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CASE REPORT

Extremity gossypiboma mimicking sarcoma: case report and review

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Abstract

A 70-year-old man with a history of bladder and colon cancer presented with an enlarging mass in his right lower extremity. Forty years before presentation, he had injured his right lower extremity in a motor vehicle accident. Imaging findings indicated suspected sarcoma, which led to biopsy. Biopsy and further surgical exploration revealed the presence of a surgical sponge and surrounding local inflammatory reaction. No neoplasm was found, and the sponge and involved tissues were removed. Gossypiboma is exceedingly rare in the extremities. Imaging of retained foreign material can appear suggestive of sarcoma because of strong inflammatory responses and local tissue mass-like derangement resulting in heterogeneous signal changes. Ultimately, biopsy must be performed to ensure that no oncological pathological condition is present.

Keywords Biopsy · Gossypiboma · Lower extremity · Retained sponge · Sarcoma · Soft tissue mass

Introduction

Retained surgical instruments or sponges are an uncommon complication estimated to occur in, at most, 0.02% of all procedures, with most reported cases occurring in large cavities, such as the abdomen or thorax [1–3]. Cases of retained instruments or sponges within the extremities are rare [4–11]. The retained sponge and surrounding inflammatory reaction is referred to by several terms, including gossypiboma [4, 7, 8, 11], textiloma [9], and cottonballoma [12]. The current case report describes a surgical sponge that was retained for more than 40 years and resulted in fibular osteolysis and other imaging characteristics consistent with malignancy.

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Case report

A 70-year-old man presented with a rapidly growing mass in his right lower leg. The patient had a history of colon and bladder cancer (both diagnosed 2 years earlier), which were treated with chemotherapy, radiation, and surgery. At the time of presentation, the patient had no known metastatic lesions. Other notable history included a motor vehicle accident that had resulted in open right fibula fracture and skin grafting 40 years earlier.

The patient reported that his right leg had always appeared slightly enlarged compared with the left leg (Fig. 1). However, he noted an increase in the size of his leg during the month before presentation, followed by weeping ulcerations on the skin surface. He had experienced no pain, weakness, loss of sensation, or any constitutional symptoms. Serum chemistries and complete blood cell count were within normal limits.

Radiographs showed a soft-tissue mass superimposing a gap in the diaphysis of the fibula (Fig. 2a). Heterotopic ossification or calcification was observed posterior to the distal portion of the remaining fibula. The distal tibia additionally had a lesion suggestive of bone infarct, better appreciated on the lateral view (Fig. 2b). A computed tomography (CT) scan revealed a lateral compartment mass with an ossification center and solid and cystic components that measured approximately 11.3 × 8.6 × 11.3 cm. Ossification spanning approximately 15.3 × 4.1 × 2.2 cm was noted within the posterior compartment abutting the flexor hallucis longus tendon, consistent with post-traumatic myositis ossificans (Fig. 3). There

Fig. 1 Clinical photograph of the patient's right lower extremity showing severe soft-tissue swelling, erythema, and ulcerations. A surgical scar was noted

Fig. 2 **a** Anteroposterior and **b** lateral radiographs of the right tibia and fibula showing a large soft-tissue mass (*short arrow*) associated with destruction of the mid-fibular diaphysis. The mass contains mixed density, including an area of dense soft tissue and calcification (*long arrow*). The distal tibia contains a lesion with a serpiginous sclerotic border and lucent center, features of an osseous infarct (*circle*). Ossification posterior to the tibia is present with a zonal pattern reminiscent of myositis ossificans (*open arrows*)

was clear evidence for osseous destruction of the fibular diaphysis.

Given the concerning imaging findings, a decision was made to proceed with open biopsy of the lesion. Possible diagnoses included infection and malignancy. A longitudinal incision was made along the lateral aspect of the leg, over the area of greatest mass prominence. As soon as the deep fascia was entered, fluid under pressure was encountered. Exploration revealed fibers consistent with suture. Further inspection showed a large mass with necrotic and inflammatory tissue (Fig. 4). Removal of a portion of the mass led to the discovery of more cotton fibers consistent with a retained surgical sponge with radiopaque markings. Because of the amount of time that had elapsed, the sponge had disintegrated and the markings were uninterpretable on imaging. Frozen sections were taken from areas suspicious for tumor. All were negative, though low-power photomicrographs showed fibers (Fig. 5). The remaining mass was then removed piecemeal. Fibers that had extended into surrounding tissues were removed, if possible. Cultures were ordered, and intravenous antibiotics were administered. Cultures grew *Alcaligenes faecalis, Staphylococcus aureus,* and group B streptococcus. The patient was discharged several days later on a 6-week course of piperacillin/tazobactam. One year after surgery the patient remains well.

Discussion

Retained surgical instruments are a concern. The overall incidence is unclear; however, it is estimated that 1 of 1000 to 1500 abdominal operations involves retained surgical instruments [3, 13, 14]. Because of recent concerns regarding safety, multiple checks are in place to count surgical instruments and prevent retention of needles, sponges, and larger instruments. If final counts are incorrect, retained objects can often be found with radiographic detection of metal instruments and radiopaque markers on sponges. Risk factors for retained

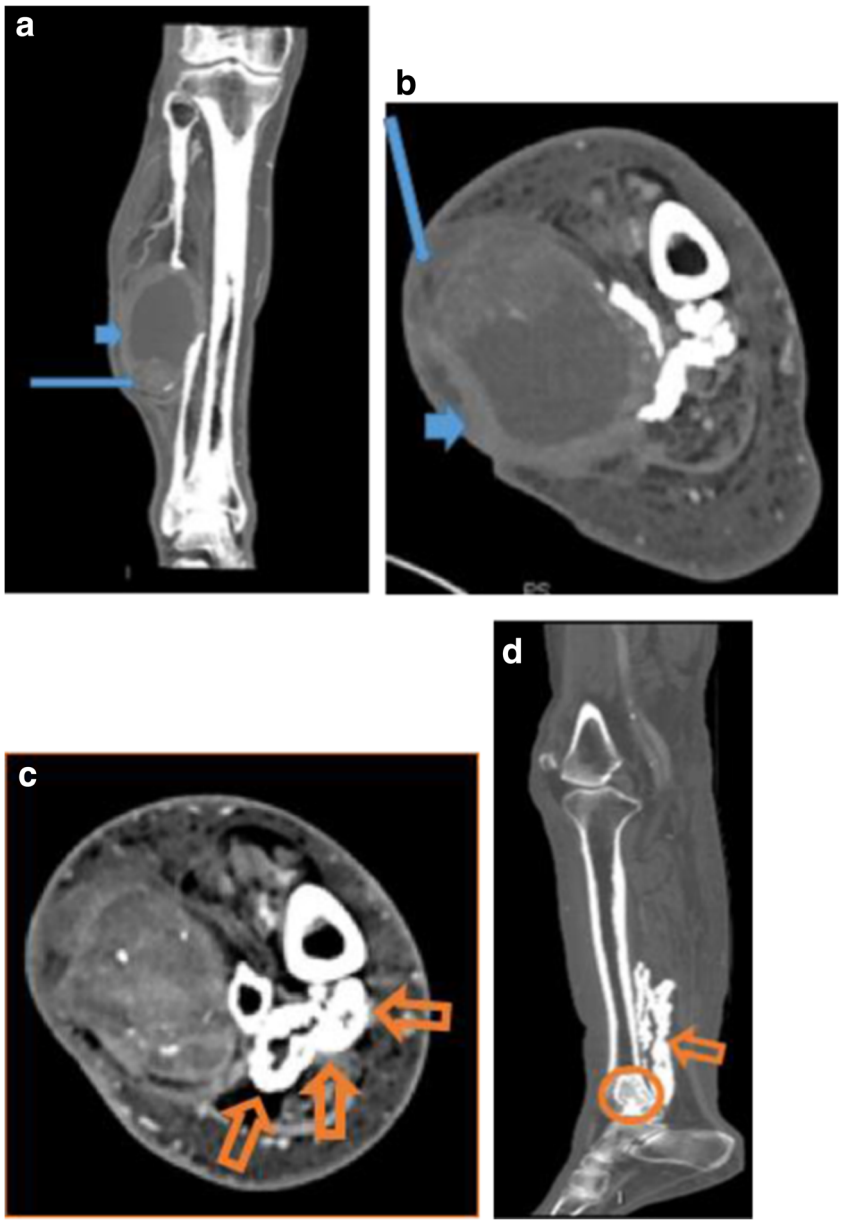

Fig. 3 **a** Coronal computed tomography image showing a large lateral compartment mass (*short arrow*) at the level of the mid-fibula, predominantly low density reminiscent of fluid, with central internal irregular high-density material and calcifications (*long arrow*). **b**, **c** Axial computed tomography images again show the large soft-tissue mass with a thick dense rim (*short arrow*) and a peripheral area of high-density material and calcification (*long arrow*). In addition, ossification in the posterior distal calf with central lucency (*open arrows*), reminiscent of myositis ossificans. **d** Sagittal computed tomography image showing a benign-appearing process with sclerotic rim, with features of a bone infarct (*circle*) and posterior calf myositis ossificans (*open arrow*)

Fig. 4 Intraoperative photograph showing the gross pathological condition consistent with a retained surgical sponge

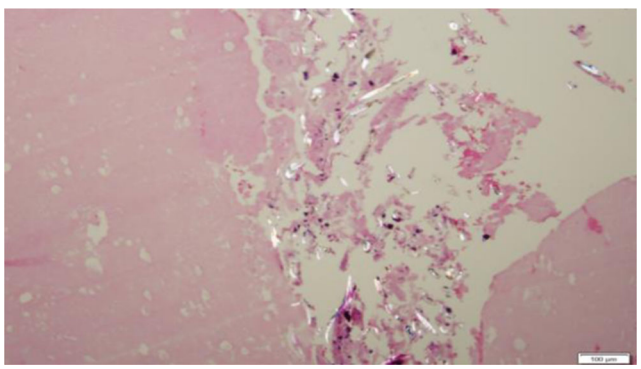

Fig. 5 Low-power photomicrograph with polarized light showing fibrin and abundant polarizable foreign fibrillar material

surgical instruments include emergency surgery, "unplanned" changes to a procedure, surgical team fatigue, and patient obesity [3].

There are many reported instances of retained sponges in the literature. Many of these describe sponges retained within the abdominal or thoracic cavity. This results in a local tissue reaction that can best be described as a foreign body granuloma and can be confused with sarcoma. Gibbs et al. [15] described two mechanisms by which a retained sponge can be discovered. In the first, an abscess can form adjacent to the sponge and can be concurrent with an inflammatory reaction. The other mechanism is thought to be an aseptic, fibrotic reaction and subsequent development of a fibrous mass.

Clinical manifestations of gossypiboma can be broad and can vary depending on the anatomic site of a retained sponge. The local inflammatory reaction that leads to gossypiboma formation can produce life-threatening complications after abdominal surgery. Abdominal gossypiboma can cause intestinal obstruction, fistulization, local infection leading to bacteremia and sepsis, gastrointestinal hemorrhage, and sinus formation [3, 16, 17]. Retained sponges within the limbs may cause local pain or swelling but may not cause major symptoms, which may lead to late discovery.

At least 19 cases of a retained sponge in an extremity have been reported (Table 1) at a median of 13 years after surgery (interquartile range: 4–30 years) [4–9, 11, 18–29]. All except one case (present in the shoulder [20]) have occurred in the lower extremity. One case was observed 3 months after resection of a myxofibrosarcoma of the buttock in an 84-year-old woman [27]. However, all other cases occurred 1.5 to 46 years after the most recent surgery. No cases had pathological testing positive for sarcoma. Although this may suggest that lesions are benign, the inflammatory reaction caused by the retained foreign body has the potential to cause osteolysis [4–11]. In two reports, patients with retained sponges for more than 13 [19] and 21 [23] years developed osteolysis leading to pathologic femur fracture. In only 2 cases (10.5%), patients presented with infection [26, 27]. In our case, the gossypiboma was associated with osteolysis and aggressive features shown on CT scan, which necessitated urgent

Table 1 Case reports of limb gossypiboma

Reference	Year	Location	Symptoms	Years since original surgery	Sarcoma formation	Postoperative course
Abdul-Karim et al. [18]	1992	Thigh	Pain/mass	35	No	Unknown
al Arabi et al. [19]	1992	Thigh	Pathological femur fracture	13	No	Unknown
Arora and Johal [4]	2014	Thigh	Pain/swelling	2[b]	No	Unknown
Bevernage et al. [20]	2006	Shoulder	Mass	1.5	No	"Uneventful"
Iwase et al. [21]	2007	Thigh	Pain/mass	12	No	Unknown
Kalbermatten et al. [5]	2001	Thigh	Swelling/weakness	25	No	No symptoms
Kominami et al. [22]	2003	Thigh	Pain/swelling	Unknown	No	Unknown
Kouwenberg and Frolke [6]	2009	Thigh[a]	Pain	7	No	Infection
Lo et al. [7]	2003	Leg	Pain/mass	5	No	No symptoms
Malot and Meena [8]	2012	Thigh	Pain/swelling	13[c]	No	Unknown
Mboti et al. [23]	2001	Thigh	Pathological femur fracture	23[d]	No	No symptoms
Mouhsine et al. [9]	2006	Leg	Swelling	3	No	Unknown
Patel et al. [24]	2007	Leg	Swelling	2	No	Unknown
Puri et al. [25]	2007	Thigh	Pain	7[e]	No	Recurrent
Sadeghifar and Saeed [26]	2013	Thigh	Pain/swelling/erythema/draining fistula	35	No	No symptoms
Sakayama et al. [11]	2005	Thigh	Swelling	40	No	No symptoms
Shiraev et al. [27]	2013	Buttock	Mass	0.3[f]	No	Unknown
Suh et al. [28]	2009	Thigh	Mass	13	No	No symptoms
Uchida et al. [29]	2010	Thigh	Mass	46	No	No symptoms
Present case	2018	Leg	Pain/swelling/ulceration	40	No	Swelling

[a] Above-knee amputation site

[b] At 4 years after fracture fixation and 2 years after fixation of periprosthetic fracture

[c] At 10 years after removal of implant and 13 years after original surgery

[d] At 21.5 years after removal of intramedullary nail and 23 years after original surgery

[e] Mass presented 7 years after original surgery and recurred 6 years after resection

[f] Mass noted 3 months after resection of high-grade myxofibrosarcoma

biopsy to rule out malignancy, especially in the setting of an oncological history.

The first-line assessment of a potential gossypiboma, as with any extremity mass, typically includes a radiograph. On radiography, the imaging features of a gossypiboma are highly variable and include a nonmineralized soft-tissue mass that may be associated with adjacent periosteal reaction and lysis or potentially osteoblastic activity; the retained sponge is radiolucent, although calcification of the foreign body may occur. (Currently, sponges are lined with radio-opaque material to aid in their detection). When calcification occurs, a thick "calcified reticular rind" has been described [30] and is better demonstrated by cross-sectional CT imaging than radiography. In addition, although gossypiboma may produce an aggressive appearance, a specific imaging feature, when present, has been described that can guide a radiologist toward the diagnosis. Gossypiboma appears as a mass containing whorl-like internal architecture and occasionally gas, produced by bacteria trapped in gauze fiber [31]. Such features are better demonstrated by cross-sectional imaging than radiography, which is another reason for using cross-sectional imaging in these cases [32]. The fiber architecture is better demonstrated by MRI than by CT, given the increased contrast resolution offered by MRI. However, the characteristic whorl-like architecture and gas are not commonly present in gossypibomas of the extremities and are more likely (although not present with a high frequency) in abdominal and pelvic gossypibomas [33] where intestinal bacteria exist.

Unfortunately, the cross-sectional imaging features of a gossypiboma of the extremities are otherwise not highly specific for this diagnosis and can vary depending on the duration of the sponge retention. Typically, the lesion is largely heterogeneous in signal, particularly on fluid-sensitive sequences, and typically hypointense by T1-weighted imaging. When an intravenous contrast agent is administered, the enhancement pattern has been described as predominantly peripheral with areas of nodularity and heterogeneous areas of enhancement [5, 11, 32], sometimes with adjacent osteolysis and periosteal reaction, similar to our case. On ultrasound imaging, an acoustic shadow is present in 64% of cases [32], although correlative radiographs are needed to rule out mineralization within the textile fibers that account for the acoustic shadow; the mass is often lacking in vascularity on ultrasound images. Ultimately, without the characteristic whorl-like interior architecture or peripheral rind of calcification and a clinical history of a retained sponge in the area of interest, the imaging features of gossypiboma can mimic those of sarcoma.

In our patient, given the impressive imaging findings on CT, MRI and ultrasound imaging were deferred because these modalities were felt to be unlikely to produce features that would change the immediate clinical plan (biopsy).

The differential diagnosis of gossypiboma includes calcific myonecrosis, which has similar imaging features and clinical history. Calcific myonecrosis has been observed almost exclusively in the calf and is thought to occur after a previous injury, resulting in a subacute or untreated chronic compartment syndrome [34, 35]. In many cases, symptoms develop several decades after the original injury [36]. Radiographs and CT scans demonstrate linear calcifications that conform to a single muscle or compartment with possible erosion into the bone [37]. Although the posterior distal calf calcifications in the current case could suggest calcific myonecrosis, the presence of an additional large lateral compartment mass hints at a different diagnosis. Therefore, biopsy was indicated in our case. If a sole diagnosis of calcific myonecrosis was determined from the imaging and clinical history, biopsy may not have been pursued given the reported risk of secondary infection and sinus tract formation and the overall benign nature of this particular diagnosis [37].

To our knowledge, there are no reports of sarcomatous transformation of an extremity gossypiboma in the setting of a retained sponge. However, this phenomenon has been reported in other locations throughout the body (Table 2) [38–41]. In all four cases, angiosarcoma was diagnosed. Notably, all locations were in the abdominopelvic region and occurred 20–38 years after the initial procedure. Two patients had distant metastases, and all four patients died within 2 months of discovery of the retained surgical sponge. Although there have been no cases showing association of retained sponges with sarcoma in the extremities, there have been many reports of secondary malignancies occurring in the environs of other retained foreign bodies [40]. These cases highlight the importance of biopsy, even if gossypiboma is the leading diagnosis on imaging. Careful biopsy along foreign body margins must be undertaken to rule out a secondary oncological process. In the current case, all margins were

Table 2 Case reports of angiosarcoma formation after retained surgical sponge

Reference	Year	Location	Presentation symptoms	Years since original surgery	Metastasis	Outcome
Ben-Izhak et al. [38]	1991	Pelvis	Rectal bleeding/anemia	25	Yes	Death
Cokelaere et al. [39]	2001	Abdomen	Flank pain	38	Unknown	Death
Jennings et al. [40]	1988	Abdomen	Abdominal mass	20	Yes	Death
Joo et al. [41]	2005	Abdomen	Constipation/mass	20	Unknown	Death

negative, necessitating only intravenous antibiotic therapy for local microbial control.

In this case, it is difficult to discern why the patient developed symptoms more than 40 years after initial surgical sponge retention. The patient may have had a unique set of recent risk factors that predisposed him to development of a local inflammatory response. Systemic chemotherapy may have suppressed his immune system, allowing for bacterial seeding at the site of the retained sponge or local immune derangement leading to unsuppressed inflammatory reaction at the site. He likely had a previous aseptic, fibrotic reaction that may have become locally infected after immune suppression from chemotherapy.

Conclusion

We report a case of a retained surgical sponge in the lower extremity leading to an acute local inflammatory reaction 40 years after the initial procedure. This led to gossypiboma formation, substantial osseous destruction, and features indicating suspected malignant transformation. Although no sarcoma was identified, it is important to carefully examine and biopsy the local tissue to rule out malignancy.

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Compliance with ethical standards

Conflicts of interest The authors declare that they have no conflicts of interest.

Informed consent All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards. Informed consent was obtained from all individual participants included in the study.

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