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ORIGINAL PAPER

Clinical management and surgical treatment of distal fibular tumours: a case series and review of the literature

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Abstract
Purpose Study reports clinical and functional outcomes of surgical treatment in a case series of nine patients with distal fibular tumours.
Methods Nine patients with distal fibular tumours were observed between 2005 and 2010. A PubMed search was performed using the terms "fibula", "lower limb tumour [cancer]", "sarcoma", "Ewing", "peroneal", "fibular metastasis", and "limb-salvage surgery".
Results In all our patients, lesions were unilateral. All patients complained of pain; limping was present in 5 of 9 tumours. Patients were managed surgically, except one who underwent local radiotherapy. In six patients, a benign or tumor-like lesion was detected. Malignancies consisted of metastatic lung adenocarcinoma (two cases) or multifocal mesenchymal cancer (one case). Non-malignant lesions were treated by curettage and filling, followed by internal fixation when needed. In malignant or locally aggressive lesions, metadiaphyseal fibular resection was performed. The literature search retrieved either case reports or small case series, reflecting the rarity of distal fibular tumours. Surgical treatment was successful in all patients with benign lesions, whereas the rate of success was 40–100 % in case of malignancies.
Conclusions Given the low incidence of distal fibular tumours, controversies exist about the optimal surgical management. Clinical observation and imaging should be reserved to asymptomatic benign lesions. In non-malignant tumours causing pain, limping, and pathological fractures; in malignancies, surgery is recommended. Finally, in patients with asymptomatic lesions of uncertain nature, biopsy and histological examination should be performed to plan appropriate management.

Level of evidence: IV

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Introduction

Malignant tumours of the lower limbs are less common and exhibit lower mortality rates compared with other sites [1–3]. The fibula is affected in 2.4% of primary bone tumours [4], with the proximal third being more frequently involved than the distal segment [2, 4]. Malignancies of the distal third of the fibula carry a better prognosis than proximal lesions [2], although some authors have not observed such prognostic difference [5].

Synovial cell sarcoma, osteosarcoma and Ewing's sarcoma are the most common mesenchymal cancers of the lower

limbs [2]. The incidence of fibular involvement in giant cell tumours (GCTs) and Ewing's sarcomas is approximately 1 % [6, 7] and 8 % [8], respectively. Osteosarcomas affect the fibula in 2–5.6 % of cases, with distal fibular localisations found in 0.47 % of patients [1, 4].

The fibula is a dispensable bone; hence, wide surgical margins are theoretically more easily achievable than in other skeletal sites. However, resections of proximal fibular tumours can be complicated by the proximity of the common peroneal nerve and the anterior tibial artery. Furthermore, this portion of fibula plays an important stabilising function for the knee. Ample resections of distal fibular lesions may be hampered by difficulties with soft tissue coverage and the possible impact on foot and ankle biomechanics [5, 9]. Effective systemic treatments are therefore necessary to avoid large resections, thus allowing the maintenance of fibular functions, while offering patients comparable prognosis [5].

In this study, we report clinical and functional outcomes of surgical treatment of a case series of nine patients with tumours of the distal fibula. A review of the literature on the subject is also provided.

Material and methods

Nine patients with distal fibular tumours were observed in our orthopaedic department between 2005 and 2010. Patients were referred to our outpatient clinic because of leg pain with or without limping. Before treatment, all patients underwent comprehensive clinical and imaging assessment, including plain radiographs, computerised tomography (CT) and/or magnetic resonance imaging (MRI).

A PubMed search was performed using the terms "fibula", "lower limb tumour [cancer]", "sarcoma", "Ewing", "peroneal", "fibular metastasis", and "limb-salvage surgery".

Results

Case series

The mean age of patients (three females and six males) was 44.3±24.8 years (range 17–76 years). All patients presented with unilateral lesions and pain either at rest or during activities, and five also limped (Table 1). In six patients, benign or tumour-like lesions were detected (Table 1). Two patients (patients 1 and 9) presented a metastatic lung adenocarcinoma, one within the distal interosseous membrane and the other in the lateral malleolus. The remaining case presented with distal fibular involvement in the context of a multifocal mesenchymal cancer (patient 8). Extraskeletal spread of disease was observed in patients 1, 8 and 9.

Surgery was performed in all but one patient, who underwent radiotherapy given his poor health status (Table 1). All operations were performed by the same orthopaedic surgeon. In patient 1 (metastatic lung cancer; Fig. 1), biopsy and histological diagnosis were obtained preoperatively. Depending on the site, extension and nature of the lesion, surgical treatment consisted in curettage plus filling with autologous bone or acrylic cement, or resection followed by internal fixation with plates with locked screws when needed (Table 1; Fig. 2). Patients 1, 8 and 9 also received chemotherapy.

The average duration of follow-up was 48 months, except for patient 1 who died 18 months after surgery, patient 8 who died six months after radiotherapy, and patient 9 who died 12 months after surgery. In the postoperative period, active and passive leg mobilisation was recommended. Weight-bearing was proscribed for one month. Subsequently, progressive load was allowed. Following surgery, the range of motion (ROM) was normal and comparable with the contralateral leg in all patients, except for patients 1 and 8, who presented a 10° limitation both in flexion and extension of the ankle. No pain or limping was reported, except for patient 8 who suffered from mild pain when

Table 1 Demographics, clinical characteristics, histology and treatment in our case series

Case number	Age (years)	Gender	Clinical presentation	Type of lesion	Treatment
1	66	Male	Pain; limping	Metastasis of lung adenocarcinoma	Metadiaphyseal fibular resection; arthrodesis
2	35	Female	Pain	Angioma	Resection; curettage; osteosynthesis
3	17	Male	Pain; limping	Aneurysmatic bone cyst	Curettage; filling
4	17	Female	Pain; limping	Fibroma	Curettage; filling; osteosynthesis
5	76	Female	Pain	Schwannoma	Resection; filling; osteosynthesis
6	35	Male	Pain	Osteochondroma	Resection
7	18	Male	Pain	Aneurysmatic bone cyst	Curettage; filling
8	66	Male	Pain; limping	Multifocal mesynchimal neoplasma	Radiotherapy
9	69	Male	Pain; limping; cutaneous rush	Metastasis of lung adenocarcinoma	Resection; filling; osteosynthesis

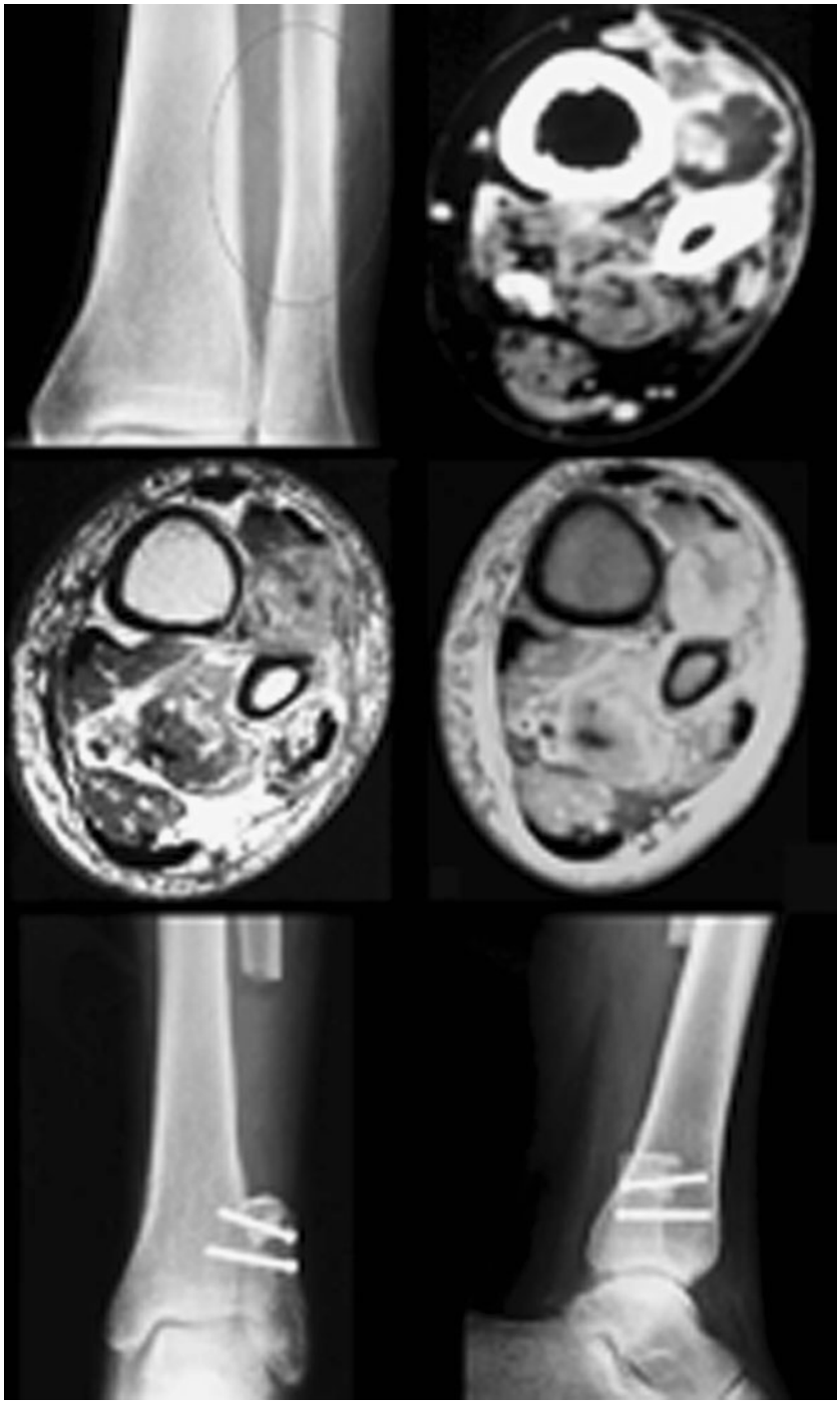

Fig. 1 The upper panels show plain radiographs and computerised tomography scan of a distal fibular lesion within the interosseous membrane consisting of a metastatic lung adenocarcinoma. The middle panels depict magnetic resonance imaging scans of the lesion. The lower panels show plain radiographs after fibular resection with tibial periosteal stripping and internal fixation with two screws

walking. In no case were varus-valgus instability, cutaneous necrosis, delayed wound healing or local infections observed. There were no local relapses.

Literature review

PubMed search did not retrieve large studies on surgical treatment of distal fibular tumours. Indeed, only single case reports or small case series have been published on the subject [3, 5, 10–31], reflecting the rarity of distal fibular tumours (Table 2). Available evidence indicates that surgery produces positive functional outcomes in all patients with benign lesions, whereas the rate of success ranges from 40 to 100 % in patients with malignancies (Table 3). These findings are in agreement with our results.

Capanna et al. [11] reviewed the outcome of 11 patients who underwent distal fibular resection for benign or malignant tumours with different techniques of reconstruction. Postoperatively, seven patients recovered a normal function, while four showed reduced mobility. One patient developed

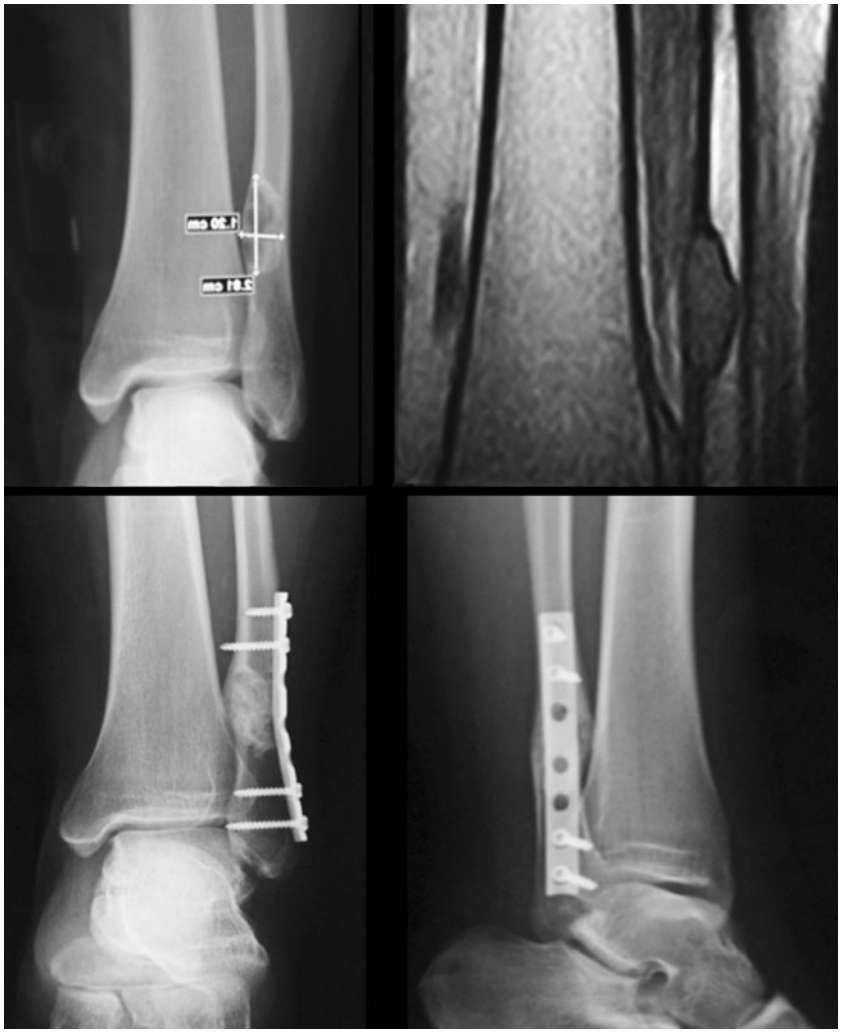

Fig. 2 The upper panels show plain radiographs and magnetic resonance imaging scan of a distal fibular fibroma. The lower panels show plain radiographs after curettage, filling and internal fixation with plate and screws

lateral subluxation of the talus. All patients were free of pain. Functional and clinical outcomes were independent of the nature of the lesion.

Dieckmann et al. [10] presented a series of 11 patients with distal fibular sarcomas or metastases, treated by resection of the whole fibula, followed by tibiotalocalcaneal arthrodesis using either a retrograde hindfoot nail or tibiotalar arthrodesis with screws. This technique failed in two patients due to osteopenic bone. Major advantages of arthrodesis with retrograde nails are increased stability and avoidance of extrinsic material in the wound area.

Lampasi et al. [17] reported nine patients with aneurysmal cysts of the distal fibula. Treatment consisted of curettage in six patients and resection in the remaining three patients. A graft from the contralateral fibula (one patient) or allografts (two patients) were positioned at the edge of the physis for reconstruction purposes. In the resection group, the number of operations, including removal of hardware, complications and time of immobilisation/orthosis were increased compared with the curettage group. These findings suggest that, although resection is effective, it should be reserved for aggressive or recurrent lesions [17]. In contrast, others have reported excellent functional outcomes in patients who had undergone resection for aneurysmal cysts [18] or osteochondromas of the distal fibula [22].

Another case series reported the outcomes in five patients with Ewing's sarcoma ($n=4$) or osteosarcoma ($n=1$) of the distal fibula [20]. In all patients, wide peroneal resections were performed. In two patients, a transtibial amputation was necessary because of inadequate surgical margins of the previous operation. The patient with osteosarcoma underwent tumour resection and ankle arthrodesis. No local relapses occurred in any case. In patients undergoing amputation or peroneal nerve resection, poor function was obtained without a prosthesis. However, all patients were disease-free over the time of follow-up (median 43 months).

Limb-salvage surgery using a tumour prosthesis appears to produce acceptable functional outcomes with clearance of the tumour in the medium term [3]. Larger studies and longer follow-up are needed to establish the efficacy of

Table 2 Publications on distal fibular tumours retrieved from PubMed ordered according to the number of patients reported

Publication	Number of cases
Capanna et al., Acta Orthop Scand (1986) [11]	11
Dieckmann et al., Int Orthopaedics (2011) [10]	11
Lampasi et al., J Bone Joint Surg Br (2007) [17]	9
Abuhassan and Shannak, J Bone Joint Surg Br (2009) [18]	8
Schneiderbauer et al., Clin Orthop and Rel Res (2006) [5]	8
Norman-Taylor et al., J Bone Joint Surg Br (1994) [21]	5
Ozaki et al., Acta Orthop Trauma Surg (1997) [20]	5
Chin et al., J Bone Joint Surg Am (2000) [22]	4
Lubliner et al., Bull Hosp Jt Dis Orthop Inst (1985) [13]	2
Yadav, Clin Orthop Relat Res (1981) [23]	2
Dogra et al., J Postgrad Med (1995) [14]	1
Durak et al., J Int Med Res (1996) [16]	1
Eger et al., Arch Orthop Trauma Surg (2004) [24]	1
Lee et al., J Bone Joint Surg Br (1999) [20]	1
Leichtle et al., J Bone Joint Surg Br (2006) [27]	1
Maccauro et al., Arch Orthop Trauma Surg (2003) [19]	1
Mohler and Cunningham, Foot Ankle Int (1997) [12]	1
Naples and Reeves, J Foot Ankle Surg (1994) [25]	1
Ongürü et al., Clin Imaging (2002) [26]	1
Palocaren et al., J Bone Joint Surg Br (2008) [31]	1
Saglik et al., Acta Orthop Belg (2008) [30]	1
Saglik et al., Foot Ankle Int (2008) [29]	1
Ramirez et al., Am J Orthop (2001) [28]	1
Uhl et al., Bull Hosp Jt Dis Orthop Inst (1990) [20]	1

prosthetic reconstruction. Careful patient selection with regard to emotional and cosmetic factors may justify this type of reconstruction [3].

Discussion

Tumours of the distal fibula are rare. However, their management poses significant challenges. Wide resections of the entire distal fibula can lead to loss of stability and critical soft tissue conditions, which require reconstruction of a stable ankle joint and sufficient skin coverage of the area. In patients with malignancies, limb-salvage surgery is rarely considered because a wide margin of resection is difficult to achieve. Notably, the incidence of inadequate margins has been reported to be higher after limb-salvage surgery for fibular osteosarcomas compared with other appendicular lesions [32]. Inadequate surgical margins are responsible for poor outcomes because of the high rate of local recurrence and metastasis [33]. This view has been recently challenged by other authors, who observed no recurrence and no impact on survival in patients with fibular osteosarcomas who had undergone intentional marginal resection and adjuvant therapy [5, 34]. It is likely that advances in chemotherapy will render limb-salvage surgery increasingly popular [3].

Surgical techniques for distal fibular tumours

Given the low incidence and diverse nature of distal fibular tumours, several solutions for different pathologies have been described [10]. For decades, below-knee amputation has been the main treatment for malignant tumours involving the distal fibula and tibia [20]. Advances in surgical techniques and chemotherapy have led to the introduction of alternative, less destructive approaches. For instance, distal fibular resection without reconstruction of the lateral side of the ankle is frequently performed [12, 15, 21, 35]. In such instances, ankle stability is obtained via either soft tissue and ligament reconstruction or tibiatalar arthrodesis

Table 3 Surgical outcomes in patients with distal fibular tumours

Publication	Number of cases	Type of lesion	Treatment	Rate of success (%)
Dieckmann et al., Int Orthopaedics (2009)	11	Sarcomas or metastasis	Distal fibular resection and tibiocalcaneal arthrodesis	73
Capanna et al., Acta Orthop Scand (1986)	11	Benign and malignant tumours	Distal fibular resection and reconstruction	64
Ozaki et al., Acta Orthop Trauma Surg (1997)	5	Ewing's sarcoma, osteosarcoma	Resection, arthrodesis, amputation	40
Norman-Taylor et al., J Bone Joint Surg Br(1994)	5	Ewing Sarcoma	Distal fibular resection	100
Lampasi et al., J Bone Joint Surg Br (2007)	9	Aneurysmatic bone cyst	Curettage or resection and bone graft	100
Abuhassan et al., J Bone Joint Surg Br (2009)	8	Aneurysmatic bone cyst	Resection	100
Chin et al., J Bone Joint Surg Am (2000)	4	Osteochondroma	Resection	100
Yadav, Clin Orthop Relat Res (1981)	2	Giant cell tumour, aneurysmatic bone cyst	Resection and ligament reconstruction	100
Lubliner et al., Bull Hosp Jt Dis Orthop Inst (1985)	2	Aneurysmatic bone cyst	Resection and allograft	100

[16, 23, 36]. In other cases, fibular resection is followed by reconstruction with allograft, autografts, pedicled vascularised epiphyseal transfers using the ipsilateral proximal fibula or a long bone graft from the iliac crest, bone transplants, or prosthetic ankle joint replacement [3, 11, 13, 24, 37–40].

At present, however, the choice of a specific procedure is frequently based on the surgeon's preference and experience, rather than being dictated by the nature and extension of the disease, reflecting the lack of guidelines and high-quality, large-scale studies on the subject.

Complications of surgery for distal fibular tumours

All surgical techniques described above present peculiar advantages and disadvantages [10]. Common complications of distal fibular resection include varus instability, valgus collapse, limited ROM and insufficient soft tissue coverage [21, 41–43]. In patients undergoing distal fibular resection and reconstruction with fibular head, donor site morbidity, lateral knee instability, peroneal nerve damage, incongruity between the proximal fibula and the talar joint surface, and non-union have been described [44, 45]. Talar collapse can occur after ankle joint replacement [3].

Conclusion

Given the low incidence of distal fibular tumours and the role of this bone in the ankle joint biomechanics, controversies exist with regard to the optimal surgical treatment. Based on our experience and the literature available on the subject, we propose a dual approach. Clinical observation with periodical imaging should be reserved to patients with asymptomatic benign or tumour-like lesions. In patients with non-malignant tumours presenting persistent pain, limping, pathological fractures or large lesions, as well as in patients with malignancies, surgery is recommended. Finally, in asymptomatic lesions of uncertain nature, biopsy and histological examination should be performed to plan treatment.

Based on our case series, benign and tumour-like lesions can be successfully managed with curettage and filling with bone substitutes, autologous bone grafts or acrylic cement. Resection and internal fixation with locked screws should be performed in case of locally aggressive benign tumours sparing the fibular epiphysis. Finally, in the presence of malignant lesions not involving the lateral malleolus and located at least 2 cm from the tibiotalar joint, metadiaphyseal fibular resection and periosteal tibial stripping is performed, followed by arthrodesis between the remaining lateral malleolus and the distal tibia, according to Capanna's technique. In malignant tumours involving the lateral malleolus, fibular resection with or without reconstruction or prosthetic ankle replacement may be recommended.

Conflict of interest The authors declare that they have no conflict of interest.

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