **Conclusion:** We describe the first association between fibromatosis and pyrocarbon biomaterial. Due to fibromatosis destructive effects, clinicians should be aware of potential complications associated with these materials and know how to accurately diagnose and treat these lesions.

Keywords: arthroplasty, fibromatosis, proximal interphalangeal joint, pyrocarbon

Proximal interphalangeal (PIP) joint pain, instability, and deformity resulting from trauma or rheumatoid arthritis are often treated with arthroplasty using silicone or pyrocarbon implants.^{2,10} The complications most commonly seen with pyrocarbon implants include dislocation, infection, and need for revision, but there has been no association between pyrocarbon and benign or malignant soft tissue changes.⁴ Although a direct causal effect cannot be established based on our findings, this report describes a unique association between small joint arthroplasty and fibromatosis and its devastating effect on joint form and function. Furthermore, we review the literature describing fibroblastic changes following the placement of various implants. Given the expanding use of pyrocarbon implants for joint arthroplasty, physicians need to be aware of this potential complication.

Case Report

A 23-year-old woman had a history of left ring finger trauma at the age of 10 that resulted in chronic traumatic arthrosis and minimal motion that she endured for 12 years. She wanted functional recovery, and therefore, she underwent a pyrocarbon PIP joint arthroplasty at an outside institution. One year later, she presented to our clinic with a 6 × 3 cm firm, nonfluctuant, soft tissue swelling encasing the dorsal aspect of the left ring finger at the PIP joint as well as

increased pain, significant deformity, and decreased motion (Figures 1a and 1b). Examination of patient's epitrochlear and axillary lymph nodes demonstrated no lymphadenopathy. Radiologic examination of the left ring finger PIP joint demonstrated volar dislocation, ulnar deviation, and a prominent soft tissue lesion extending from the left fourth web space to immediately distal to the PIP joint (Figures 1c and 1d). The patient had Middle East ethnicity with no family history of soft tissue malignancy, fibromatosis, Dupuytren disease, or other connective tissue disorders. There was no clinical evidence of infectious etiology, and laboratory tests demonstrated no leukocytosis or elevation of inflammatory markers. Due to concern for sarcoma, a first-stage incisional biopsy was performed for a definitive diagnosis.

We proceeded with the incisional biopsy by making an incision along the plane where we planned to ultimately resect the tumor allowing us adequate exposure for sampling of the lesion. Exposure of the mass demonstrated a 6

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Figure 1. (a) Dorsal and (b) volar aspects of the left hand showing a firm, nonfluctuant soft tissue swelling and ulnar deviation of her ring finger PIP joint. (c) posterior anterior and (d) lateral radiographic images of the left ring finger PIP joint depicting volar dislocation with ulnar deviation of the implant. A prominent soft tissue lesion is also visualized extending from the left fourth web space to immediately distal to the PIP joint.

Note. PIP, proximal interphalangeal.

× 3 cm firm, fibrous mass encasing the dorsal aspect of the left ring finger proximal phalanx and PIP joint. Biopsies were obtained for definitive tissue diagnosis. The mass did not give any clinical evidence of infection, and therefore, no tissue cultures were taken. Final histopathological examination of the soft tissue mass revealed uniformly appearing elongated spindle cells with no atypical cellular nuclei or anaplasia—an appearance consistent with fibromatosis.

Once final pathology of fibromatosis was confirmed, we proceeded with a second stage for definitive management of the PIP joint mass by marginal excision through the same incision as previously noted. Through a dorsal approach, we exposed the mass by retracting the extensor tendon and

resected the 6 × 3 cm tumor off the PIP joint (Figures 2a, 2b, and 2c). The entire pyrocarbon implant was intact and removed (Figure 2d). We filled the residual defects in the medullary cavities with corticocancellous bone allograft (Community Tissue Services, Kettering, Ohio) and stabilized the joint with a Kirschner wire and 1.5-mm plate (Synthes, Inc, West Chester, Pennsylvania) fusing the finger at 30° of flexion. This allowed increased stabilization to restore finger length in a young woman who wanted to wear a wedding ring. Histopathology of the mass showed results similar to those seen at the initial biopsy consistent with fibromatosis. No implant debris was observed (Figure 3) and she had a stable fusion of the PIP joint (Figure 4). The

Figure 2. (a) Dorsal exposure of soft tissue mass. (b) Excised mass measuring 6×3 cm. (c) Pyrocarbon implant visible in the left ring finger proximal interphalangeal joint. (d) Removal of the pyrocarbon implant.

Figure 3. (a) Histology demonstrating well-circumscribed soft tissue mass that lacks a surrounding capsule. (b) High-power magnification shows elongated, spindle-shaped cells of uniform appearance surrounded by dense bands of collagen. Minimal mitotic figures noted with no cellular atypia or hyperchromatic nuclei.

Figure 4. (a) Left ring finger proximal interphalangeal joint after fusion with (b) posterior anterior and (c) lateral radiographic images.

Kirschner wire was removed 4 weeks later and the internal plate remained in place.

At her nine-month follow-up, her fusion remained stable and she was doing well functionally. She was pregnant and therefore declined radiographs. She had minimal residual swelling on the ulnar border of her finger and will continue with surveillance for recurrence (Figure 5).

Discussion

Fibromatosis represents a nonneoplastic lesion characterized by fibroblastic proliferation that is often locally aggressive and has no metastatic potential.⁷ Clinically, fibromatosis may present as either a rapidly expanding or slow growing lesion. Patients often complain of pain, pressure, and deformity or contracture owing to fibrosis of underlying fascia, tendons, nerves, or muscles. Because the clinical presentation is similar to soft tissue sarcomas, a tissue sample is often required for definitive diagnosis.

Histologically, these tumors are often well circumscribed lacking a true capsule. They are composed of elongated spindle cells of uniform appearance with intermittent collagen and reticular fibers. Although these masses may be rapidly growing, mitoses are infrequent with minimal variation in cellularity and no cells demonstrating atypical nuclei or anaplasia.¹³

Fibromatosis is a rare tumor with 2 to 4 cases per million people reported annually.³ The age of onset is generally between 30 and 50 years; however, hand involvement has been seen in children and adolescents.⁶ Fibromatosis of the hand is classified into 2 types: superficial and deep. Superficial fibromatosis affects the fascia whereas deep fibromatosis involves tendon and muscle.

The etiology of fibromatosis remains unclear, but multiple causative factors have been reported including diabetes, alcoholic cirrhosis, smoking, genetic, and local trauma.¹³ Of these factors, our patient's only positive finding was local trauma to the PIP joint. Furthermore, literature review

Figure 5. Three-month follow-up (a) posterior anterior and (b) lateral radiographs demonstrating fusion of the PIP joint. (c) Volar and (d) dorsal nine-month follow-up pictures illustrating well-healed scar and minimal residual swelling over the ulnar border of her ring finger. Note. PIP, proximal interphalangeal.

did not reveal any association between pyrocarbon and fibromatosis;⁴ however, there are a few cases reported of fibromatosis following other implant materials including a total hip replacement,¹⁴ breast implants from both silicone and saline implants,¹⁵ and one report of fibromatosis following an internal jugular catheter placement.¹ Reports of pyrocarbon implant complications demonstrate two cases of local reactions that include joint swelling secondary to joint capsule irritation and osteophyte formation. These cases were treated by extension splinting and excision respectively.^{2,10}

In contrast, sarcomas are histologically distinct in that they demonstrate poorly differentiated mesenchymal cells with marked nuclear abnormality, increased mitotic rate, high cellularity, and anaplasia.⁸ The cause of soft tissue sarcoma is poorly understood. Because the treatment of sarcoma is aggressive and may require radical resection, radiation, and systemic chemotherapy, it is critical to obtain a tissue sample to confirm that the tumor is nonmalignant.⁸

Despite fibromatosis being histologically benign, the clinical presentation of the tumor is similar to soft tissue sarcomas in that both conditions are locally invasive and have a high rate of recurrence.^{8,12} The resection of fibromatosis with negative margins is widely accepted as the treatment of choice.⁷ Nonsurgical options are available including radiation, hormonal therapy, chemotherapy, and biological therapies (tyrosine kinase inhibitors), but these options have only been successful in select cases.¹¹ Clinical and pathological features suggestive of fibromatosis recurrence are extremity location of the tumor, incomplete resection, local trauma, young age, increased mitotic index, areas of necrosis, and inflammation within the tumor.^{13,5}

Because of the similar presentation between benign and malignant soft tissue masses and the lack of documented association between pyrocarbon implants and soft tissue masses, we chose to stage our procedure to obtain a tissue diagnosis. Given the sudden onset of the mass and progressive deformity that occurred so acutely after implant placement, we hypothesize that the etiology of her neoplasm is

highly suggestive of a reactive process secondary to pyrocarbon arthroplasty. Furthermore, although anecdotal, previous associations have been described in the literature between foreign materials/implants and fibromatosis as noted previously. However, to this point no association has been made between pyrocarbon biomaterial and fibromatosis. The main objective of this report is to present a potentially serious and destructive complication associated with pyrocarbon implant arthroplasty, given its increasingly wide use for joint arthroplasty. However, no direct connection can be made between pyrocarbon biomaterial and fibromatosis, only an association.

To this point, the causative factors of fibromatosis are poorly understood and additional studies are needed to further delineate this relationship. Owing to the close temporal relationship of this patient's fibroblastic changes with her PIP joint arthroplasty, potential etiologies may include repeated local trauma from pyrocarbon implant failure and joint instability, or a reactive process following initial pyrocarbon arthroplasty. The inciting trauma leading to the original joint injury was in her remote past with no evidence of fibroblastic changes. This likely has less contribution to her current situation particularly given previous reports in the literature documenting the inciting trauma 3 to 4 months prior to fibromatosis formation.⁹

We describe an unusual association between pyrocarbon implants and fibromatosis, and demonstrate the diagnostic workup involved in this rapidly expanding soft tissue mass of the hand. Once the diagnosis is confirmed, the mainstay of treatment is wide resection with the preservation of local structures. Because the risk of recurrence in fibromatosis can be as high as 78%, close follow-up is crucial.

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Authors' Note

The content is solely the responsibility of the authors and does not necessarily represent the official views of the National Institutes of Health.

Ethical Approval

This study was approved by our institutional review board.

Statement of Human Rights

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008.

Statement of Informed Consent

Informed consent was obtained from all patients for being included in the study.

Declaration of Conflicting Interests

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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