

Information Statement

Inappropriate Excision of Unrecognized Soft Tissue Sarcoma

This information Statement by the Musculoskeletal Tumor Society (MSTS) was developed as an educational tool based on the available literature and opinion of the authors. It is not a product of a systematic review. Readers are encouraged to consider the information presented and reach their own conclusions.

Summary Statement

The Evidence Based Medicine Committee of the Musculoskeletal Tumor Society recommends the following:

1. Soft tissue masses that are **larger than 5 cm or enlarging** have a significant likelihood of being malignant and should be promptly referred to a sarcoma specialist. MRI of the entire tumor with and without contrast may be considered prior to referral.
2. Inappropriate excision of soft tissue sarcomas should be avoided whenever possible as it exposes patients to additional morbidity from a second, possibly more complex, surgery. In the case of large, deep sarcomas patients may experience worse oncologic outcomes.
3. The literature reflects that inappropriate excisions of soft tissue sarcomas are being performed by general and plastic surgeons, in addition to orthopedic surgeons, so efforts at physician education should include multiple specialties .

Statement of Clinical Concern

Patients with unrecognized soft tissue sarcomas often undergo inappropriate biopsy or non-oncologic excision before referral to appropriate specialists. In addition to increased expense and anxiety for the patient, non-oncologic excision can increase treatment-associated morbidity. One study even showed increased mortality in the case of inappropriate excision of Stage III sarcomas [9].

Despite ample evidence advising against inappropriate excision, this problem has persisted in clinical practice accounting for an estimated 30% of sarcoma referrals, with reported rates ranging from 18-53% [5, 11, 12,14]. In part, we believe this may be due to knowledge gaps or barriers to information dissemination. In some cases, surgeons may be working outside the normal scope of their training.

In the past, much of the of the literature on this subject has referred to these events as “inadvertent excisions.” However, because many of these procedures were likely elective, scheduled surgeries, and to emphasize the risk of additional morbidity to the patient, we suggest a more apt description would be “inappropriate excisions of an unrecognized sarcoma.”

Background and Literature Review

In 1982, Mankin et al published the manuscript "The hazards of biopsy in patients with malignant primary bone and soft-tissue tumors" [6]. In this study, the authors surveyed members of the Musculoskeletal Tumor Society to determine the frequency and consequences of inappropriate biopsies or attempted excisions of bone and soft tissue malignancies before being referred to sarcoma centers. They found that biopsy-related errors occurred "three to five" times more often when the biopsy was performed at the referring rather than at the treating institution. In 15/329 (8.5%) patients, an amputation was required for local control as a direct consequence of a poorly-executed biopsy or resection. This paper stimulated a society-wide educational effort to reduce the frequency of these adverse events.

In 1992 the same authors repeated the study, collecting data on 579 patients from 21 institutions [13]. Disappointingly, they found the rate of improperly performed outside biopsies negatively impacting treatment planning had not changed (19% vs 18%). They also found that improper biopsies were performed much more frequently for soft tissue versus bone tumors (17% vs 5%) and questioned if educational efforts directed toward orthopaedic surgeons alone was not sufficient, as general surgeons also commonly assess and treat soft-tissue tumors.

More recently, Potter et al reviewed their experience in 203 high-grade soft tissue sarcomas, of which 32% had undergone non-oncological excision, i.e. an excision that was not performed with the intent of obtaining adequate margins for cure, at an outside institution [8]. In addition to the surprisingly high number of patients who received inappropriate treatment, they also found that these patients required more complex surgery, such as skin grafts or flap coverage. Despite this additional surgery, these patients still had an increased rate of local recurrence (34% vs 6%), demonstrating the real cost to patients in terms of increased morbidity and worsened outcomes arising from inappropriate sarcoma excision.

Siegel et al reviewed the surgical treatments required for 54 patients referred after inappropriate excision of soft tissue sarcomas. Of these, four required amputation and 11 (20%) required rotational or free flaps for coverage. Operative errors documented included transverse incisions in four patients, exposure of major neurovascular structures in three patients and extensive hematoma in three patients [10].

Referring Specialties

Several authors have documented the specialty of referring physicians for patients with inappropriate excisions of soft tissue sarcomas.

Siegel et al found that 38 of 54 (70%) of their patients who underwent inappropriate excision were previously treated by general surgeons, 11 of 54 (20%) by orthopaedic surgeons, and 5 of 54 (10%) by plastic surgeons [10]. We suspect this type of practice variation could present an opportunity for education and improvement.

In a 2012 report from the UK, Venkatesan et al found that 38% (16/42) of their patients with an inappropriate biopsy or excision had been treated by a general surgeon, 14% (6/42) by a plastic surgeon and 14% by an orthopedic surgeon [12]. In a series from Denmark, the largest proportion of inadvertent resections was performed by general surgeons (38%), followed by orthopedic surgeons (30%) [2].

Other US authors describe the patient characteristics of those referred after inappropriate excisions but do not specify the referring specialties. Alamanda et al found that among 147 patients referred after previous excision, neither distance from sarcoma center nor insurance status was associated with attempted excision prior to referral [1]. Potter et al report a high percentage of patients (64/203 [32%]) were referred after previous excision to their center but did not report distribution of referring specialties [8]. In a large but older series from Memorial Sloan-Kettering Cancer Center, 407 of 1092 patients (37%) had undergone resection before referral but again the referring specialties were not specified [5].

Anatomic Locations

Not all inappropriate excisions or biopsies are equal. With regard to soft tissue sarcomas, referral after resection of a superficial sarcoma (outside the muscle fascia) is not clearly associated with worsened oncologic outcomes. At least two retrospective studies have shown similar overall survival between patients who underwent primary vs re-excision of sarcomas [3, 5].

The similarity in oncologic outcome may reflect that the majority of inadvertent excisions occur in small, superficial soft tissue sarcomas which have an inherently good prognosis. Potter et al found that deep tumors comprised a smaller proportion of inappropriately excised tumors referred, ranging from 15% - 17% [5] [8]. It is worth remembering, however, that despite the similarity in oncologic outcome, patients undergoing re-excision still required two surgeries for their tumors.

In contrast to patients with small superficial sarcomas, patients who have undergone an inappropriate excision of a deep sarcoma are at increased risk of oncologic failure, even after adequate re-resection and radiation. Qureshi et al showed that the initial stage of the soft tissue sarcoma had a large impact on outcome. They compared the outcomes of 134 patients with

unplanned excision of soft tissue sarcomas with matched group of 209 patients with planned resections. The difference in the local recurrence rates for Stage I and Stage II tumors ranged from 12-15%. However, for stage III tumors, there was a 23% increase (38% vs. 14%) in local recurrence. Even more concerning, they found decreased disease-free and overall survival for Stage III patients. The authors commented that previous studies which did not demonstrate different outcomes for re-resection patients failed to match groups by initial stage [9, 3, 5].

Proposed Intervention and Management

Venkatesan et al suggest that the inadvertent excision of a small superficial sarcoma may be characterized as "bad luck" but that an inadvertent excision of a large deep sarcoma as "bad medicine." [12] We agree with this assessment. We should not accept these events as simply unfortunate but rather a failure to educate our peers.

Traditionally, a large tumor size, a history of growth, symptoms of pain and location deep to the fascia have been advocated as features that are predictive of sarcoma. However, a recent report found that presence or absence of pain was not a useful predictor of malignancy. Furthermore, depth of a tumor can be difficult to assess by examination [7].

Therefore, for soft tissue lesions, the most important clinical and radiological features concerning for malignancy should be a history of growth and size greater than 5 cm. These clinical findings should prompt treating physicians to consider prompt referral to a sarcoma specialist. At a minimum, these lesions require further imaging, which may lead to a biopsy, preferably at the direction of a sarcoma specialist involved in performing the definitive surgical management. For a referring provider to make a diagnosis after excising a tumor should not be considered appropriate care and strongly discouraged.

In 2006, Robert Grimer from Birmingham, UK published "Size Matters for Sarcomas" in which he argued that a soft tissue mass bigger than 5 cm and enlarging should be considered highly suspicious for sarcoma [4]. The familiar image of a golf ball (4.3 cm) was proposed to quickly communicate this size. In a follow up study utilizing Bayesian analysis, size greater than 4.3 cm and enlargement were the most predictive of malignancy [7]. The same authors also found static masses less than 3 cm in young (< 32y) patients had only 6% chance of malignancy. Finally, Grimer reported that an educational program, wherein a leaflet and golf ball imprinted with "is this a sarcoma?" were mailed to referring physicians, resulting in a 37% increase in referrals [15]. Efforts such as this should be applauded, and the lessons learned in the UK should be extrapolated to other health systems worldwide.

In summary, inappropriate excisions of unplanned sarcomas continues to be an issue that leads to significant morbidity and increased burden of disease while being a preventable occurrence.

Summary Statement

The Evidence Based Medicine Committee of the Musculoskeletal Tumor Society recommends the following:

1. Soft tissue masses that are larger than 5 cm and enlarging have a high likelihood of being malignant and should be promptly referred to a sarcoma specialist. MRI of the entire tumor with and without contrast may be considered prior to referral.
2. Inappropriate excision of soft tissue sarcomas should be avoided whenever possible as it exposes patients to additional morbidity from a second, possibly more complex, surgery. In the case of large, deep sarcomas patients may unfortunately experience worse oncologic outcomes.
3. The literature reflects that inappropriate excisions of soft tissue sarcomas are being performed by general and plastic surgeons, in addition to orthopedic surgeons, so efforts at physician education should include multiple specialties .

These recommendations reflect our professional opinion and are a pragmatic application of the available literature. More research to clarify the effects and epidemiology of this problem will likely be beneficial. However, we do not believe efforts at improved education of surgeons has to await more data.

While increased efforts to educate orthopaedic and general surgery trainees may minimize non-oncological sarcoma biopsies and excisions in the next generation of surgeons, there is ample evidence that this problem is substantial presently. We believe that improved patient outcomes are possible with prudent and timely referral of soft tissue sarcomas prior to invasive actions and we hope that this endpoint is achievable through improved education of referring physicians.

References

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