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Damage control orthopaedics: State of the art

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Abstract

Damage control orthopaedics (DCO) originally consisted of the provisional immobilisation of long bone - mainly femur - fractures in order to achieve the advantages of early treatment and to minimise the risk of complications, such as major pain, fat embolism, clotting, pathological inflammatory response, severe haemorrhage triggering the lethal triad, and the traumatic effects of major surgery on a patient who is already traumatised (the “second hit” effect). In recent years, new locations have been added to the DCO concept, such as injuries to the pelvis, spine and upper limbs. Nonetheless, this concept has not yet been validated in well-designed prospective studies, and much controversy remains. Indeed, some researchers believe the indiscriminate application of DCO might be harmful and produce substantial and unnecessary expense. In this respect, too, normalised parameters associated with the acid-base system have been proposed, under a concept termed early appropriate care, in the view that this would enable patients to receive major surgical procedures in an approach offering the advantages of early total care together with the apparent safety of DCO. This paper discusses the diagnosis and treatment of severely traumatised patients managed in accordance with DCO and highlights the possible drawbacks of this treatment principle.

Key words: Damage control orthopaedics; Early total care; Early appropriate care; Polytrauma; Resuscitation; External fixation; Packing; Embolisation; Compartment syndrome

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Core tip: Damage control orthopaedics (DCO) is the treatment of lesions that provoke major bleeding and pathological inflammatory response, whilst avoiding the traumatic effects of major surgery in a patient who is already traumatised (the “second hit” effect). The concept of DCO has not previously been validated and much controversy remains as

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to whether the indiscriminate application of DCO might be clinically and economically harmful. In addition, parameters associated with the acid-base system have been published with the idea that the existence of normalised parameters will enable patients to receive major surgical procedures, under a concept termed early appropriate care. This paper discusses the above concepts.

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INTRODUCTION

The concept of damage control orthopaedics (DCO)^[1] originally concerned the provisional immobilisation of long bone fractures - mainly the femur - in the severely traumatised patient (STP) in order to minimise the traumatic effects of non-life saving surgical procedures, termed the "second hit" effect^[2-5]. In recent years, new locations have been added to the DCO concept, such as pelvis fractures, spine fractures and upper limb injuries^[6].

Haemorrhage is a major cause of acute morbidity and mortality in the STP, and it also worsens the evolution of the generalised inflammatory response^[3-5]. Although haemorrhage complicates the generalised inflammatory reaction, transfusion may also aggravate the general traumatic syndrome, as this therapy can provoke complications in a patient who already presents a pathological inflammatory response. Massive transfusion can also provoke coagulation abnormalities, ion disorders and immunosuppression with subsequent infection as well as proclivity to lung injury and hypothermia^[6,7]. Therefore, blood transfusion can be a life-saving procedure, but it may also provoke a "second hit" reaction. This ambivalent effect can also arise from surgical procedures. Even interventions aimed at stopping a haemorrhage can provoke the release of molecules that aggravate the coagulation mechanism and heighten the inflammatory response. The fundamental goal of DCO is to do as little as possible in order to avoid further damage, and, therefore, only life-saving procedures should be performed when the patient's condition is acute.

The idea of doing "as little as possible" but "sufficient" to save the patient's life, the philosophy on which DCO is based, remains ill-defined. Although DCO is currently applied worldwide, the concept has not been validated in well-designed prospective studies, and controversy remains as to whether the indiscriminate application of DCO might be harmful and incur substantial unnecessary expense^[8]. The Polytrauma Study Group of the German Trauma Society reviewed 63 controlled trials of DCO but found no generalised management strategy^[9]. Similarly, a study conducted in the United States reported DCO implementation rates in reputed institutions ranging from 12%^[10,11] to 57%^[12]. Thus, there is a need to propose a better definition of the general physiopathology of major trauma in response to the need for a universal validation of DCO.

In addition to the above concerns, molecular-mediated mechanisms responsible for trauma-inducing coagulopathy^[13,14], susceptibility to infections^[13] and fracture-healing impairment^[15] all remain poorly understood^[16]. In consequence, the relationship between levels of inflammatory biomarkers and the "second-hit" effect is not firmly established. In this respect, only a few small prospective studies have been undertaken.

In a related area, a small non-comparative study was performed to consider the immediate impact of intramedullary femoral nailing, as the second hit, on multiple trauma patients, measuring various indices of haemodynamic stability, coagulation, fibrinolysis, oxygenation and inflammatory cytokines in the blood, using a pulmonary artery catheter before nailing. However, this analysis did not enable firm conclusions to be drawn^[17].

Some indices (the thrombin/anti-thrombin complex, tissue plasminogen activator and interleukin-10) present maximum values at the time of admission before surgery (first hit), while others (tissue factor, plasminogen activator inhibitor, tumour necrosis factor- α , interleukin-6 and pulmonary shunting) increase later, at 48-72 h after surgery. None of the remaining indices considered are significantly affected, other than a transient increase in pulmonary vascular resistance at around 2 h after

surgery^[17].

Besides the trauma severity itself, genetics is also thought to play an important role in the inflammatory response^[18-20], but in this respect, much remains to be determined before any clinical application can be made.

In summary, the concept of DCO is far from being universally accepted and validated, and the cornerstone of major trauma survival continues to be the control of bleeding and the inflammatory response. Although in the case of major bleeding, blood haemoglobin concentration sensitivity may be very low, this is a key variable, together with blood pressure, to be taken into account when rapid treatment decisions must be taken.

Attention should also be paid to other laboratory markers (evidence grade 1B)^[21]. Serum lactate and base deficit are very sensitive measures for detecting and monitoring the extent of bleeding and shock (evidence grade 1B) in conjunction with repeated combined measurements of prothrombin time, activated partial thromboplastin time, fibrinogen and platelets (evidence grade 1)^[21].

The current debate on DCO *vs* early appropriate care (EAC) has led to much discussion regarding the significance of various laboratory markers^[22-32]. In addition, it has been claimed that definitive early treatment of major fractures can be achieved under an EAC regime and more frequently than *via* DCO^[23-25].

Timely resuscitation enhances the initial treatment of fractures, and definitive fixation appears to be associated with a low incidence of complications. Therefore, early fixation usually results in better general and local outcomes, as well as being more cost-effective, and it has long been a major research goal to identify parameters associated with early fracture fixation. Since 2011, the Cleveland group^[23-25] has highlighted parameters believed to be associated with the acid-base system, based on the idea that if excessive base and lactate values can be normalised, patients will be better able to withstand major surgical procedures. The importance of this approach is that the consideration of any other metabolic parameter in the severely traumatised patient can then be dismissed. Under these circumstances, patients could be treated under a quasi-early total care (ETC) regimen, *i.e.* EAC, in an approach that might provide the advantages of ETC but the safety of DCO.

Authors who have supported the DCO concept^[22,30] in preference to that of EAC^[23-25] accept that the use of blood lactate levels is the main parameter to be considered in the management of patients with sepsis and/or septic shock^[31]. However, a normal acid-base situation does not necessarily mean that the patient's clinical condition is satisfactory or even that a surgical procedure can be performed^[25]. In this respect, other concepts such as the "triad of death", taking into account other indicators, can also be useful^[30].

In any case, the concept of EAC, as such, does not require the application of one or more specific surgical techniques, as is the case with ETC (*e.g.*, regarding intramedullary nailing versus ExFix under DCO). EAC is more a concept of metabolic permissiveness for the performance of ETC. Furthermore, the concept of DCO, which at present seems to be internationally accepted, is currently under review because of the belief that use of this technique is being abused. What EAC actually does more frequently approaches ETC. Some researchers have called for the validity of the above concepts and that of DCO in particular to be re-examined^[27-33].

RESUSCITATION

The number one priority in resuscitation is to stop the bleeding, while that of any treatment in the acute phase of the STP is to avoid hypovolaemic shock and the "lethal triad"^[34], and then to establish DCO. The time elapsed between injury and surgical intervention to control the bleeding should be minimised. Sustained systolic blood pressure of less than 80-90 mm Hg after treatment with vasoactive drugs is considered a sign of active bleeding, making the basis of resuscitation the prompt initiation of surgical treatment to stop the bleeding, together with the use of colloids to avoid the need for blood transfusion, if possible. Hypotonic solutions such as Ringer's lactate should not be administered to patients with significant brain injury^[21]. Therefore, surgical treatment is the baseline approach to resuscitation, and attention should be focused on the possible sources of severe bleeding: extensive skin lesions, injuries to the chest, abdomen, pelvis or lower limbs and long-bone fractures, particularly the femur. Fractures in the skeleton, especially the pelvis or the femur, are major causes of bleeding and can provoke highly dangerous or even fatal haemorrhages.

Pelvic fractures may be accompanied by ruptures to major vessels or injuries to vascular plexuses. When the STP is still haemodynamically stable, a contrast

computed tomography (CT)-scan should be performed before any X-ray projection, as pelvic and spinal fractures can be missed by conventional radiological studies^[35,36]. A fracture or dislocation of the pelvis due to an anteroposterior trauma provokes a broadening of the pelvic cavity and can be associated with vertical instability. These fracture patterns are the most severe and require prompt attention to close and stabilise the pelvic ring diameter to normal dimensions^[37].

The use of pelvic binders, a technique that dramatically reduces mortality rates^[38], is currently considered the gold standard for pelvic ring closure. Whenever possible, therefore, this approach should be taken in acute situations, whenever the pelvic ring is enlarged. Moreover, binders can be applied rapidly and simply, allowing transfer to the CT-scan with the pelvic ring closed^[39]. Either a commercially-manufactured device or a conventional sheet can be used as a binder^[40-43], although they can incite skin sores^[44] if maintained for more than 2-3 d^[45].

ExFix provides more stability than binders, avoiding anterior abdominal cavity compression and also the risk of skin necrosis^[45]. When properly applied, ExFix facilitates laparotomy, stabilising the pelvic bones^[6,7]. However, the ExFix technique is much more time-consuming and aggressive. It requires anaesthesia and an operating room and is more upsetting for patients. Moreover, the use of ExFix can lead to the development of pin track infection, and the use of subcutaneous fixation is not yet fully understood^[46]. The experience of other complications, too, has led ExFix, whether conventional or subcutaneous, to fall out of favour, and for the binder approach to be preferred^[47,48]. In any case, the conversion of ExFix to internal fixation should be performed as soon as possible^[49].

ExFix presents other problems, too, in relation to biomechanics. Although many attempts have been made^[50], posterior pelvic lesions are poorly stabilised by anterior frames^[49]. However, C-clamp devices^[50-53] applied to posterior lesions can achieve good bone fracture reduction^[54]. Nevertheless, due to the wide range of patient types and results that can be encountered, universal conclusions cannot be drawn, even from systematic reviews^[55]. C-clamping can be a dangerous technique, and iatrogenic lesions may occur as a result, even when the method is applied by an experienced surgeon. The main complications reported in this respect are migration into the pelvic cavity, which can provoke intestinal piercing or further bone fractures^[51,56].

Even in an emergency, a less invasive method, such as iliosacral screw internal fixation, an X-ray-guided technique, can often be used, if the surgeon is familiar with this procedure^[53,56-59]. Iliosacral screws usually produce better outcomes than the C-clamp, although pelvic dysmorphism can make this method technically demanding even for experienced surgeons^[52,56,57], particularly in an emergency setting^[58]. Moreover, both C-clamps and iliosacral screws require full integrity of the iliac bone, in the case of a pure iliosacral dislocation or sacral fractures^[53,57-60].

When there is major retroperitoneal bleeding that remains uncontained after pelvic ring closure, the indicated approach could be packing or embolisation^[6,7]. However, since pelvic ring closure produces a tamponade effect, stabilises the pelvis and occludes fractures, thus enhancing cessation of the haemorrhage, pelvic stabilisation is advised before any attempts are made at packing or embolisation^[6,7,37], although if necessary the latter could be performed even in the case of an unfixed, unstable pelvic fracture^[61].

Extraperitoneal packing is a safe and useful technique that facilitates the repair of any abdominal or pelvic cavity bleeding. Injuries to major vessels can also be treated by the extraperitoneal approach^[62]. It is especially useful for “in extremis” patients when a CT-scan is not advisable or when ring closure is impractical and also if further bleeding takes place following pelvic ring closure. Training in this technique is necessary, as it is not yet widely known^[63], although it is straightforward for a trauma surgeon and can be performed more rapidly than angiography plus embolisation^[64]. Moreover, the latter methods require the presence of a specialised radiology team, a resource that is not always available. Even when such a team is in full-time attendance, any embolisation during the night or weekend can make treatment schedules more complex and result in higher mortality^[65]. In addition, embolisation may only address arterial bleeding, and not that provoked by major vessels, and is very time consuming^[61,66]. Finally, this technique is associated with an increase of up to 10% in overall complications, including gluteal muscle necrosis, surgical wound breakdown, deep or superficial infections, impotence and bladder necrosis^[67].

In view of these considerations, the complementation of packing with angiography and embolisation appears to be a reasonable strategy^[68]. A systematic review in this respect concluded that pelvic packing, as part of a DCO protocol, provides crucial time for a more selective management of haemorrhage^[69]. Other technologies such as temporary partial intra-iliac balloon occlusion during internal pelvic fixation are also in use but to our knowledge have not yet been validated^[70].

Open pelvic injuries require special attention. A study of 29 battlefield trauma

patients reported mean blood requirements of 60.3 units during the first 24 h. Ring closure in these patients is often not possible, and other circumstances such as vascular, bowel, genital and bladder injuries are often coexistent^[71]. In this context, haemorrhage control, concurrent regional lesions and soft tissue lesion with infection prevention are the main issues to be addressed^[72-75]. Apart from clinical inspection, including consideration of possible injuries to the bowel and urinary systems, a contrast CT-scan is mandatory when the patient is haemodynamically stable. When there is a bowel lesion, early diverting colostomy is usually necessary^[72-75]. Experience with battlefield pelvic wounds is often valuable in the subsequent treatment of civilian patients, particularly in relation to open blast injuries to the pelvis^[71].

FEMUR FRACTURES

Femur bone fractures can also provoke acute life-threatening bleeding. Therefore, high-energy femur fractures must be promptly recognised and immobilised^[76]. Since these fractures can provoke major limb deformity, diagnosis is usually immediate by simple inspection, and treatment under a DCO regime advises speedy immobilisation by ExFix^[76,77] in order to avoid poorer outcome, further surgical interventions, more blood transfusion and a longer hospital stay^[78,79]. Nonetheless, for “in extremis” patients, a non-compressive garment or skeletal traction can be appropriate^[80], as a fracture fixation method would not produce a useful effect on the incidence of systemic complications in severely traumatised patients^[12]. Moreover, some authors have found that early intramedullary nailing can reduce the need for mechanical ventilation, and decrease treatment costs^[81]. The presence of concurrent lesions, particularly abdominal injuries^[77,82] or a bilateral femur fracture is, in any case, a very important variable for worsening the outcome, particularly with respect to abdominal injuries^[77,82]. In these cases, at least, management with ExFix is advisable.

SPINAL FRACTURES

Multiply-injured patients frequently present spinal trauma^[83]. Thus, 93% of victims of fatal traffic accidents present a cervical fracture, while among survivors of such accidents, up to 40% have cervical fractures, and 10%-30% have thoracolumbar fractures, which can provoke a permanent neurological deficit. In order to prevent these complications, it is important to apply the ATLS®-protocol^[84], bearing in mind possible vertebral lesions and how to prevent the aggravation of neurological injuries. Correct performance of the log-roll manoeuvre and maintaining sufficient blood perfusion, especially for patients with injuries to the central nervous system, is mandatory. In the case of spinal shock, hypotension must be treated rapidly with vasoactive drugs^[85].

Spinal injuries can frequently be overlooked in severely traumatised patients, and so a full body CT-scan should be performed, as an appropriate diagnostic test to detect possible spinal fractures^[85,86,88].

The prompt diagnosis and proper management of spinal lesions are aspects of overriding importance. However, questions may arise as to what type of treatment is most appropriate for severely traumatised patients with associated spinal column fractures. According to most studies, early fixation is preferred; this approach is safe, decreases the incidence of pulmonary complications and neurological damage, reduces the duration of intensive care, lowers morbidity and enhances survival and neurological recovery^[89-92]. Hence, spinal DC is a staged procedure of immediate posterior fracture reduction and instrumentation within 24 h^[93,94]. Although immediate reduction and posterior stabilisation of spinal fractures is desirable, if necessary an interbody completion fusion can be performed, together with a large anterior decompression. Nevertheless, if possible, this should be carried out at a later stage, as further blood loss and a “second hit” with extensive soft tissue exposure can aggravate the patient’s general economy. Depending on the fracture type, additional anterior instrumentation may also be added^[95].

When sufficient closed reduction is feasible, posterior, less-invasive stabilisation systems (LISS) are to be preferred. When there is neurological damage, speedy open decompression may be required^[96,97]. LISS techniques provide various benefits, such as decreased blood loss, surgical time, patient morbidity, postoperative pain and infection rates, and improved outcomes^[98-100]. Studies comparing percutaneous fixation without fusion to traditional techniques have demonstrated similar outcomes in long-term follow-up and according to radiological parameters^[91,92,101,102]. In summary, whenever possible, LISS is a highly recommended option within algorithms for spinal

decompression.

UPPER LIMB FRACTURES

The presence of complex trauma in the upper limb is a challenge for the surgeon because it requires outstanding knowledge of the anatomy at risk^[103]. Soft tissue cleansing, the extraction of foreign bodies and radical debridement are needed to provide an appropriate base on which to stabilise the fractures^[104]. For forearm bone fractures, either ExFix or plates can be used as osteosynthesis methods. It is essential to preserve longitudinal vascular, nerve and tendon functioning and viable structures^[105] by using venous grafts to preserve circulation and by direct suture tension in peripheral nerves^[106]. Definitive coverage by means of skin and muscle flaps must be undertaken when the patient's general state allows^[107]. Currently, the development of negative pressure therapy systems facilitates delaying the repair of soft tissues and decreases the complexity of the reconstruction by diminishing the size of the wound^[108]. Avoiding postoperative rigidity is an important objective for patients who require that attention be paid to other, more urgent areas^[109].

PHARMACOLOGICAL MANAGEMENT

Tranexamic acid

Severe haemorrhage is one of the most important causes of death in the STP. To address this condition, tranexamic acid (TXA), an antifibrinolytic product, has recently been added to the pharmacological resources available and is now the only specific pharmacological treatment currently recommended^[110]. Nevertheless, doubts remain about its management, such as the appropriate dose and the characteristics of the patients who would most benefit from this treatment. Although most guidelines recommend a 1 g bolus, there is great variability of opinion regarding subsequent doses. Moreover, the mechanism of action responsible for its effects was not determined in the Clinical Randomisation of an Antifibrinolytic in Significant Haemorrhage-2^[111] or the Military Application of Tranexamic Acid in Trauma Emergency Resuscitation studies^[112,113] and remains unknown.

Researchers have concluded that in patients with more severe injuries the use of TXA is associated with a higher mortality rate regardless of the time of administration^[114]. Nonetheless, the latter was a retrospective study with a sample of 300 patients, and prospective studies are needed in order to identify the threshold of the beneficial effects of TXA.

While the prompt use of TXA is recommended, a much-debated topic is that of the time and site of its administration. Some recent studies have advocated the prehospital use of the drug, proposing that when in a given site it is not available for prehospital use, the patient should be transferred to another nearby hospital in order to receive this treatment even if it does not have sufficient infrastructure for trauma patients^[115,116].

A randomised clinical trial, conducted to evaluate the efficacy and safety of recombinant active factor VII rFVIIa as a complement for direct haemostasis in polytraumatised patients, concluded that the use of rFVIIa reduced the use of blood products but did not affect mortality compared to placebo treatment^[117]. Ongoing research studies seek to analyse the use of other agents for the control of trauma-induced coagulopathy. Thus, an experimental study in a porcine model was recently published regarding the effects of factor-based resuscitation on shock and trauma-induced coagulopathy and of prothrombin complex concentrate, TXA and fresh frozen plasma - both individually and in combination - on acute trauma-induced coagulopathy. The authors concluded that no benefit was obtained from the use of prothrombin complex concentrate or TXA, either as single agents or in combination, for resuscitation from haemorrhagic shock. However, the concurrent administration of plasma with these agents seems to provide good results in the treatment of haemorrhagic shock, by alleviating hypotension, decreasing lactic acidosis, improving coagulopathy and enhancing clot formation and quality^[118]. In conclusion, although controversies persist, the early use of TXA, within 3 h of the trauma, and even in the prehospital phase, is currently included in the initial management guidelines for severely traumatised patients.

THROMBOEMBOLIC PROPHYLAXIS

For years, it has been known that STPs are at high risk of suffering deep vein thrombosis (DVT) and pulmonary thromboembolism (PE), both of which are frequent causes of death. In each case, the incidence varies greatly from one study population to another, according to the diagnostic criteria used. Diagnoses of DVT and PE are becoming more frequent^[119], and pharmacological antithrombotic prophylaxis (AP) is needed, together with mechanical therapies. The questions of when AP should be initiated and which patients are at most risks remain highly controversial. Studies have suggested that risk factors include age older than 40 years, pelvic and lower extremity fractures, spinal cord injury with paralysis, cranial trauma, more than 3 d of mechanical ventilation, vascular injuries and shock at the time of patient admission and major interventions^[120].

The clinical variability observed means that it is currently impossible to protocolise AP. While the concurrent presence or risk of major haemorrhage is a major challenge, the start of AP is often delayed. Nevertheless, as DVT develops within the first days after trauma, AP should be set up as soon as possible^[119]. It has been demonstrated that in severely traumatised patients, initial hypocoagulability lasts for some 24 h. Accordingly, AP must be started after that time^[121]. On the other hand, in patients with cranial trauma presenting haemorrhage or with massive visceral lesions, spinal cord injuries or uncorrected coagulopathy, AP should be delayed. By starting AP 72 h after the traumatism, the incidence of DVT seems to decrease without progression of the haemorrhage^[122,123]. However, this outcome is not evidence-supported, and so it is advisable not to delay AP for the above-mentioned patients^[121].

Low molecular weight heparin (LMWH) seems to be the drug of choice for AP^[124], although the evidence in this respect remains insufficient. In a study carried out with 743 high-risk polytraumatised patients, in whom the AP was carried out with LMWH, the treatment was started once the patients were haemodynamically stable and the bleeding was under control. For patients with intracranial haemorrhage or spinal trauma, the AP was started when, according to the CT study, the intracranial haemorrhage was inactive. In patients with epidural analgesia, the AP with LMWH was started after removal of the epidural catheter. These patients were given a dose of 5000 units, once daily, administered by subcutaneous injection, and the AP was continued until the patient could walk independently. This treatment was maintained under the same regimen, even when the patient required further surgical treatment. The study concluded that this daily regimen with LMWH provided similar levels of safety and efficacy to those reported in previous studies when LMWH was given twice a day. In addition, the once-daily regimen, regardless of the need for further invasive procedures, obtained better results in terms of compliance^[125]. Although recent attempts have been made to improve these results by dosing LMWH adjusted for thromboelastography, conclusive data have not yet been obtained^[126].

Other preventive measures to avoid DVT and PE events involve the use of mechanical compression. These systems, as well as being of unproven efficacy, may be impossible to use in certain patients with trauma or who require surgery of the lower limbs, particularly if ExFix has been applied. Even so, they are in common use and are usually associated with pharmacological AP^[120]. Vena cava filters (VCF) have also been evaluated in this regard. This type of prophylaxis is proposed for extremely high risk patients in whom it is not feasible to perform AP by mechanical or pharmacological methods. In these patients, the use of VCF is aimed at achieving the prophylaxis of PE, as it does not prevent DVT^[127]; however, its use remains hotly debated^[128]. In summary, most current guidelines for AP advice the use of chemoprophylaxis with LMWH as soon as possible, associated with mechanical methods whenever feasible; the use of VCF is not yet recommended as routine prophylaxis.

ANTIBIOTIC PROPHYLAXIS

Infection is a frequent problem in polytraumatised cases, and sepsis is the second leading cause of death in these patients after haemorrhage. The prevention of infection is a complex matter. It is very difficult to establish protocols or guidelines in this regard, as patients often develop a disturbance of immunity secondary to trauma and their injury patterns vary greatly. Infection also depends on the type and severity of injuries presented, and many doubts arise concerning its treatment, in areas such as the time of administration, antibiotic selection and the duration of administration.

Nevertheless, it is generally accepted that the guidelines for antibiotic use do not change according to whether the patient being treated is severely traumatised. In patients with one or more open fractures, antibiotics should be administered at an early stage, and if possible within 3 h of the trauma^[129]. Strict measures to prevent

infections should be taken, with aseptic, rigorous management and care of wounds.

There is a consensus that the presence of multiple traumas does not justify changing the autologous blood-derived product regimen or prolonging it from that used in open fractures^[130,131]. Nonetheless, antibiotic doses in these patients should be individualised in accordance with the general organic function, since renal function impairment varies from one patient to another^[132].

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Observational Study

Inter- and intra-rater reliability of vertebral fracture classifications in the Swedish fracture register

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Institutional review board

statement: The regional Ethical Review Board in Stockholm, Sweden, approved the study, No. 2016/897-31/1. All patients were investigated and treated according to clinical guidelines. No interventions were made.

Informed consent statement:

Patient consent was not required for the retrieval of radiological images from the hospital. Verbal consent was obtained from the physicians that classified the images.

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Abstract**AIM**

To investigate the inter- and intra-rater reliability of the vertebral fracture classifications used in the Swedish fracture register.

METHODS

Radiological images of consecutive patients with cervical spine fractures ($n = 50$) were classified by 5 raters with different experience levels at two occasions. An identical process was performed with thoracolumbar fractures ($n = 50$). Cohen's kappa was used to calculate the inter- and intra-rater reliability.

RESULTS

The mean kappa coefficient for inter-rater reliability ranged between 0.54 and 0.79 for the cervical fracture classifications, between 0.51 and 0.72 for the thoracolumbar classifications (overall and for different sub classifications), and between 0.65 and 0.77 for the presence or absence of signs of ankylosing disorder in the fracture area. The mean kappa coefficient for intra-rater reliability ranged between 0.58 and 0.80 for the cervical fracture classifications, between 0.46 and 0.68 for the thoracolumbar fracture classifications (overall and for different sub classifications) and between 0.79 and 0.81 for the presence or absence of signs of ankylosing disorder in the fracture area.

Data sharing statement: Data can be provided on request by the corresponding author.

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CONCLUSION

The classifications used in the Swedish fracture register for vertebral fractures have an acceptable inter- and intra-rater reliability with a moderate strength of agreement.

Key words: Vertebral; Spine; Fracture; Classification; Swedish fracture register; Inter-rater; Intra-rater; Reliability

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Core tip: The Swedish Fracture register gathers national data on fractures. We adapted commonly used classifications for spine fractures and studied inter and intra-rater reliability as a basis for future usage of the register, including research on outcome after spine fractures.

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INTRODUCTION

One in six women and one in twelve men will at some point in their lifetime sustain a vertebral fracture^[1]. The current knowledge of the epidemiology of vertebral fractures in Sweden is based on the national patient register, which only classifies fractures based on large anatomical areas^[2,3]. Classification of vertebral fractures is important to describe epidemiology and allow treatment comparisons. By using large quality registers, we have the possibility to improve knowledge, increase our understanding of vertebral fracture epidemiology and possibly have an impact on future fracture treatment. The recently introduced Swedish Fracture Register offers such an opportunity^[4].

Most of the previous studies on inter-rater and/or intra-rater classification reliability have been performed in expert settings^[5-10], while fewer studies have been performed in more clinical settings with physicians of different experience levels^[11,12]. Registration of vertebral fractures was recently introduced in the Swedish Fracture Register. The reliability of the classification system has not been tested. Our aim was therefore to test the classification before large scale epidemiologic or observational studies in this register are attempted. Our hypotheses were that inter- and intra- rater reliability in this register are acceptable and similar to previously published studies.

MATERIALS AND METHODS

Fracture classification

Vertebral fractures have been possible to enter into the Swedish Fracture Register since 2015^[13]. The fracture classification is performed with the help of drawings that provide information about typical fracture patterns. Since classifications were to be performed not only by experts in spinal trauma care, but also by interns, residents and emergency physicians, the aim was to use classifications that was easy to comprehend without previous experience in the subject. Earlier published classifications were used as a foundation, but some of them were modified compared to their original descriptions. Appendix I (available as supplementary material) show the fracture classifications used in the Register.

Atlas (C1) fractures are classified according to Jackson^[14]. Axis (C2) fractures are classified according to Anderson-D'Alonzo^[15] for odontoid fractures and according to Effendi^[16] and Levine-Edwards^[17] for traumatic spondylolisthesis of the axis (hangman's fracture). Subaxial (C3-Th1) fractures are classified with a slightly modified version of the sub-axial cervical spine injury classification (SLIC)^[18]. Thoracic and lumbar (Th2-L5) fractures are classified with a modified version of the 2013 version of the "Arbeitsgemeinschaft für Osteosynthesefragen" (AO) spine injury

classification system^[19].

In addition, the classifying physician is also asked to determine whether there are signs of ankylosing spinal disorders in the fracture area, such as diffuse idiopathic skeletal hyperostosis (DISH) or ankylosing spondylitis (AS).

Study population

We identified consecutive patients with vertebral fractures, 50 with cervical spine fractures and 50 with thoracolumbar spine fractures, from the medical records at the Karolinska University Hospital Huddinge. It is the primary hospital for the southern part of Stockholm, but also contains one of two referral centers for surgical treatment of vertebral fractures in the Stockholm County, which has a total population of 2.3 million inhabitants. The radiological images (plane x-ray, computed tomography, and/or magnetic resonance imaging; MRI) were collected from the radiological archive. The mean age at the time of the radiological examination was 60 years (range 15-93) for patients with cervical fractures and 55 years (range 9-96) for patients with thoracolumbar fractures. Low-energy trauma (defined as a fall from standing height or less) was the cause for 23 (46%) of the cervical fractures and 20 (40%) of the thoracolumbar fractures. Non-surgical treatment was provided to 40 (80%) of the patients with cervical fractures and to 35 (70%) of the patients with thoracolumbar fractures.

Reliability tests

In all, six physicians of different experience level and one medical student were involved in the classifications; five of which were involved in each of the cervical and the thoracolumbar classifications with two of the physicians and the medical student involved in both classifications (Table 1). The raters only had information on patient's age and date of the investigation. No other clinical information or information about the fractures, such as radiological image evaluations, was provided to the raters. The raters classified the fractures independently without knowledge of the results of the others. The classifications were made with a document containing the Swedish Fracture Register fracture classification and subheadings for each fracture type (Appendix I).

Gold standard

After the classifications were completed, the results from the first test of the two most experienced raters, (Rater 1 and 2) were compared. In case a classification was not identical, Rater 1 and 2 reviewed the available radiological images (plain radiographs, computed tomography and/or magnetic resonance images) until consensus was achieved.

Statistics

The classifications made by the raters were analyzed using Cohen's kappa^[20] and are shown as kappa value (asymptotic standard error, standard deviation or range). Besides analyzing the result of all cervical and all thoracolumbar classifications, the classifications made by the raters were divided into subgroups analyzing only C1, C2 or C3-Th1 injuries in the cervical spine and A-, B- or C-types of injuries in the thoracolumbar spine (Th2-L5). All statistical analyzes were made in IBM SPSS Statistics for Windows, Version 24.0.

RESULTS

Gold standard

Rater 1 and 2 reached independent consensus in 21 patients with cervical fractures and 20 patients with thoracolumbar fractures. For presence or no presence of ankylosing spinal disorders, independent consensus was reached in 45 patients with cervical fractures and 46 patients with thoracolumbar fractures. After discussion, final consensus was reached for all fracture classifications and signs of ankylosing spinal disorder. The final distribution of fracture classifications is presented in Table 2. Not all classifications were represented.

Inter-rater reliability for vertebral fracture classifications

Inter-rater reliability mean kappa was 0.54 (0.26-0.70) for the cervical fracture classifications (C1-Th1) and 0.51 (0.18-0.69) for the thoracolumbar fracture classification (Th2-L5). Inter-rater reliability for individual raters is presented in Table 3.

For the cervical subgroups the inter-rater reliability was 0.79 (0.61-1.00) for C1 fractures, 0.76 (0.56-0.89) for C2 fractures and 0.68 (0.45-0.80) for C3-Th1 fractures.

Table 1 Rater experience level and time between the test occasions

	Experience level	Time between first and second test for cervical fractures	Time between first and second test for thoracolumbar fractures
Rater 1	Senior consultant in orthopaedics and spine surgery as well as responsible for the vertebral fracture classification in the Swedish Fracture Register.	1 mo	7 mo
Rater 2	Senior consultant in orthopaedics and spine surgery.	1 mo	7 mo
Rater 3	Specialist in orthopaedics.	7 mo	
Rater 4	Resident in orthopaedics.	7 mo	
Rater 5	Medical student, trained by Rater 1 to be knowledgeable on the topic.	1 mo	1 mo
Rater 6	Resident in orthopaedics.		1 yr
Rater 7	Resident in emergency medicine.		1 yr

Raters 1, 2 and 5 were actively working with classifications and vertebral fractures in the time between the tests while Raters 3, 4, 6 and 7 were not.

Inter-rater reliability for the thoracolumbar classifications was 0.72 (0.34-0.88) for A-type of injuries, 0.60 (0.41-0.76) for B-type injuries and 0.70 (0.51-0.89) for C-type of injuries.

Signs of ankylosing spinal disorder inter-rater reliability

For presence or absence of signs of ankylosing spinal disorder the inter-rater mean kappa coefficient was 0.65 (0.20-0.91) for cervical fractures and 0.77 (0.31-1.00) for thoracolumbar fractures. Inter-rater reliability for individual raters is presented in [Table 3](#).

Intra-rater reliability for vertebral fracture classifications

Intra-rater reliability mean kappa was 0.58 (0.40-0.72) for cervical fracture classifications (C1-Th1) and 0.46 (0.16-0.62) for the thoracolumbar fracture classification (Th2-L5). Intra-rater reliability for individual raters is presented in [Table 4](#).

For the cervical subgroups the intra-rater reliability was 0.80 (0.67-0.93) for C1 fractures, 0.78 (0.63-0.93) for C2 fractures and 0.69 (0.53-0.82) for C3-Th1 fractures. Intra-rater reliability mean kappa for the simplified thoracolumbar classifications was 0.63 (0.31-0.83) for only A-type of injuries, 0.56 (0.32-0.78) for only B-type injuries and 0.68 (0.47-0.90) for only C-type of injuries.

Signs of ankylosing spinal disorder intra-rater reliability

For presence or absence of signs of ankylosing spinal disorder the intra-rater mean kappa was 0.79 (0.66-1.00) for the cervical spine and 0.81 (0.60-1.00) for the thoracolumbar spine. Intra-rater reliability for individual raters is presented in [Table 4](#).

DISCUSSION

According to Landis and Koch^[21], the overall inter- and intra-rater reliability had a moderate strength of agreement for both cervical and thoracolumbar fracture classifications. When dividing the classifications into subgroups, both inter- and intra-rater reliability mean kappa coefficients increased. Inter- and intra-rater kappa coefficients were also generally higher for the more experienced physicians and the medical student who received training, which was most evident in the thoracolumbar classification resulting in the largest variability in inter-rater and intra-rater Kappa values. Similarly, our overall results for signs of ankylosing spinal disorder inter-rater reliability had a substantial strength of agreement while the intra-rater reliability have a substantial to almost perfect strength of agreement for vertebral fractures^[21].

Previous studies on fracture classifications used in the Swedish Fracture Register include humerus and tibia fracture classifications that have produced results similar to or better than our overall inter- and intra-rater reliability for the cervical and thoracolumbar classifications^[5,9]. Both studies used only one classification system and had a different study design. The humerus classifications had 12 possible alternatives

Table 2 Distribution of fractures according to the gold standard

			Number of patients	Number of patients with signs of ankylosing spinal disorder
Cervical spine	C1-Th1		50	6
	C1	Posterior arch	2	0
		Burst	5	1
		Lateral mass	1	0
	C2	Odontoid	11	1
		Hangman's	2	0
		Unclassifiable	4	0
	C3-Th1	Compression	13	3
		Burst	0	0
		Translation/rotation	7	2
		Other	11	0
Thoracolumbar spine	Th2-L5	Injury through axial compression (A-type injuries)	40	6
		Injury through anterior and posterior structures causing displacement (C-type injuries)	10	10
	Th2-L5 A-type injuries	Wedge shaped compressions	17	2
		Fracture through the middle part of the vertebral body	2	0
	Th2-L5 B-type injuries ¹	Burst fracture	21	4
		No injury of posterior structures	24	1
		Rupture of the posterior tension band structures through bone	9	4
		Rupture of the posterior ligament	5	0
	Th2-L5 C-type injuries	Unable to assess	2	1
		Hyperextension injury without translation	9	9
		Translation injury/dislocation through bone or disc/ligament	1	1

¹Classification of B-type injuries are only assessed in patients with A-type injuries. C: Cervical vertebra; L: Lumbar vertebra; Th: Thoracic vertebra.

while the tibia classifications had 27 possible alternatives for the raters to choose from^[5,9]. This can be compared to our study using 5 different classification systems with 3-14 possible alternatives in each separate classification system (in total 33 alternatives).

One study on C2 odontoid classifications and another study on the AO Spine subaxial cervical injury system (C3-7) have produced slightly poorer inter-rater reliability when compared to our subgroups (C2 and C3-Th1)^[7,8]. Previous studies on the AO Spine thoracolumbar spine injury classification system and Thoracolumbar Injury Classification and Severity Score (TLICS) have produced results that are generally better than our results for the Th2-L5 fracture classification^[6,11,19]. However, two more recently published studies concerning the AO Spine thoracolumbar spine injury classification system had poorer or similar inter-rater reliability compared to our study^[10,12]. The differences in results between studies are most likely due to differences in methods, study populations and the limitations in using Kappa, but we interpret our inter-rater results for vertebral fracture classifications used in the Swedish Fracture Register to be within what others have previously considered acceptable for a classification system^[5,7-10,12].

Our results for intra-rater reliability was at best similar to that of some previous studies^[5-10], while similar to or better than the results from a study with raters of

Table 3 Inter-rater reliability compared to the gold standard shown as kappa value (asymptotic standard error) for vertebral fracture classifications

		Kappa									
		Cervical spine					Thoracolumbar spine				
		C1-Th1	C1	C2	C3-Th1	Signs of ankylosing spinal disorder ¹	Th2-L5	Th2-L5 A-type injuries	Th2-L5 B-type injuries	Th2-L5 C-type injuries	Signs of ankylosing spinal disorder ¹
Test 1, Rater	1	0.59 (0.07)	0.61 (0.10)	0.81 (0.07)	0.78 (0.06)	0.81 (0.13)	0.64 (0.08)	0.88 (0.06)	0.70 (0.08)	0.72 (0.10)	0.91 (0.07)
	2	0.59 (0.07)	0.72 (0.10)	0.89 (0.06)	0.71 (0.07)	0.91 (0.09)	0.67 (0.07)	0.86 (0.06)	0.64 (0.08)	0.89 (0.08)	1.00 (0.00)
	3	0.51 (0.07)	0.79 (0.10)	0.66 (0.08)	0.66 (0.07)	0.67 (0.15)					
	4	0.47 (0.07)	0.86 (0.09)	0.74 (0.07)	0.61 (0.07)	0.32 (0.11)					
	5	0.59 (0.07)	0.93 (0.07)	0.74 (0.07)	0.73 (0.07)	0.77 (0.13)	0.44 (0.08)	0.67 (0.08)	0.63 (0.09)	0.57 (0.11)	0.91 (0.07)
	6						0.46 (0.08)	0.76 (0.08)	0.49 (0.09)	0.81 (0.09)	0.75 (0.10)
	7						0.45 (0.08)	0.67 (0.08)	0.48 (0.09)	0.61 (0.13)	0.31 (0.13)
Test 1 Mean		0.55 (0.06)	0.78 (0.12)	0.77 (0.08)	0.70 (0.07)	0.70 (0.23)	0.53 (0.11)	0.77 (0.10)	0.59 (0.10)	0.72 (0.13)	0.77 (0.27)
Test 2, Rater	1	0.70 (0.07)	0.93 (0.07)	0.81 (0.07)	0.80 (0.06)	0.81 (0.13)	0.58 (0.08)	0.82 (0.07)	0.76 (0.07)	0.77 (0.09)	0.91 (0.07)
	2	0.65 (0.07)	0.65 (0.10)	0.89 (0.06)	0.78 (0.06)	0.77 (0.13)	0.60 (0.08)	0.82 (0.07)	0.62 (0.08)	0.78 (0.10)	0.86 (0.08)
	3	0.26 (0.06)	0.66 (0.09)	0.56 (0.07)	0.45 (0.07)	0.52 (0.14)					
	4	0.44 (0.07)	0.72 (0.10)	0.65 (0.08)	0.63 (0.07)	0.20 (0.08)					
	5	0.63 (0.07)	1.00 (0.00)	0.85 (0.07)	0.70 (0.07)	0.74 (0.15)	0.69 (0.07)	0.82 (0.07)	0.76 (0.07)	0.82 (0.10)	0.95 (0.05)
	6						0.35 (0.08)	0.58 (0.09)	0.46 (0.09)	0.51 (0.13)	0.70 (0.11)
	7						0.18 (0.07)	0.34 (0.09)	0.41 (0.10)	0.52 (0.14)	0.38 (0.13)
Test 2 Mean		0.54 (0.18)	0.79 (0.16)	0.75 (0.14)	0.67 (0.14)	0.61 (0.25)	0.48 (0.21)	0.62 (0.21)	0.60 (0.17)	0.68 (0.15)	0.76 (0.23)

For test 1 and 2 mean kappa (standard deviation) is shown. ¹Ankylosing spondylitis or diffuse idiopathic skeletal hyperostosis. C: Cervical vertebra; L: Lumbar vertebra; Th: Thoracic vertebra.

different experience levels^[12]. The extended time (up to a year) in between the two reliability tests in our study needs to be taken into consideration, as well as lack of specific training and lack of detailed data on injury and patient history. Most other studies have about 4-6 wk between tests^[5,7,9,10,12], with the longer times being 6-8 mo^[6,8]. Some studies have also provided more information about the patients to the raters such as; clinical information (patient history, injury mechanism, associated injuries, clinical examination and neurological status), operation notes, follow up information, fracture level or radiological image evaluations^[6,7,9-11,19]. Therefore, our study could be said to represent the minimum level, which could have affected both our inter- and intra-rater reliability results negatively compared to other studies that provide more information.

In the discussions to decide a gold standard some of the common problems that came up were disagreement whether the example image, description text or the assumed injury mechanism should take priority. Classifications that involve injury to ligaments also created uncertainty among the raters in cases in which no MRI were provided. Still MRI is generally not necessary for thoracolumbar fractures^[22] and is not always available for cervical fractures. From the results it is apparent that thoracolumbar B-type injuries may be the most difficult to classify (Table 2). This is not surprising since this corresponds to our clinical experience; to determine whether there is a rupture of the posterior ligament complex is not always easy.

It has also previously been suggested that descriptions of the mechanisms of injury and ligamentous injury should not be included in a spinal injury classification^[23]. Yet it must also be taken into the consideration that the classifications need to be clinically relevant and must be associated with relevant patient outcomes in context with specific fracture management plans^[23,24].

The subgroups and simplified classification with fewer alternatives have a higher mean kappa coefficient for both inter- and intra-rater reliability, the most obvious reason for this increase is the limitations in using Cohen's kappa as it does not take in consideration if the difference between two classifications is small or large. Another possible reason for the increase is that more choices make it harder to choose the right classification, especially if they are similar to each other. Improved descriptions could

Table 4 Intra-rater reliability for vertebral fracture classifications shown as kappa value (asymptotic standard error) for all raters. Raters are compared to themselves.

		Kappa										
		Cervical spine					Thoracolumbar spine					
		C1-Th1	C1	C2	C3-Th1	Signs of ankylosing spinal disorder ¹	Th2-L5	Th2-L5 A-type injuries	Th2-L5 B-type injuries	Th2-L5 C-type injuries	Signs of ankylosing spinal disorder ¹	
Rater		1	0.72 (0.07)	0.67 (0.10)	0.93 (0.05)	0.80 (0.06)	1.00 (0.00)	0.59 (0.08)	0.76 (0.08)	0.69 (0.08)	0.84 (0.09)	1.00 (0.00)
	2	0.62 (0.07)	0.72 (0.10)	0.93 (0.05)	0.75 (0.07)	0.70 (0.14)	0.62 (0.07)	0.83 (0.07)	0.64 (0.08)	0.90 (0.07)	0.86 (0.08)	
	3	0.40 (0.07)	0.86 (0.09)	0.63 (0.07)	0.55 (0.07)	0.66 (0.12)						
	4	0.44 (0.07)	0.79 (0.10)	0.64 (0.08)	0.53 (0.07)	0.72 (0.10)						
	5	0.72 (0.07)	0.93 (0.07)	0.78 (0.07)	0.82 (0.06)	0.85 (0.10)	0.60 (0.08)	0.75 (0.07)	0.78 (0.08)	0.69 (0.10)	0.95 (0.05)	
	6						0.32 (0.07)	0.52 (0.09)	0.37 (0.10)	0.49 (0.14)	0.60 (0.13)	
	7						0.16 (0.07)	0.31 (0.09)	0.32 (0.10)	0.47 (0.17)	0.63 (0.19)	

¹Ankylosing spondylitis or diffuse idiopathic skeletal hyperostosis. C: Cervical vertebra; L: Lumbar vertebra; Th: Thoracic vertebra.

possibly improve the classifications. Reduced number of classifications could result in greater agreement between observers, but will decrease details of fracture data. Implementing web based training/example videos could possibly increase the agreement between physicians.

Strength and limitations

There are currently no methodological standard^[25] and we chose to use Cohen's kappa in our study as it is commonly used in other similar studies^[5,8,9,12,25]. A clinical setting with raters of different experience levels classifying vertebral fractures is similar to how data collection of vertebral fractures to the Swedish Fracture Register works and thus, this study provides us with an insight on the reliability of the data in the Swedish Fracture Register. Unfortunately this also make comparisons to other studies harder as different methods, study populations, classification systems and/or comparisons for the classification system is used^[6,7,11,19]. From this, we can assess that our results are not necessarily applicable to other registers or studies using one or more of the five individual classification systems used in the Swedish Fracture Register. The study population in this study cannot be generalized for the entire population or every type of hospital. It can also be argued that the study population in this study is too small for the cervical spine and that 50 patients should be selected for each classification system within the cervical spine in order to get a better understanding of the inter- and intra-rater reliability for these individual classification systems. Nevertheless, it would be hard to find large numbers of cervical fractures for a similar study, especially C1 fractures, which only represents 2% of all vertebral fractures^[4] but could be the focus for future studies.

Conclusions

The classifications used in the Swedish Fracture Register for vertebral fractures have an acceptable inter- and intra-rater reliability with a moderate strength of agreement. With specific training or many years of experience, a higher consistency in inter-rater reliability could be achieved. The results indicate that the Swedish Fracture Register data may be used for studies on epidemiology. Studies comparing treatment outcome should consider reclassifying images to ensure correct classification.

ARTICLE HIGHLIGHTS

Research background

The Swedish Fracture register gives the possibility to attain nationwide data on fractures.

Research motivation

Classification of vertebral fractures have recently been introduced in the register.

Research objectives

We tested the inter- and intra-rater reliability of the vertebral fracture classifications in the

Swedish Fracture register.

Research methods

Radiological images of consecutive patients with vertebral fractures were classified by 5 raters with different experience levels at two occasions.

Research results

The mean kappa coefficient for inter-rater reliability ranged between 0.51 and 0.79 for the different classifications (overall and for different sub classifications). The mean kappa coefficient for intra-rater reliability ranged between 0.46 and 0.81.

Research conclusions

The classifications have an acceptable inter- and intra-rater reliability with a moderate strength of agreement.

Research perspectives

The results indicate that the Swedish Fracture register is ready for use in epidemiological studies. Studies comparing treatment outcome should consider reclassifying images to ensure correct classification.

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Observational Study

Validation of the Danish version of the musculoskeletal tumour society score questionnaire

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Author contributions: Saebye CKP collected the majority of the data; Saebye CKP, Keller J and Baad-Hansen T designed the study and analyzed the data; Saebye CKP, Keller J and Baad-Hansen T wrote the manuscript.

Institutional review board

statement: The study was preapproved in accordance with the national ethical guidelines, since this type of study does not require approval in Denmark, furthermore the study is in accordance with the Helsinki Declaration.

Informed consent statement: All patients gave their verbally informed consent for participating in the study, however signed consent was not required according to Danish guidelines, since the study was based on questionnaires.

Conflict-of-interest statement: All authors declare: no support from any organization for the submitted work; no financial relationships with any organizations that might have an interest in the submitted work in the previous three years; no other relationships or activities that could appear to have influenced the submitted work.

STROBE statement: The authors have read the STROBE Statement-checklist of items, and the

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Abstract

BACKGROUND

The musculoskeletal tumour society score (MSTS) is a well-known questionnaire for measuring functional outcome in patients with neoplasms in the extremities. Standardized guidelines for cross-cultural translation and validation ensure the equivalence of content between the original and translated versions. The translation and validation provide the possibility to compare different sarcoma populations on an international level. This study is based on the hypothesis that the Danish MSTS questionnaire is a valid tool for measuring the end result after surgery for neoplasms in the extremities.

AIM

To validate the Danish version of the upper and lower extremity version of the MSTS.

METHODS

The translation of the MSTS was conducted in accordance with international guidelines. Patients operated for sarcomas and aggressive benign tumors were invited to participate in the study. The psychometric properties of the Danish version of the MSTS were tested in terms of validity and reliability and for the risk of floor or ceiling effect. Spearman's rank coefficient was used to test the validity by comparing with the Toronto Extremity Salvage Score (TESS). The Intraclass Correlation Coefficient (ICC) was used to evaluate inter-rater reliability. Cronbach's alpha was used to test for internal consistency. Spearman's rank coefficient was used to compare the MSTS lower extremity version with the objective test, Timed Up and Go (TUG).

RESULTS

The upper extremity version demonstrated an ICC of 0.95 in the inter-rater reliability test. The lower extremity version had an ICC of 0.88 in the inter-rater reliability test, respectively. Both MSTS versions showed a ceiling effect. The

manuscript was prepared and revised according to the STROBE Statement-checklist of items.

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validity of the MSTS was measured by Spearman's rank correlation coefficient by comparing the MSTS with the TESS and found it to be of 0.80 ($P < 0.01$) and 0.83 ($P < 0.01$) for the upper extremity and lower extremity version, respectively. A Spearman's rank correlation coefficient of -0.26 ($P < 0.01$) was found between the TUG and the MSTS questionnaire. A Spearman's rank correlation coefficient of -0.38 ($P < 0.01$) was found between the TUG and the lower extremity version of the TESS questionnaire.

CONCLUSION

The Danish version of the MSTS questionnaires were found to have good reliability and validity, however a substantial ceiling effect was identified.

Key words: Sarcoma; Patient outcome assessment; Clinical oncology; Surgical oncology; Patient satisfaction

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Core tip: The Danish version of the musculoskeletal tumour society score (MSTS) was found to have an overall good reliability and validity, however a substantial ceiling effect as well as a possibility of measurement error in the MSTS score was found. These findings must be taken into consideration when using the MSTS questionnaire.

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INTRODUCTION

The Musculoskeletal Tumour Society Score (MSTS) questionnaire was developed in 1985 and revised in 1993 as a physician-completed questionnaire to measure functional outcome in patients with neoplasms^[1]. The MSTS has been widely used in sarcoma research^[2-6]. However, the English version of the MSTS has never been properly validated^[1]. The lower extremity version of the MSTS has been translated and validated into Brazilian Portuguese, Chinese and Japanese^[7-9]. To our knowledge, the MSTS has never been properly translated and validated for Danish-speaking patients.

Guillemin *et al*^[10] were some of the first to construct a standardized guideline for cross-cultural translation. The cross-cultural translation is intended to ensure the equivalency of content between the original and translated version. Equivalency is achieved by ensuring not only correct linguistic translation but also cultural adaptation. Others have since created recommendations regarding ways of assessing the psychometric properties of such an instrument^[11,12].

The Timed Up and Go (TUG) test was introduced in 1991 as measure of dynamic balance and basic mobility skills needed for daily living^[13]. It has been shown to have good validity and reliability in lower extremity patients who have undergone unilateral amputation^[13]. The TUG has not been properly validated for use in sarcoma patients who have undergone limb-sparing surgery, although Marchese *et al*^[14] have validated the TUG as part of a larger functional outcome assessment method in sarcoma patients.

Only few studies have investigated the correlation between objective measurements and questionnaires, such as the MSTS^[14,15]. However, Marchese *et al*^[14] found a fair to moderate correlation between the TUG and the MSTS and TESS questionnaires.

In order to compare Danish sarcoma patients' functional outcomes internationally, the aims of this study have been: (1) to validate the psychometric properties of the Danish translation of the lower and upper extremity version of the MSTS questionnaire; and (2) to investigate the correlation between functional outcomes as measured by questionnaires, such as the MSTS and the Toronto Extremity Salvage Score (TESS), and the objective measurement, TUG, in patients with lower extremity tumors.

MATERIALS AND METHODS

Study design

The translation of the MSTS questionnaire into Danish was conducted at the Department of Orthopedic Surgery at Aarhus University Hospital between May and August 2015. The validation of the Danish translation was carried out among patients operated for sarcoma or aggressive benign tumors who attended the outpatient clinic at Aarhus University Hospital (Aarhus, Denmark) between August 2015 and June 2016. The study was reported to and approved by the Danish Data Protection Agency (file No. 1-16-02-650-15). Informed consent was obtained from all patients participating in this study. The study was preapproved in accordance with the national ethical guidelines and in accordance with the Helsinki Declaration. The translation method used was based on published international guidelines for the process of cross-cultural translation of an instrument^[10,16]. The cross-cultural translation and validation consists of several stages.

Translation

Stage I: Forward-translation: Two independent translators translated the upper and lower extremity version of the MSTS questionnaire (including the instructions to the user) from the original English version. The two independent translators were fluent in English and Danish but had Danish as their native language (both held diplomas in English and one was also a linguist). The two translators had different backgrounds in order to achieve the best possible translation. The first translator was a physician with clinical experience and was therefore considered an “informed” translator. The second translator had no clinical experience or relation to health care and was therefore considered a “naive” translator.

Stage II: Synthesis of a combined translation: The two translations were compared, and any discrepancies were discussed and resolved by the two forward-translators. A combined translation was finally made from the original English version and the two independent translations.

Stage III: Backward-translation: Two new independent translators conducted a backward-translation based upon the combined translation. They were blinded for the initial two forward-translations and the original English version. Both backward-translators were fluent in English (both held diplomas in English) and had Danish as their native language. The first backward-translator was a highly experienced researcher in health care, however with no prior knowledge to the concepts of the MSTS questionnaire. The second translator had no prior clinical experience or relations to health care.

Stage IV: Committee: The authors of this paper reviewed all the translated versions and components of the questionnaire, and the discrepancies were discussed following consensus concerning the final wording and formatting of the Danish version of the MSTS questionnaire.

The validation process

The validation was designed as a cross-sectional design, requiring physicians to complete the MSTS questionnaire and patients to complete the TESS. In case patients had been operated in the lower extremities, they also completed the TUG test.

Study population

All patients, age 18 or above, who had undergone surgical treatment for sarcomas or aggressive benign tumors in the extremities, were consecutively asked to participate in the study when attending the outpatient clinic. All patients were required to read and speak Danish to be able to participate. Patients were excluded if they had competing diseases affecting their physical function. A total of 240 out of 267 were included in the study.

Measurements

The MSTS is based on factors related to the patient as a whole and of those specific to the upper or lower extremity. It consists of six items of which the first three are identical in both MSTS versions: Pain, daily function and emotional acceptance. The upper extremity version also encompasses items measuring hand positioning, dexterity and lifting ability. As for the lower extremity version, this consists of items measuring the use of aids, walking ability and gait^[1]. Each of the aforementioned items is assigned a value of 0 to 5 points, and the final score is calculated as a percentage of the maximum obtainable score. The original English version of the MSTS was never tested for psychometric properties. However, the lower extremity

version was translated and validated into Brazilian Portuguese, Chinese and Japanese^[7-9].

The TESS assesses functional outcome in musculoskeletal tumor patients aged 12-85 years^[17]. The TESS consists of upper and lower extremity versions which have 29 items and 30 items, respectively. The final TESS score ranges between 0 and 100. The TUG test measures the time needed to stand up from a chair, walk 3 meters, turn around, walk back and sit back down in the chair^[13]. A stopwatch was used to document the time used.

Analysis of the data

Data was analyzed in Stata, version 12.1. Descriptive statistics were used for the patients' clinical demographic. All variables were examined to ascertain data distribution.

The psychometric properties were evaluated by assessing different domains, such as reliability and validity, as well as the estimate for possible floor and ceiling effects. Furthermore, the patients were stratified into groups according to tumor types.

The domain of reliability could be further divided into subdomains such as internal consistency, reliability and measurement error. The internal consistency was measured by Cronbach's α . A Cronbach's α between 0.70 and 0.95 was considered good^[18]. The inter-rater test was conducted by having two different physicians independently complete the MSTS questionnaire in the outpatient clinic. The inter-rater reliability was measured by intraclass correlations coefficient (ICC). The measurement error of the MSTS questionnaire was assessed by Bland-Altman plots in the inter-rater test^[19].

In this study, the construct validity was assessed by comparing the MSTS score with the TESS score^[18]. This was evaluated by either the Pearson's r or the Spearman's rank correlation coefficient, depending on the distribution of data. Floor and ceiling effects were considered present if > 15% of the patients received the lowest or highest possible score, respectively^[18].

The correlation between functional outcome measured by questionnaires (MSTS and TESS) and an objective measurement (TUG) was found by calculating correlation coefficients between MSTS and TUG as well as TESS and TUG by using either Pearson's r or Spearman's rank correlation coefficient, depending on the distribution of data.

RESULTS

Translation

The results showed an overall high consistency between the two forward-translations and the two back-translations when compared with the original English version. Only the item concerning emotional acceptance was found to be slightly different due to the differences in how Danish people express their feelings concerning their health and the treatment. In the original English version words such as "enthusiastic" and "like it" are used for describing the feelings of the surgical treatment, however a Danish patient might find this wording culturally strange for describing the feelings of a cancer diagnosis and the treatment of this. The Danish version does however take this into account, hence the emotional acceptance item was still found appropriate.

Validation

The demographic data concerning the patients is listed in [Table 1](#). The participation rate was 89.9%. [Table 2](#) describes the reasons for the 27 patients not participating in the study. The median MSTS scores for upper and lower extremity versions were 93 (Interquartile range (IQR): 80-100) and 87 (IQR: 73-97), respectively. The median TESS scores for upper and lower extremity versions were 98 (IQR: 83-100) and 93 (IQR: 81-98), respectively. The median TUG time (in seconds) was 6.4 (IQR: 5.4-8.0).

The test for internal consistency resulted in a Cronbach's α of 0.85 for the upper extremity version and 0.79 for the lower extremity version. The inter-rater reliability was also measured by the ICC and was found to be 0.95 (95%CI: 0.92-0.97) for the upper extremity version and 0.88 (95%CI: 0.84-0.91) for the lower extremity version. [Figures 1](#) and [2](#) present the limits of agreement in a Bland-Altman plot for the upper extremity versions and the lower extremity versions, respectively.

The analysis for construct validity found a Spearman's rank correlation coefficient of 0.80 ($P < 0.01$) and 0.83 ($P < 0.01$) for the upper and lower extremity versions, respectively, between the MSTS and the TESS.

[Table 3](#) presents the floor and ceiling effects found in the MSTS questionnaire. A Spearman's rank correlation coefficient of -0.26 ($P < 0.01$) was found between the

Table 1 Baseline characteristics of the patients

Characteristics	Patients (n = 240)
Age (mean)	52.6 ± 18.4
Range in age	18-89
Years since surgery (median)	2.2
Range in years	0.5-8.1
Gender	
Male	125
Female	115
Location	
Upper extremity	78
Lower extremity	162
Type	
Soft tissue sarcoma	149
Bone sarcoma	63
Aggressive benign tumor	28

TUG and the lower extremity version of the MSTS questionnaire. **Figure 3** illustrates the correlation between the TUG and the MSTS. A Spearman's rank correlation coefficient of - 0.38 ($P < 0.01$) was found between the TUG and the lower extremity version of the TESS questionnaire. **Figure 4** illustrates the correlation between the TUG and the TESS.

DISCUSSION

This Danish translation of the MSTS questionnaire was found to have good internal consistency, reliability and construct validity. However, the MSTS does have limitations as shown by the identification of a ceiling effect and possible measurement error between raters. In addition, poor correlations were found between the MSTS/TESS and the TUG. When using an existing measurement, it is important that it has undergone a proper cross-cultural translation in order to ensure that it measures the same concept as the original measurement^[10,16]. We have used well-known standardized guidelines to translate and validate the MSTS questionnaire into Danish^[10,16,18]. The lower extremity version of the MSTS questionnaire has also been translated and validated into Brazilian Portuguese, Chinese and Japanese according to these guidelines^[7-9]. To our knowledge, it is the first time the upper extremity version of the MSTS questionnaire has been translated into a foreign language following a standardized guideline. In the Japanese upper extremity version of the MSTS questionnaire, Wada *et al*^[20] tested the construct validity, but it was not mentioned whether that version had undergone systematic cross-cultural translation. Lee *et al*^[21] also reported the validity and reliability of both versions of the Korean version of the MSTS questionnaire, but also without reporting if the translation into Korean had been done according to the standardized guidelines.

The original validation of the English version of the MSTS unfortunately did not report a Cronbach's α ^[1]. The good internal consistency found in this study is however comparable to those found by Rebolledo *et al*^[7], Xu *et al*^[9] and Iwata *et al*^[8]. The inter-rater reliability also showed excellent results for both versions, and is in accordance with those found by Rebolledo *et al*^[7] and Xu *et al*^[9]. The original validation of the MSTS questionnaire also reported good inter-observer reliability, although no correlation coefficient was reported^[1]. **Figures 1 and 2** show low mean bias on all plots, however the limits of agreement are wide, which indicates a possible high measurement error. No previous studies have tested the measurement error in the MSTS^[1,7,8,20,21]. The test for measurement error is an important part of the validation process, since only a change in the MSTS score larger than the measurement error can be considered a possible 'real' change in the functional outcome^[22].

The construct validity of the MSTS has been determined as good in this study by comparing the MSTS with the TESS. This can be compared with similar results found by Rebolledo *et al*^[7], Xu *et al*^[9] and Iwata *et al*^[8]. Wada *et al*^[20] found a good correlation between the upper extremity version of the MSTS and the disability of the arm, shoulder and hand questionnaire.

Table 2 Reasons for exclusion from the study

	n (%)
Did not wish to participate	10 (37)
Incomplete questionnaire	9 (33)
Could not read Danish	4 (15)
Competing disease affecting function	3 (11)

A general ceiling effect was found in both the upper and lower extremity versions of the MSTS. However, when stratified there was no ceiling effect in patients with lower extremity bone sarcoma (8.9%, $n = 45$) or aggressive benign tumors (11.1%, $n = 18$) (Table 3). These results are similar to those reported by Rebolledo *et al*^[7] (7.4%, $n = 67$) in the lower extremity version of the MSTS, while the finding of a substantial ceiling effect in the pooled data is consistent with the results found by Iwata *et al*^[8] (23%, $n = 100$) and Wada *et al*^[20]. A study by Tanaka *et al*^[15] with the aim of predicting the knee extension strength and post-operative function has also shown a noticeable ceiling effect in the MSTS questionnaire (22.2%, $n = 18$). These results question the role of the MSTS in evaluating function in all musculoskeletal tumor patients, as a ceiling effect results in difficulties distinguishing between patients with superior function. Against this backdrop, it is important to consider the future role of the MSTS. A possibility could be to further develop this questionnaire to make it more appropriate for measuring physical function, or perhaps abandon this instrument entirely and instead develop a new and more precise one.

The current mainstay treatment of musculoskeletal tumors in the extremities directly influences the musculoskeletal system which accentuates the importance of an instrument that measures functional outcome precisely. We found a poor correlation between the TUG and the lower extremity version of the MSTS and the TESS. Marchese *et al*^[14,23,24] also found a generally poor correlation between the TUG and the MSTS/TESS in three studies, while Tanaka *et al*^[15] found a moderate to good correlation between the MSTS/TESS and the extension strength of the knee. This highlights the importance of choosing the correct instrument for measuring the desired concept of function. The purpose of the TUG is to measure the balance and mobility skills needed for daily living^[13], while the purpose of the TESS is to measure the patients' perception of function^[17]. In this way, two various subconstructs of the concept of function are measured. Although both can be of an importance, in exploring a hypothesis they may differ in significance.

A great strength of this study is the number of participating patients. This study included 78 patients with upper extremity tumors and 162 patients with lower extremity tumors. Previous guidelines concerning the validation of instruments have set a minimum of 100 patients as an excellent sample size, while 50 to 99 patients constitutes a good sample size^[25].

This study also has a main limitation, *i.e.*, the presence of the possibility of selection bias, as only the patients attending the outpatient clinic were asked to participate in the study. Patients with progressive disease and patients who were not satisfied with their treatment were less likely to attend the outpatient clinic.

In conclusion, the Danish versions of the upper and lower extremity MSTS questionnaires were found to have good reliability and validity. The Danish versions are comparable to the other translated MSTS questionnaires. It is however of concern that a ceiling effect was found in both versions. When using the MSTS questionnaire, it is important to take into account which concept of function is intended to be measured.

Table 3 Floor and ceiling effects of the musculoskeletal tumour society score questionnaire

	<i>n</i>	Percent floor	Percent ceiling
All patients			
Upper extremity version	78	0%	38.5%
Lower extremity version	162	0%	23.5%
Bone sarcoma patients			
Upper extremity version	18	0%	27.8%
Lower extremity version	45	0%	8.9%
Soft tissue sarcomas patients			
Upper extremity version	50	0%	44%
Lower extremity version	99	0%	32.3%
Aggressive benign tumor patients			
Upper extremity version	10	0%	30%
Lower extremity version	18	0%	11.1%

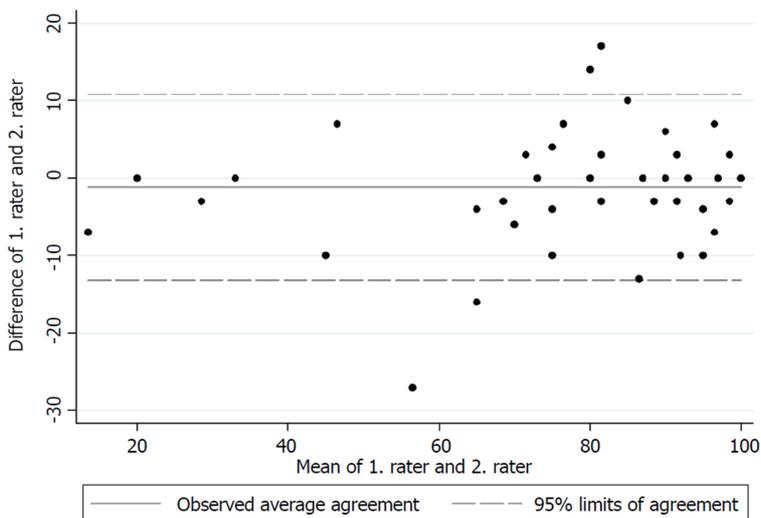


Figure 1 A Bland-Altman plot for the upper extremity version between raters.

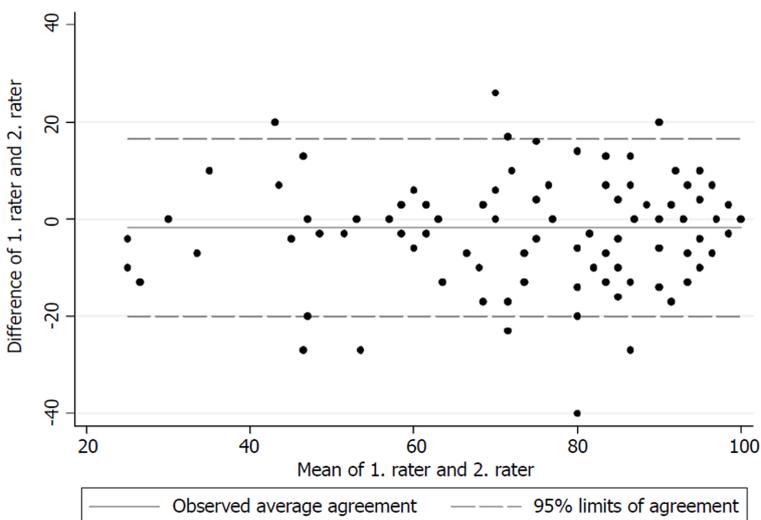


Figure 2 A Bland-Altman plot for the lower extremity version between raters.

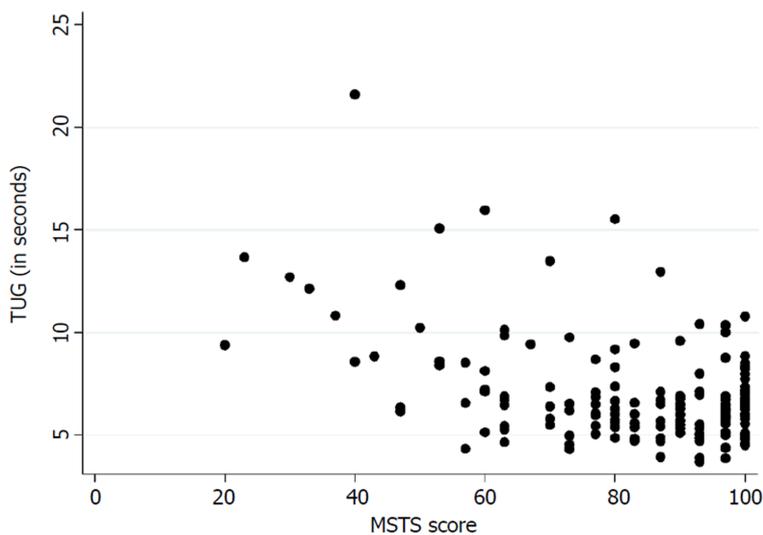


Figure 3 Correlation between musculoskeletal tumour society score and Timed Up and Go. MSTS: Musculoskeletal tumour society score; TUG: Timed Up and Go.

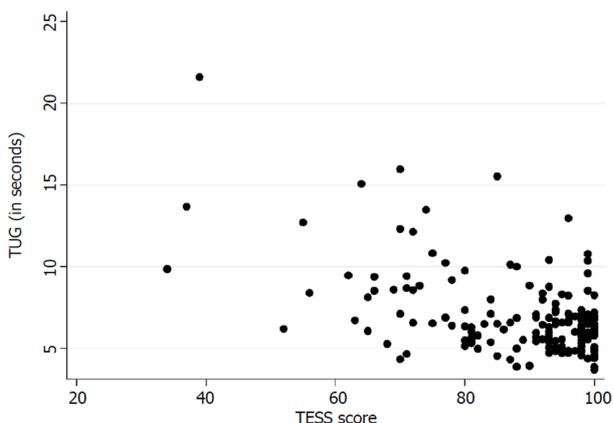


Figure 4 Correlation between Toronto Extremity Salvage Score and Timed Up and Go. TESS: Toronto Extremity Salvage Score; TUG: Timed Up and Go.

ARTICLE HIGHLIGHTS

Research background

The musculoskeletal tumour society score (MSTS) questionnaire is a physician/patient-completed questionnaire designed to assess functional outcome for patients with sarcomas in the extremities. The MSTS questionnaire was originally developed in English. Over the past decades there has been increased focus on the aptness of questionnaires to measure correctly. This also includes the aptness of questionnaires after being translated from one language to another.

Research motivation

To ensure that the Danish version of the MSTS questionnaire measures the same aspects of functional outcome in sarcoma patients as the English version, it is important to validate the measurement properties of the Danish version of the MSTS questionnaire and compare it to other language versions of the questionnaire. Furthermore, cultural differences need to be considered during the translation process, as this is a part of ensuring the original measurement properties. This rigorous process provides the possibility to compare results from national studies with other international studies.

Research objectives

The objectives of this study were: (1) to validate the Danish version of the MSTS questionnaire; and (2) to investigate the correlation between functional outcomes as measured by questionnaires, such as the MSTS, and the objective measurement, Timed Up and Go (TUG).

Research methods

The translation of the MSTS was conducted in accordance with international guidelines. Patients, age 18 or above, operated for sarcomas and aggressive benign tumors were consecutively invited

to participate in the study. The psychometric properties of the Danish version of the MSTS were tested in terms of validity and reliability and for the risk of floor or ceiling effects. Spearman's rank coefficient was used to compare the MSTS lower extremity version with the objective test, TUG.

Research results

The upper extremity version of the MSTS questionnaire demonstrated an excellent intra- and inter-rater reliability. The lower extremity version of the MSTS questionnaire showed an excellent intra- and inter-rater reliability. A ceiling effect, however, was found in both versions. Both versions of MSTS questionnaire were shown to have good validity. The MSTS questionnaire showed a possible presence of a measurement error. A poor correlation was found between the objective measurement, TUG, and the functional outcome measured by questionnaires.

Research conclusions

The Danish version of the MSTS questionnaire was found to have good reliability and validity, however a substantial ceiling effect as well as the possibility of measurement error were identified. The Danish version of the MSTS questionnaire can be used to measure functional outcome in sarcoma patients and to compare these results with other international studies.

Research perspectives

The measurement errors and ceiling effects are concerns which are not to be overlooked. It is highly recommendable to further investigate these issues and the measurement properties of the MSTS questionnaires, such as its aptness in detecting significant clinical changes in the functional outcome.

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Contralateral trapezius transfer to treat scapular winging: A case report and review of literature

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Institutional review board

statement: The Institutional Review Board (IRB) of Hospital Universitario Virgen del Rocío gave approval for the study, No. JGA-TCEA-2017.

Informed consent statement:

Written informed consent was given by the participant before the beginning of the study.

Conflict-of-interest statement: The authors of the study have no potential conflicts of interest to declare.

CARE Checklist (2016) statement:

The guidelines of the "CARE Checklist - 2016: Information for writing a case report" have been adopted.

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Abstract

BACKGROUND

No dynamic technique, such as tendon transfer, has been described for scapular winging due to levator scapulae or rhomboid major and minor palsies resulting from an isolated dorsal scapular nerve injury. Thus, we evaluated how the contralateral trapezius compound osteomuscular flap transfer would work in stabilizing lateral scapular winging, and the case is reported here. A literature review was also conducted, and articles relevant to the case are presented.

CASE SUMMARY

A 37-year-old male patient who had sustained an isolated dorsal scapular nerve injury underwent reconstructive surgery using the contralateral trapezius compound osteomuscular flap transfer technique to treat scapular winging and the consequent pain, and to restore function from the shoulder impairment. As a result, the involved shoulder showed an improved Constant-Murley score, from 19.5% to 81.88%.

CONCLUSION

Contralateral trapezius osteomuscular flap transfer succeeded in stabilizing scapular winging in this case, improving shoulder function and affording pain relief.

Key words: Trapezoid muscle; Osteomuscular flap; Scapular winging; Rhomboid muscles; Nerve paralysis; Dorsal scapular nerve; Case report

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Core tip: This report describes a case of scapular winging as a result of dorsal scapular nerve injury. Only few cases of winged scapula due to rhomboid paralysis are reported in

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the literature. Although scapular winging can seriously impair shoulder function, no dynamic techniques have been described to treat this condition. This case reveals that contralateral trapezius compound osteomuscular flap transfer can successfully stabilize lateral scapular winging due to a dorsal scapular nerve injury, while improving shoulder function and affording pain relief.

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INTRODUCTION

Winging of the scapula was first described over 200 years ago^[1]. It remains a cause of serious impairment of upper limb function, decreasing shoulder range of motion and strength; ultimately, it affects activities such as lifting, pushing and pulling heavy objects^[2], not to mention the pain associated with doing so^[3,4]. In addition to these functional restrictions, it affects the shape of the back and can become an aesthetic and psychological issue^[5].

To understand this pathology, the anatomy of the shoulder must be fully known. The minor rhomboid muscle originates from the cervical spinous processes 6-7 and the major rhomboid muscle from the thoracic spinous processes 1-4. The major and minor rhomboid muscles are both innervated by the dorsal scapular nerve which originates from C5 or C5 and C6^[6], inserts at the scapula medially, presses the scapula on the thorax and pulls it to the spine. This anatomy preconditions for retroversion and internal rotation in antagonization of the serratus anterior muscle which is innervated by the long thoracic nerve which originates from C5, C6 and C7^[7], and inserts at the scapula medially. This anatomy preconditions for anteversion and external rotation. With rhomboid muscle insufficiency, elevation of the arm is still possible but for full movement both preconditions have to be met.

The other involved muscles (function) in scapular girdle motion are: Pectoralis major muscle (anteversion, internal rotation); pectoralis minor muscle (adduction, internal rotation); coracobrachialis muscle (anteversion); deltoid muscle (abduction up to 90°, strongest anteversion with its clavicular part, internal rotation with its spinal part); subscapular muscle (strongest internal rotation); supraspinatus muscle (external rotation and additionally abduction); infraspinatus muscle (strongest external rotation); teres major muscle (strongest retroversion, internal rotation); teres minor muscle (external rotation); latissimus dorsi muscle (adduction, retroversion, internal rotation); and, trapezoid muscle (elevation/abduction of more than 90° with its descending part). The trapezoid muscle is innervated by the spinal accessory nerve, a cranial nerve which originates from the first five to six spine segments before entering the cranium through the foramen magnum (Ramus externus or Radices spinales) and is then accompanied by its cranial part that originates from the oblongata medulla (Ramus internus or Radices craniales). Both parts run together with the vagal nerve, through the jugular foramen, before innervating the sternocleidomastoid muscle and the trapezoid muscle. Contributions to trapezius innervation by the cervical plexus have also been described but findings thus far suggest it has limited significance^[8].

Many etiologies can cause scapular winging. Most cases, however, are attributed to neurological lesions. These include long thoracic, spinal accessory and dorsal scapular nerve injuries, which are linked to this problem due to paralysis of the serratus anterior, trapezius and/or rhomboid muscles, respectively. Normally, the synchronized actions of these muscles push the medial border of the scapula against the thoracic wall; dysfunction of any of them will consequently lift the medial border of the scapula away from the back. In any case, dorsal scapular nerve lesions are quite rare and the literature contains few case reports of winged scapula due to rhomboid paralysis in particular^[9-11].

The direction of displacement of the inferior angle of the scapula determines whether winging will be medial or lateral. The former is due to a lack of function of the serratus anterior muscle, and the latter to a lack of function of the trapezius and/or rhomboid muscles. Winging can be also classified as primary, secondary, or

voluntary. Causes of primary winging are neurological injuries, usually involving the long thoracic nerve, the accessory nerve or the dorsal scapular nerve. These occur in association with tumor on the ribs or scapula or malunited fractures of the scapula, as well as following the rupture or absence of periscapular muscles. Secondary winging is due to glenohumeral and subacromial disorders. Voluntary winging of the scapula is very rare and psychological factors play an important role^[12].

Scapular winging presents a diagnostic challenge, as evidenced by the many patients that are initially misdiagnosed^[13,14]. Electromyography of the serratus anterior, trapezius and rhomboid muscles is useful to inform the diagnosis and reveal which muscles are involved as well as the degree of denervation^[15,16]. Electromyographic testing typically shows resting denervation potentials, decreased motor unit recruitment, and polyphasic motor unit potentials during volitional activity^[17,18] if any recovery is taking place. Imaging studies are rarely diagnostic but should be performed to ascertain any structural abnormalities and aid in ruling out other diagnoses^[10,15,19,20].

Transfers of several muscles have been described to treat scapular winging. In this sense, transfer of the rhomboid major and minor has been suggested to treat scapular winging due to spinal accessory nerve injury and trapezius paralysis. Transfer of the pectoralis major is widely used when scapular winging is present following a long thoracic nerve injury and serratus anterior paralysis. To the best of our knowledge, there are no reports of any muscle transfer for primary scapular winging due to an isolated dorsal scapular nerve injury.

With written informed consent, we report herein the unusual case of an adult patient who developed scapular winging on the right side due to a dorsal scapular nerve injury. We chose to treat the case with contralateral trapezius transfer, and obtained approval from the local Ethics Committee (No. JGA-TCEA-2017). The surgical technique provided successful restoration of scapulothoracic joint function and pain relief, and may be a useful means by which to treat scapular winging due to an isolated dorsal scapular nerve injury.

CASE PRESENTATION

A 37-year-old male patient was referred to our clinic with a history of right shoulder indirect injury sustained 2 years prior (**Figure 1**). He worked carrying heavy marble stones. The injury had occurred when he was trying to prevent one from falling. He reported having made a sudden arm movement, followed by tremendous effort to hold the stone. Following the incident, the patient experienced right scapular winging, shoulder function impairment, and pain.

Clinical examination revealed scapular winging at rest and during both passive and active shoulder flexion (**Figure 2**), together with decreased active shoulder movement on the right side, having 60° flexion and 45° abduction (**Figure 3**). In terms of shoulder rotations, the patient was able to put his hands behind his head with elbows forward but was unable to reach his back. The patient underwent evaluation of overall function of the shoulder using the Constant-Murley score (CMS; a 100-point scale composed of several parameters that define the level of pain, function, range of motion and strength^[21]). The patient's preoperative CMS was 19.5.

Imaging studies (plain X-rays and magnetic resonance imaging) did not reveal any structural injury. Electrophysiological studies showed an isolated dorsal scapular nerve lesion and proper function of other nerves, such as the long thoracic and spinal nerves.

FINAL DIAGNOSIS

Dorsal scapular nerve injury.

TREATMENT

The case was considered to be an irreparable nerve lesion given the time elapsed between nerve injury and the patient's referral to our clinic. Nerve surgery was obviated and contralateral trapezius compound osteomuscular flap transfer was proposed to correct the scapular winging.

Surgery was performed under general anesthesia. The patient was placed in the prone position. Superficial anatomical landmarks were drawn, including scapular silhouette on both the recipient and donor sites, lateral edge of the trapezius muscle,

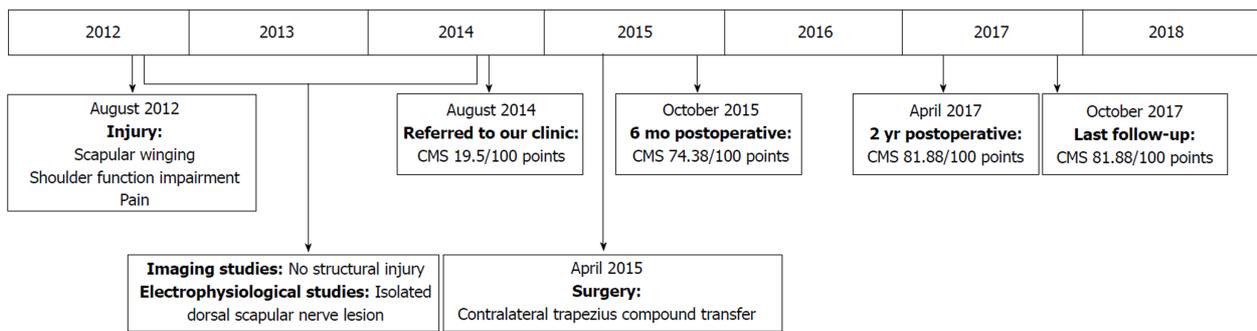


Figure 1 Case timeline. CMS: Constant-Murley score.

and spinal processes from T4 to T12. Centered on a line bisecting the angle formed between midline and the lateral edge of trapezius, an “s” shaped incision was made with the caudal flap designed to expose the lowest thoracic spinal processes. A second incision was made along the medial border of the scapula at the site of injury, at the level of the scapular spine (Figure 4). At the donor site, the trapezius flap was raised and freed from its bed, from lateral to medial, and detached from the spinal processes except from the last two (T11 and T12), which were osteotomized to include two hemispinal processes in the compound flap. Bone bleeding in the flap indicated bone viability (Figure 5). The trapezius flap was detached from its insertion at the spinal processes, from T4 to T10. Continuity of the posterior elements of the spine was not interrupted and posterior interspinous ligament was spared. The compound osteomuscular trapezius flap was raised and passed through a subcutaneous tunnel towards the second incision (Figure 6). Prior to attaching the flap to the recipient site, the shoulder of the injured site was pushed backward to bring the medial border of the scapula closer to the midline. In this position, we marked the sites where the two hemispinal processes of the compound flap were to be placed on the scapular spine with the flap under maximal tension. At these sites, we created two beds in the spine of the scapula so that bone-to-bone fusion could take place between them, providing a footprint for bone-to-tendon fusion. The flap was then attached using anchors in the beds and transosseous sutures through the scapular spine (Figure 7). Nonabsorbable sutures were used. Layered closure was performed and drainage was placed in the donor site. A diagram of the surgery is provided in Figure 8.

The scapulothoracic joint was controlled postoperatively with a brace (Actimove® Clavícula; BSN Medical Inc., Rutherford College, North Carolina, United States), with both shoulders positioned “at attention” to avoid flap elongation for 6 wk. Five days later, the patient was discharged from in-patient care. Skin sutures were removed after 2 wk. Subsequently, the patient first underwent daily check-up by the therapist for a period of 2 mo, then twice weekly for another 2 mo.

OUTCOME AND FOLLOW-UP

CMS was measured monthly using an automated dynamometer (microFET®2; Hoggan Scientific LLC, Salt Lake City, Utah, United States). The patient then learned to dissociate donor from recipient movement by performing recipient movement without consciously thinking about donor movement (Video 1).

The patient improved his CMS from day 1 preoperative (19.5%) to 74.38% at 6 mo postoperative and 81.88% at 2 year postoperative. No additional improvement or impairment has been reported up to the last follow-up revision (30 mo postoperatively).

The patient was asked to record his preoperative and postoperative pain using a 10-cm visual analogue scale (commonly known as VAS). The preoperative level of pain of the injured shoulder was 5 on a 0-10 VAS. Following surgery, this score fell to 3 at 6-mo follow-up, and it was 0 at 2-year follow-up. Soreness was reported at the donor site for 6-mo postoperative.

Shoulder motion was measured preoperatively and in the follow-ups. Preoperative shoulder flexion and abduction were 60° and 45°, respectively; external rotation allowed the patient to reach the occipital area with elbows forward but internal rotation was severely impaired, since the patient was unable to reach his back. Postoperatively, all active ranges of motion were measured, and at 6 mo the patient achieved maximum postoperative mobility of the shoulder. This was maintained



Figure 2 Scapular winging with right shoulder in passive flexion.

through to the last follow-up, with the patient having 155° flexion, 95° abduction, full elevation and ability to reach T12 on his back (Figure 9).

With respect to strength, the patient was able to lift up to 9.1 kg with no pain and shoulder flexed at 90° with the arm positioned in the axis of the scapula at the most recent measurement. No preoperative strength measurements were performed, since the patient was not able to achieve 90° of active flexion.

No impairment of the contralateral shoulder or scapular girdle was detected postoperatively. Scapulothoracic rhythm was not altered in the donor site, while it was improved in the recipient site.

DISCUSSION

We searched PubMed from January 1990 up to October 2018 using the search terms “dorsal scapular nerve”, “dorsoscapular nerve”, “nervus dorsalis scapulae”. Sixty-nine articles were retrieved, of which thirteen were deemed relevant and were used in this report. In addition, other topically relevant articles are discussed in order to support the treatment approach used on our patient; the treatment decision was based on the fact that, to the best of our knowledge, no dynamic technique (*i.e.*, tendon transfer) had yet been described for scapular winging due to rhomboid major and minor palsy resulting from an isolated dorsal scapular nerve injury.

From an etiological point of view, dorsal scapular nerve injury usually occurs in the context of compressive neuropathy. The compression of the nerve has been described in situations related to sports medicine. Ravindran *et al*^[22] presented the cases of 2 brothers who practiced volleyball at a high level, with both having developed a winged scapula as a consequence of neuropathies of the suprascapular nerve in association with neuropathy of the dorsal scapular nerve. The association of dorsal scapular nerve injury with suprascapular nerve injury has also been described by Lee *et al*^[23]. Also in sports medicine, Jerosch *et al*^[24] described the case of a judoka who, after suffering a dislocation of the glenohumeral joint, had a winged scapula due to injury to the long thoracic nerve and the dorsal scapular nerve.

There have also been reports of cases unrelated to sports medicine. Debeer *et al*^[25] reported a case of compression exerted by a poorly adjusted orthosis in a patient with scoliosis. In our case, the patient suffered a dorsal scapular nerve injury in the workplace, after making an effort to lift a heavy weight. On this same topic are the case reports from Argyriou *et al*^[26] (describing a patient with a winged scapula and work history of carrying about 8 kg of weight repetitively) and of Akgun *et al*^[9] (describing a patient who developed an acute winged scapula after lifting heavy objects above the head). For these two patients, the cause was a compressive neuropathy of the dorsal scapular nerve.

Iatrogenic lesions of the scapular dorsal nerve also occupy an important niche in this research field. A case reported by Lee *et al*^[27] developed as a consequence of a trigger point puncture in the rhomboid major muscle. Saporito^[28] concluded that there is potential risk of damaging the dorsal scapular nerve and long thoracic nerve in the anesthetic blocks of the brachial plexus in the interscalene region. This researcher then recommended ultrasonographic localization of both nerves to avoid injuring them; this idea was based on the possible ultrasound localization in most cases as published by Hanson *et al*^[29].

Regarding clinical manifestations, according to Muir *et al*^[30] neuropathy of the dorsal scapular nerve can cause winged scapula to varying degrees, depending on the



Figure 3 Decreased shoulder movement with 60° active flexion. Scapular winging is observed with shoulder flexion.

weakness of the major and minor rhomboid muscles. Sultan *et al*^[31] defined, in addition to the winged scapula, other symptoms related to entrapment of the dorsal scapular nerve, such as interscapular pain. These authors attributed such symptoms to sensitization of nociceptors of the nerve sheath; although, interscapular pain may also be a consequence of the tension affecting the cutaneous medial branches of the dorsal primary rami of thoracic spinal nerves, that develops when the winged scapula occurs. We believe that the latter reasoning fits the cause of pain in our patient best, given that in our case the pain presented by the patient before the operation had a value of 5 in the VAS and that after the surgery the pain decreased in intensity without the need of perform any type of intervention on the nerve, only treating the winged scapula.

Another symptom reported by Sultan *et al*^[31] is the loss of range of motion of the shoulder. In our case, the patient lost mobility in every arc of movement of the shoulder, which was recovered after the surgery. These features suggested to us that the loss of mobility was due to an alteration of the dynamics of the scapulothoracic joint and the unfavorable position by which the winged scapula affects all muscle groups of the scapular girdle. Although, in our patient, we did not observe the profile of symptomatology described by Sultan *et al*^[31], such as cervicogenic dorsalgia, notalgia paresthetica and posterolateral arm pain, possibly related to dorsal scapular nerve neuropathy. Chen *et al*^[32] have also reported discomfort and soreness of the neck, shoulder and back region too as part of the symptomatology.

Argyriou *et al*^[26] considered conservative treatment of dorsal scapular nerve neuropathy suitable for mild or moderate cases, as did Benedetti *et al*^[33] who obtained satisfactory results through a multidisciplinary rehabilitation approach. However, for those cases in which conservative treatment is not effective, Argyriou *et al*^[26] recommended surgical treatment. In this sense, Chen *et al*^[32] considered that the symptoms derived from the compression of the scapular dorsal nerve can be improved if the middle scalene muscle and its tendinous tissue are sectioned in relation to the compressed nerve.

Nevertheless, nerve acute injuries are difficult to treat, given that such lesions are often diagnosed late. Motor endplate atrophy leads to suboptimal results when reconstruction is performed more than 3-6 mo after the nerve damage occurs. However, even immediate restoration of original function does not always result in satisfactory outcomes^[34]. In these cases, tendinous transfers are widely used for the treatment of peripheral nerve injuries that are in the sequelae phase and otherwise not repairable. These dynamic procedures are currently the standard of care for restoration of shoulder function after chronic scapular winging from nerve palsy in younger, active people^[35].

To the best of our knowledge, no dynamic technique, such as tendon transfer, has been described for scapular winging due to rhomboid major and minor palsy resulting from an isolated dorsal scapular nerve injury. Medial static stabilization of

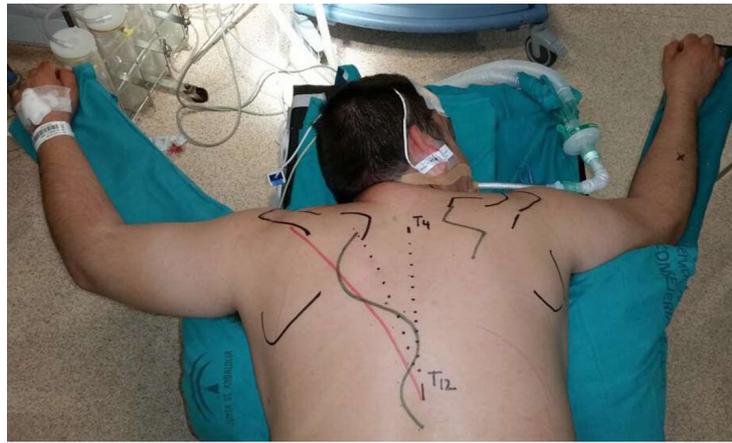


Figure 4 Superficial anatomical landmarks and incision planning.

the scapulothoracic joint has been performed using strips of fascia lata. This technique was described by Ketenjian in 1978^[36] and posteriorly modified by Atasoy and Majd^[37], who obtained good results. Scapulothoracic fusion has also been suggested, although disparate results have been reported after fusion with complications such as pneumothorax, pleural effusion, atelectasis, fracture of the scapula, and pseudarthrosis^[38].

Terzis *et al*^[39] found that contralateral low trapezius transfer was a useful technique to treat scapular winging in patients with brachial plexus birth palsy (known as BPBP). Scapular winging in children with sequelae of BPBP is due not only to weakness of periscapular musculature but also to glenohumeral dysplasia, which results from an imbalance in the internal and external rotators of the shoulder. This dysplasia also leads to abnormal scapulothoracic movement during shoulder motion, due to both the anatomical and structural alterations of the glenohumeral joint. Moreover, most patients who suffer from BPBP sustain postganglionic root lesions where dorsal scapular, long thoracic and spinal nerves are usually spared. In these cases, scapular winging also develops, revealing that this condition is caused mainly by shoulder rotator imbalance instead of weakness of the periscapular muscles.

As described by Elhassan *et al*^[40], contralateral trapezius transfer to the infraspinatus insertion appears feasible and has been used to restore external rotation of the shoulder by transferring to the infraspinatus insertion^[41]. Given that suture is performed to join two tendons, weakness at the junction should not be a main concern in these cases, as it is when tendons are transferred to bone. The best healing interface is obtained when repair is performed between homogeneous tissues (*i.e.*, bone-to-bone and tendon-to-tendon) and shows better healing quality with respect to mechanical and histological assessments, as compared to the healing between heterogeneous tissues (*i.e.*, bone-to-tendon)^[42]. Consequently, and given that at the recipient site muscle and bone are the only tissues available, we decided to take bone from the donor site to perform bone-to-bone fusion. Bone tissue was obtained without interrupting the continuity of the posterior elements of the spine, taking only the ipsilateral hemispinal processes and sparing the posterior interspinous ligament, not only in adjacent spinal processes (T10 and L1) but also in the affected ones (T11 and T12).

For our patient, scapulothoracic rhythm was preserved in the donor site and it was improved in the recipient site. Measurements were performed with visual inspection systems. Digital inclinometers have been recommended for quantifying shoulder and scapular kinematics^[43], and it is possible that these devices would have reported subtle changes in the donor site.

We have found that contralateral trapezius compound osteomuscular flap transfer was able to successfully stabilize lateral scapular winging due to an isolated dorsal scapular nerve injury in the case reported, while improving shoulder function and affording pain relief. Further studies are needed with a larger sample size to determine whether the contralateral trapezius compound osteomuscular flap transfer technique is the best choice for patients with scapular winging due to an isolated dorsal scapular nerve injury.

EXPERIENCES AND LESSONS

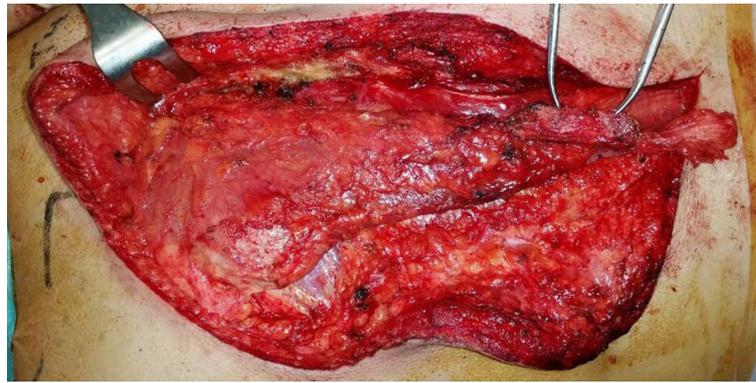


Figure 5 Bone tissues can be seen at the tips of the mosquito clamp coming from T11 and T12 spinal processes.

Whole shoulder girdle exploration is needed when the shoulder function is impaired, since dorsal scapular nerve injury may go unnoticed without a high degree of suspicion. The contralateral trapezius compound osteomuscular flap is a technique that we consider to be not especially complex if performed by surgeons with experience in shoulder and periscapular surgery. Given the good result obtained with this dynamic technique for scapular stabilization when the scapular winging was due to dorsal scapular nerve injury, we have abandoned the static techniques that were carried out previously and that yielded worse results with higher associated morbidity.

In our patient, no complications were observed at the donor site, except for soreness reported at the 6-mo postoperative follow up. The conclusion we have come to from our experience and literature review is that contralateral trapezius transfer is a safe technique, even when the two lowest hemispinal processes are included in the flap.



Figure 6 The compound osteomuscular trapezius flap was raised and passed through a subcutaneous tunnel (Penrose drain) to the second incision.



Figure 7 The flap was fixed using anchors in the beds and transosseous sutures through the scapular spine to attach the tendon to the footprint.

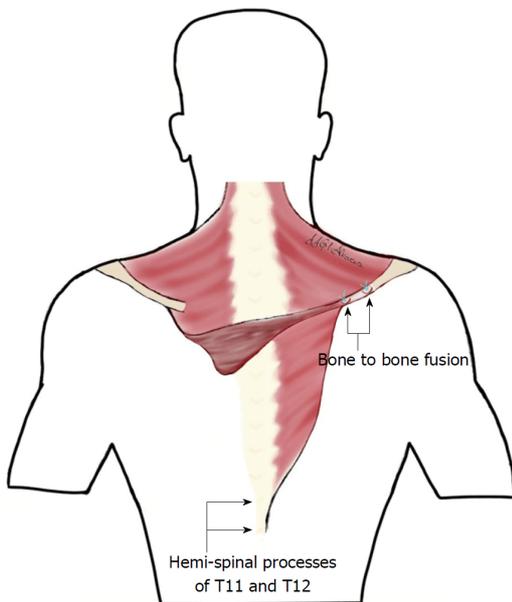


Figure 8 Diagram of the contralateral trapezius compound osteomuscular flap transfer.



Figure 9 Active range of motion at the 6-mo follow-up. A: Flexion (notice flap working); B: Abduction; C: External rotation; D: Internal rotation and normal lift-off test.

ARTICLE HIGHLIGHTS

Case characteristics

A 37-year-old male presented at 2 years after a right shoulder indirect injury, with right scapular winging, shoulder function impairment, and pain.

Clinical diagnosis

Physical examination of the patient suggested a lateral scapular winging. Definitive diagnosis was obtained after neurophysiological studies revealed an isolated dorsal scapular nerve injury.

Differential diagnosis

Lateral scapular winging is due to lack of function of the trapezius and/or rhomboid muscles, so these two entities should be considered. Given that in obese patients it is difficult to differentiate medial from lateral scapular winging, medial scapular winging due to lack of function of serratus anterior muscle should also be taken into account.

Laboratory diagnosis

Electrophysiological studies showed an isolated dorsal scapular nerve lesion and proper function of other nerves, such as the long thoracic and spinal nerves.

Imaging diagnosis

Imaging studies (plain X-rays and magnetic resonance imaging) did not reveal any structural injury.

Pathological diagnosis

Not applicable in this case.

Treatment

Scapular stabilization was performed by attaching, to the spine of the scapula, a compound osteomuscular flap obtained from the contralateral trapezius and T11 and T12 hemispinal processes.

Term explanation

Scapular winging: Scapular malposition with its inferior angle prominent, which leads to impairment of shoulder function and pain.

Experiences and lessons

Contralateral trapezius compound osteomuscular flap is a technique that we consider not

especially complex if performed by surgeons with experience in shoulder and periscapular surgery. Given the good result obtained with this dynamic technique for scapular stabilization when the scapular winging was due to dorsal scapular nerve injury, we have abandoned the static techniques that were carried out previously and that yielded worse results with higher associated morbidity.

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Peroneus longus tendon rupture: A case report

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Abstract

BACKGROUND

Peroneal tendinopathies are an under-diagnosed and potentially under-treated pathology. If left untreated it can be a cause of chronic lateral hindfoot pain. Its diagnosis is challenging owing to its low incidence and vague clinical presentation.

CASE SUMMARY

We share a case of a patient who experienced a chronic lateral ankle pain exacerbated after alighting from a bus. This patient came to our attention only after failing conservative management on two separate occasions. Plain radiographs and magnetic resonance imaging revealed rupture of the peroneus longus tendon (PLT). Findings were confirmed intra-operatively and tenodesis of the PLT to the peroneus brevis was performed. Patient was kept non-weight-bear with his foot everted and in plantarflexion before being converted to an off-loading boot at two weeks. Patient was started on a progressive rehabilitation programme at six weeks and was able to return to work shortly after with excellent outcomes.

CONCLUSION

We aim to share our experience in managing this patient and propose some pointers guided by available literature to avoid missing this commonly overlooked pathology.

Key words: Chronic lateral ankle pain; Peroneus longus rupture; Peroneal tendinopathy; Tenodesis; Diagnostic challenge; Case report

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Core tip: Peroneus tendon pathologies are a group of under-appreciated cause of lateral ankle discomfort. Diagnosis is difficult as presenting complains are vague and concomitant ligamentous injury might be present. It is prudent to screen for peroneal

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tendon pathologies in patients complaining of lateral hindfoot tenderness or swelling.

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INTRODUCTION

Ruptures of the peroneus longus tendon (PLT) are rare. The mechanism of injury is due to a combination of mechanical and anatomical factors. The peroneus longus undertakes a tortuous course from its origin at the proximal tibia and fibula to its insertion at the first metatarsal and medial cuneiform. The tendon travels within the posterior fibula groove, passes under the peroneal tubercle of the calcaneus and bends sharply along the cuboid groove before inserting medially at the base of the first metatarsal. These sites are potential sources of attrition^[1] and avascularity^[2-4]. Incompetence of the superior peroneal retinaculum over the fibula groove can predispose PLT subluxation^[1,3]. Varus deformities of the hindfoot or lateral ankle ligamentous laxity can subject the PLT to increased tension and frictional forces at transitional points described above^[3]. In addition, anatomical variability such as the presence of an os peroneum in approximately 20% of the population may predispose to PLT rupture^[5-9].

PLT pathologies can present acutely or insidiously, typically presenting with chronic pain. Patients who are able to relate an initiating injury are defined as acute; whilst patients who were unable to are classified as chronic^[6,10]. Non-surgical management rarely yield acceptable outcomes^[11,12]. Due to the PLT's role in mid and hindfoot stability, surgical correction is often warranted with most authors achieving reproducible results. Timely intervention yields best results^[6,10].

Its incidence remains ill-defined and the condition has only been described in case series or cadaveric studies. Dombek *et al*^[12] documented 5 cases of isolated peroneus longus tears in their series of 40 patients with peroneus tendon pathologies. In contrast, Sobel *et al*^[13] did not report any PLT ruptures in a cadaveric study of 124 ankles. Due its low incidence, peroneus tendon pathologies are often overlooked and often misdiagnosed. The duration between injury and diagnosis of PLT tear ranges from 6 to 48 mo^[6,10,12]. In addition, the ambiguity of its symptoms makes PLT pathologies a diagnostic challenge. In the largest retrospective study available, only 60% of peroneal tendon injuries were accurately diagnosed on initial consult^[12]. Prudent history-taking, adequate examination and appropriate investigations cannot be overemphasized. We aim to share our experience managing a case of PLT rupture and strive to highlight the diagnostic challenges involved in this unique case.

CASE PRESENTATION

Chief complaints

A 51-year-old male flight attendant complained of chronic lateral ankle pain without prior trauma or twisting injury.

History of present illness

Three-month history of left lateral ankle pain. He sought treatment at other Orthopaedic centres but failed conservative management on two separate occasion. He subsequently presented to our care after he exacerbated his lateral ankle pain and heard a "click" whilst alighting from a bus. He denied any traumatic or inversion injury.

History of past illness

Known history of gout and previous left total hip replacement secondary to left hip avascular necrosis.

Personal and family history

None.

Physical examination

On clinical examination, he presented with an antalgic gait, swelling and tenderness over the posterolateral hindfoot. He demonstrated weakness in eversion

Laboratory examination

Not performed.

Imaging examination

Radiograph of the left foot showed a proximally displaced os peroneum compared to contralateral foot (Figures 1 and 2). Peroneal tendon pathology was suspected and magnetic resonance imaging (MRI) of the left foot revealed a full-thickness rupture of the PLT distal to the os peroneum with underlying tenosynovitis (Figure 3).

FINAL DIAGNOSIS

Full-thickness rupture of the PLT with underlying tenosynovitis.

TREATMENT

Surgical repair of the PLT was performed 4 wk after injury. Patient was positioned lateral with a thigh tourniquet applied. Incision was centred over the os peroneum and made from the left fibula tip to the base of the fifth metatarsal (Figure 4). The os peroneum was identified and excised. Unhealthy portions of the PLT and devitalised synovium were debrided. Proximal and distal ends of the PLT was mobilised and the defect was measured (Figure 4F). During surgery, a 1.5 cm longitudinal split of the peroneus brevis was noted and was repaired *via* tubularisation using vicryl sutures (Figure 4G). Side-to-side tenodesis of the PLT to the peroneus brevis tendon was performed using size 2-0 braided polyethylene sutures (ULTRABRAID Suture, Smith and Nephew, York, United Kingdom). The wound was irrigated with normal saline and hemostasis was performed. Layered closure was performed and a backslab was applied over the operated foot, keeping it in plantarflexion and eversion.

OUTCOME AND FOLLOW-UP

Patient was discharged on post-operative day one and was kept non-weight bear on the operated leg. Follow up was done in the outpatient setting at two weeks and six weeks post-operatively. At two weeks, the wound was inspected, skin sutures removed. The backslab was converted to a tall-aircast with heel wedges for the next four weeks and the patient was allowed to perform partial weight-bearing. At six weeks after surgery, patient was allowed full weight-bearing on the aircast and started on a rehabilitation programme aimed at improving range of motion and strength through stretching and isometric exercises.

At three months after surgery, patient demonstrated excellent clinical outcomes (Table 1) with significant improvements in American Orthopedic Foot and Ankle Score, visual analogue pain score as well as the Physical Component Score. He was allowed full weight bearing on normal footwear and returned to ground duties at the airport. At 6 mo, he had returned to sport and was allowed to return to flying duties. This patient continued to demonstrate excellent clinical outcomes at 12 mo post op (Table 1).

DISCUSSION

In a patient with posterolateral hindfoot tenderness and swelling refractory to conservative management, peroneal tendon pathology should be considered. A thorough history detailing the initial injury, exacerbating symptoms, evidence of instability or clicking should be elicited. Associated conditions such as rheumatoid arthritis, neuropathies, gout, local steroid injections and known ankle pathologies (*e.g.*, enlarged peroneal tubercle) can predispose peroneal tendinopathy^[14-18].

The presence of cavovarus or hindfoot varus may increase peroneal tendon tension and increase risk of rupture due to the unopposed action of the posterior tibialis. Physical examination, in particular, palpation should be focused along the course of the peroneal tendons. Common sites of discomfort and swelling include posterolateral hindfoot, cuboid groove and insertion site over at the plantar and medial aspect of the

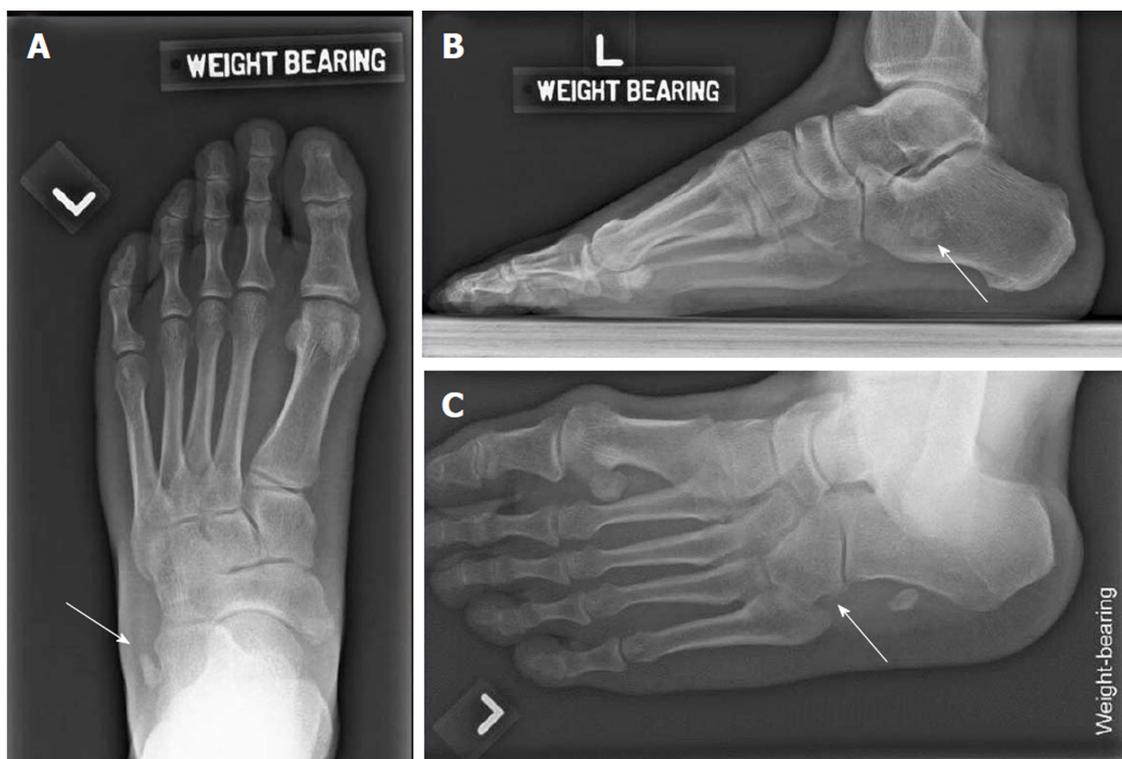


Figure 1 Initial weight bearing radiographs of the injured foot. A: Weight-bearing dorsoplantar view of left foot showing well-defined bony fragment (white arrow) lateral to the anterior left calcaneum; B: Left lateral weight-bearing views shows a bony fragment at the level of the calcaneum; C: Left oblique view. White arrow shows the absence of the os peroneum at its usual anatomical. Instead, it was displaced proximally to the level of the anterior calcaneum process.

foot^[3]. Eversion weakness should increase clinical suspicion of PLT pathology as the peroneal tendons contributes to 63% of eversion strength - with the peroneus longus contributing more than peroneus brevis^[19]. However, it must be noted that the absence of weakness doesn't preclude peroneal tendinopathy. Tenderness on active eversion, passive inversion or during plantarflexion of first ray may also suggest peroneal tendinopathy with the latter specific for the peroneus longus. Lateral ankle ligamentous laxity should also be assessed to rule out concomitant anterior-talofibular ligament (ATFL) incompetence.

Despite having a known history of gout, our patient did not require pharmacological control for his condition and denied any previous local steroid injections. Extra-articular tophaceous gout can predispose to PLT tendinopathy but rarely involves peroneal tendons. To date, there have only been two documented cases of PLT tendinopathy secondary to gout. De Yoe *et al*^[17] described a patient with peroneus brevis tendon rupture and ATFL attenuation secondary to gouty infiltration. Radice *et al*^[6] reported longitudinal tears of both peroneus longus and brevis tendons caused by urate crystal deposition. In our patient, intra-operative findings revealed no gouty infiltration.

Imaging investigations should be guided by clinical findings. Appropriate initial investigations involves weight-bearing anteroposterior, lateral and oblique views of affected foot (Figure 1). Although not routinely performed, the contralateral foot should be imaged in suspected PLT pathologies as it will be helpful in detecting subtle anatomical differences (Figure 2). In our patient, fracture of the os peroneum resulting in proximal migration of the os is pathognomonic of PLT ruptures. This is best seen on oblique and lateral views. There are numerous cases documenting the association of an os peroneum with PLT tears^[5-9]. Some authors however disagree, reporting the lack of association in their series^[3,6]. In addition, the oblique foot view also enables assessment of the peroneal tubercle. Enlargement of the peroneal tubercle alters biomechanics of the peroneus longus and is associated with tears of the PLT^[20,21]. In our patient, the peroneal tubercle was not enlarged.

Ultrasound of the peroneal tendons allow for dynamic assessment, particularly useful in examining peroneal tendon subluxation. It has been shown to provide 90% to 94% diagnostic accuracy of peroneal tendon tears with excellent sensitivity and specificity^[22,23]. MRI is also widely employed, especially since the clinical presentation of PLT can be difficult to diagnose initially^[6]. T2-weighted images allow detection of tendinosis or oedema within the tendons. However, there are a high rate of false

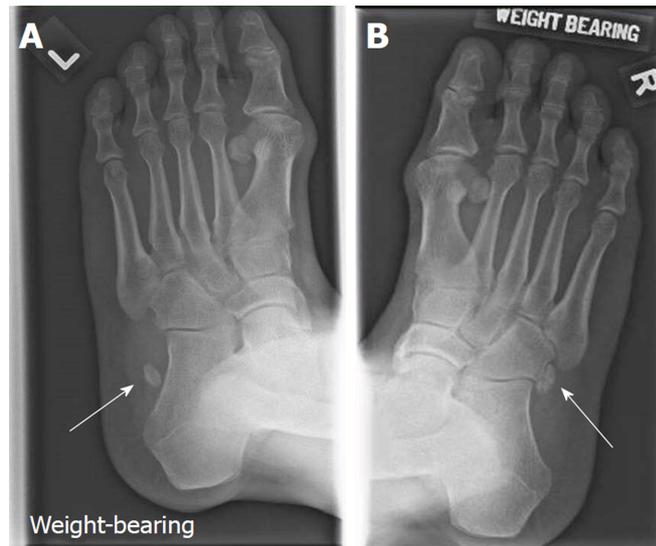


Figure 2 Comparing radiographs of bilateral oblique foot views. A: Weight-bearing oblique view of left foot show a bony fragment lateral to the anterior left calcaneum; B: Shows the position of the undisplaced os peroneum on the right foot. The displaced os peroneum over the left foot is better appreciated when compared against the contralateral foot.

positives and false negatives attributed to signal artefacts^[10,24,25]. O'Neil *et al*^[26] believes that the magic angle effect takes place as the PLT curves postero-inferiorly around the lateral malleolus - mitigatable through plantarflexion of the foot^[26].

In minimally symptomatic patients without ankle instability or loss of function, a trial of conservative management can be offered. Non-surgical treatment protocols are poorly defined but consist of non-steroidal anti-inflammatory medication, rest, orthosis or immobilisation with a short leg cast. Graded rehabilitation programs aiming to increase flexibility and strength are normally started two to four weeks post-immobilisation^[6,19]. Outcomes with non-surgical management remains poor - especially for PLT ruptures^[6,25]. In a series involving 40 patients with peroneal tendon tears, 36 patients reported undergoing unsuccessful non-surgical treatment^[12].

Surgery is the mainstay of treatment for PLT ruptures. Sammarco proposed surgery when a trial of non-operative management has been unsuccessful or when an unstable or varus foot is noted. In the presence of an os peroneum, excision is indicated before repair is attempted. In the absence of an os peroneum, an MRI is indicated to identify cause of rupture or concomitant ligamentous or bony injuries^[6].

Redfern and Myerson's algorithm of treatment considers the condition of the peroneus brevis^[25]. Type I lesions occur when both tendons appear grossly intact; Type II lesions on the other hand occurs when one tendon is torn, whilst the other tendon remains intact and finally Type III lesions occur when both tendons are torn. In the case of a PLT rupture with a usable PBT, this is classified as a Type II lesion in the algorithm and can be managed by tenodesis. Similarly in our patient, the post-debridement gap was too large (four centimetres) (Figure 4F) and did not allow for a direct repair despite maximal mobilization of the proximal and distal ends of the PLT. The use of tendon transfers and autografts have been proposed within literature, but evidence is limited and mostly employed in concomitant PBT ruptures^[27,28]. At present, all studies surrounding the management of peroneal tendon tears or ruptures evolve around Level IV or V evidence and the consensus is that surgical intervention yields the most consistent and reproducible results.

Rehabilitation post-operatively is crucial in achieving positive outcomes. Most authors kept patients non-weight bearing for at least two weeks. During this period, a backslab or hinged boot can be employed to prevent inversion/dorsiflexion which may disrupt the tendon repair. Between two to eight weeks post-surgery, early mobilisation is encouraged through the use of an off-loading walker boot. Redfern and Myerson employs the use of stirrup brace for an additional six weeks and have a low threshold to extend the use of the boot should there be concerns regarding the repair or patient compliance^[25]. It is at this point that most patients are started on a range of motion and strengthening rehabilitation programme^[3,10,12,19,25].

Peroneal tenosynovitis, tendinosis, subluxation or dislocation, stenosing tenosynovitis, os peroneum fracture, acute and chronic tendon tears are part of a spectrum of peroneal tendinopathies. These conditions may co-exist or precipitate one

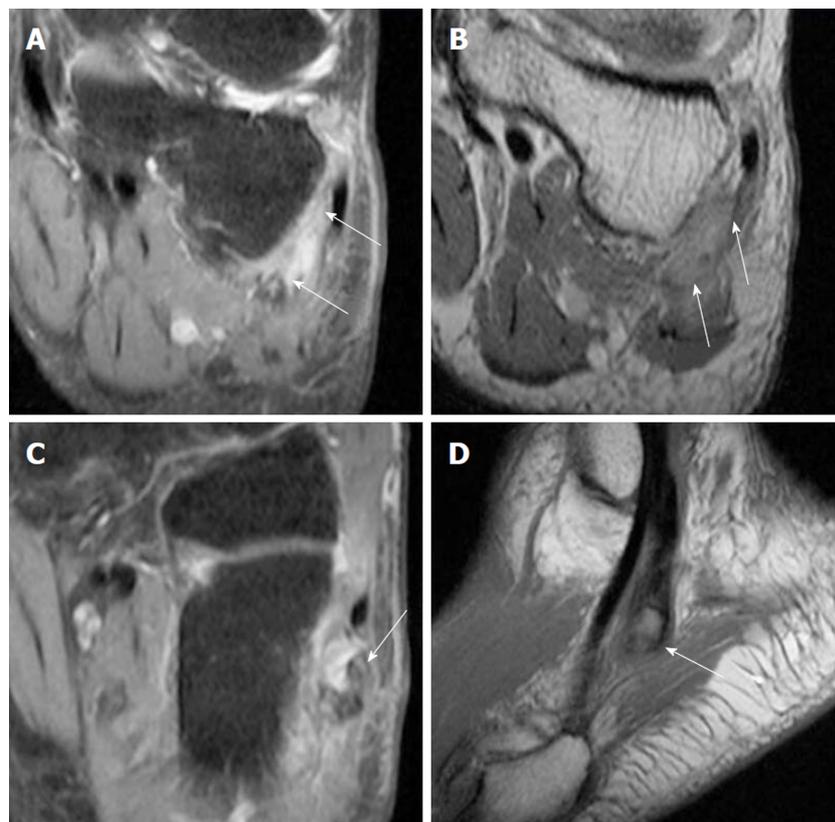


Figure 3 Magnetic resonance imaging of left foot. A: (Coronal) Peroneal tenosynovitis; B: (Coronal) Both peroneal tendons run within a flat peroneal groove with intact superior peroneal retinaculum. No fixed lateral subluxation of the peroneal tendons; C, D: (Sagittal view). Five millimeter os peroneum within the peroneus longus tendon (PLT), with full-thickness rupture of the PLT. The tear measures 2 cm in length, with proximal migration of the tendon at the level of the cuboid bone.

another. In the case discussed, peroneal tendinosis likely precipitated PLT rupture. Peroneal tendinopathy was not considered and therefore, inadequate conservative treatment was prescribed.

We can appreciate the difficulties faced in arriving at this diagnosis. Many other authors have described similar challenges^[6,10,12]. Common differential diagnosis such as lateral ankle ligament injury, base of 5th metatarsal fractures and sinus tarsi syndrome are often considered before investigating for peroneal tendinopathy. In their series of 6 cases, Arbab *et al*^[10] reported requiring an average of 10.8 mo to achieve diagnosis. Sammarco shared similar findings, with a series of 14 cases symptomatic between 7 to 48 mo before definitive diagnosis^[6]. Patients who present acutely and receive timely surgical intervention, achieve the best outcomes. It is therefore essential to diagnose these peroneal tendinopathies early so that appropriate treatment can be commenced promptly. We propose an investigation pathway when reviewing patients with lateral ankle discomfort (Figure 5). Paramount to this is an astute clinical history and examination as well as focused radiographic investigations of the affected ankle and foot. Most differential diagnosis of lateral ankle pain can be ruled out at this juncture. In the event of raised clinical suspicion, radiographic presence or displacement of the os peroneum, persistent symptoms or the absence of a clear diagnosis - early MRI is warranted.

Tenodesis of the proximal stump of the ruptured peroneus longus to the intact peroneus brevis is a useful technique allowing for early return to activity as well as excellent post-operative clinical outcome (Table 1). More studies are required to further evaluate this technique as well as compare clinical outcomes against other techniques available within literature.

CONCLUSION

PLT ruptures are rare and form part of a spectrum of peroneal tendon pathologies that is being increasingly recognized today. This case demonstrates how elusive

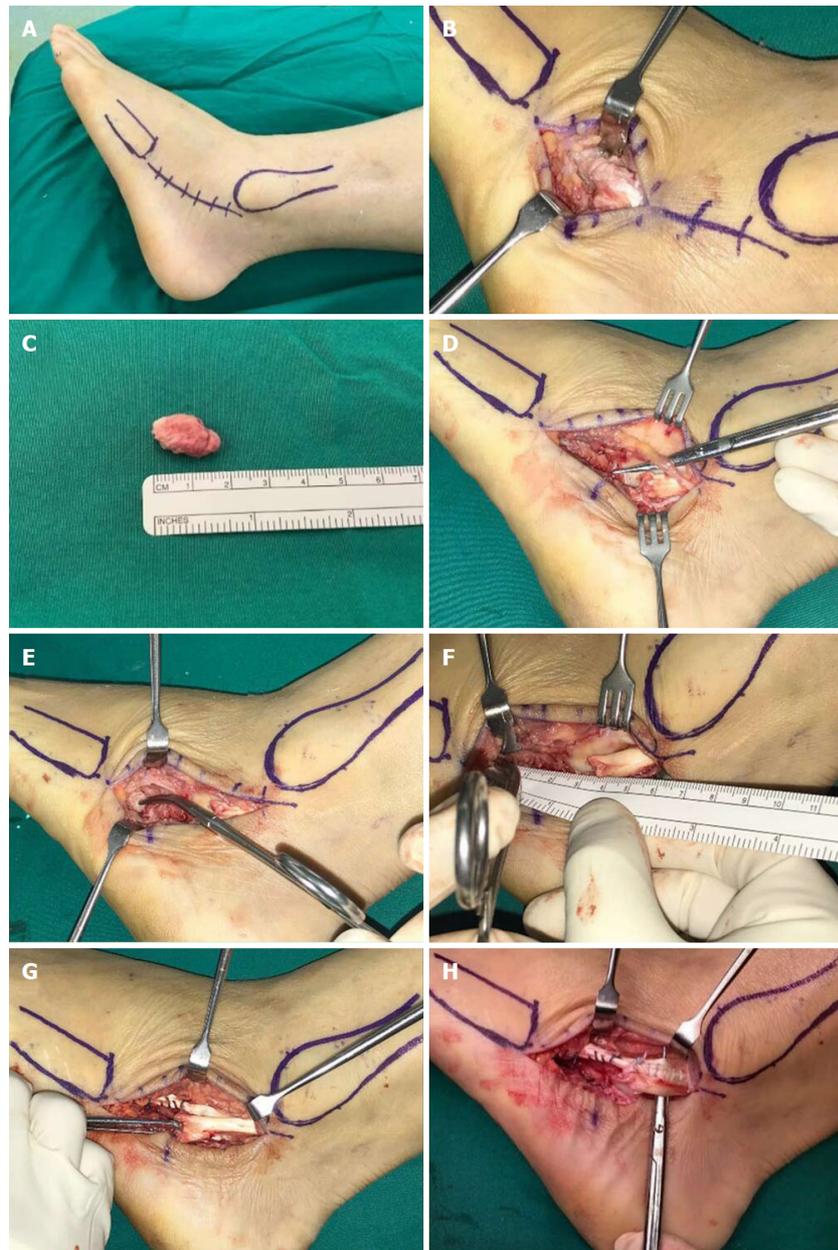


Figure 4 Intra-operative images. A: Lateral position adopted with a thigh tourniquet applied; B: Incision was made from the left fibula tip to the base of the fifth metatarsal - centred around the os peroneum; C: The os peroneum was identified and excised and unhealthy tendon and devitalised synovium were debrided; D: The sural nerve was identified and protected during the surgery; E and F: Proximal and distal ends of the peroneus longus tendon (PLT) was mobilised and defect gap measured; G: The longitudinal split tear in the peroneus brevis was repaired; H: Side-to-side tenodesis of the PLT to the peroneus brevis tendon was performed.

diagnosing peroneal tendon pathology is - especially when it is rarely considered. We shared our experience in promptly diagnosing and surgically treating this patient. Tenodesis of the peroneus longus to the peroneus brevis is a useful technique in the management of peroneus longus ruptures. We achieved excellent early clinical outcome measures that remained excellent 12 mo after surgery.

Table 1 Significant improvement in clinical outcome scores at 6 mo after surgery (excellent results were maintained at 12 mo after surgery)

	Pre-operative	Post-operative	
		6 mo	12 mo
AOFAS Score	60	97	97
Physical Component Score	18.87	63.91	66.78
Visual Analogue Score	4	0	0

AOFAS: American Orthopedic Foot and Ankle Score.

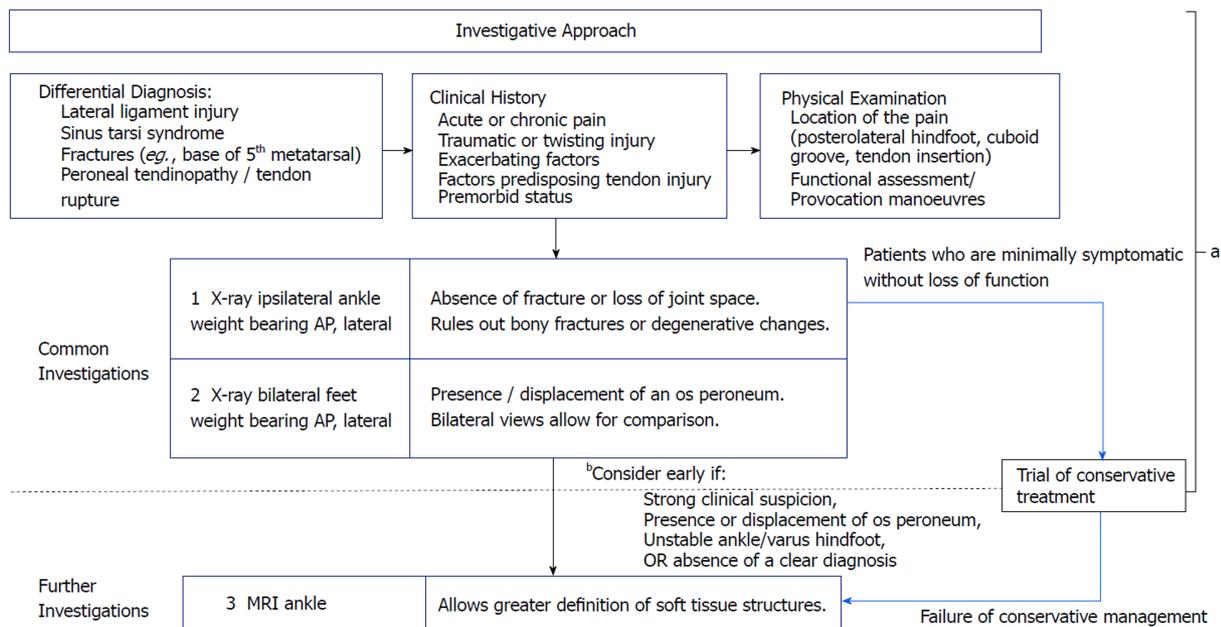


Figure 5 Investigative pathway when approaching patients with lateral ankle discomfort.^aAt this juncture, most differential diagnosis can be ruled out with astute clinical and radiological findings; ^bEarly magnetic resonance imaging ankle is warranted should there remain a strong clinical suspicion, persistently symptomatic patient and even the absence of a definitive diagnosis.

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