The outcome of scapulothoracic fusion for painful winging of the scapula in dystrophic and non-dystrophic conditions

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Scapulothoracic fusion (STF) for painful winging of the scapula in neuromuscular disorders can provide effective pain relief and functional improvement, but there is little information comparing outcomes between patients with dystrophic and non-dystrophic conditions. We performed a retrospective review of 42 STFs in 34 patients with dystrophic and non-dystrophic conditions using a multifilament trans-scapular, subcostal cable technique supported by a dorsal one-third semi-tubular plate. There were 16 males and 18 females with a mean age of 30 years (15 to 75) and a mean follow-up of 5.0 years (2.0 to 10.6). The mean Oxford shoulder score improved from 20 (4 to 39) to 31 (4 to 48). Patients with non-dystrophic conditions had lower overall functional scores but achieved greater improvements following STF. The mean active forward elevation increased from 59° (20° to 90°) to 97° (30° to 150°), and abduction from 51° (10° to 90°) to 83° (30° to 130°) with a greater range of movement achieved in the dystrophic group. Revision fusion for nonunion was undertaken in five patients at a mean time of 17 months (7 to 31) and two required revision for fracture. There were three pneumothoraces, two rib fractures, three pleural effusions and six nonunions. The main risk factors for nonunion were smoking, age and previous shoulder girdle surgery.

STF is a salvage procedure that can provide good patient satisfaction in 82% of patients with both dystrophic and non-dystrophic pathologies, but there was a relatively high failure rate (26%) when poor outcomes were analysed. Overall function was better in patients with dystrophic conditions which correlated with better range of movement; however, patients with non-dystrophic conditions achieved greater functional improvement.

In patients with neuromuscular disorders affecting the shoulder girdle, selective weakness of the muscles that stabilise the scapula to the chest wall, particularly serratus anterior, trapezius and the rhomboids, may result in scapulothoracic dyskinesia with pain in the shoulder, reduced strength and limitation of movement.1,2 When the scapula loses stability, deltoid works at a mechanical disadvantage and painful winging of the scapula may result from altered kinetics or muscle fatigue.3

Most patients with symptomatic winging of the scapula, scapulothoracic pain or crepitus respond to non-operative measures that involve analgesia, local cortisone injections and physiotherapy to strengthen the scapular stabilisers and thoracohumeral muscles. When these measures fail, Copeland and Howard1 were the first to report good results from scapulothoracic fusion (STF) that produces a stable fulcrum for glenohumeral movement and improves the efficiency of muscle contraction.1,2,4-10 Most papers on STF have analysed the outcome in patients with facioscapulohumeral muscular dystrophy (FSHD),7-11 The outcome of STF in non-dystrophic conditions such as isolated serratus or trapezius weakness, polio, neuralgic amyotrophy, scapular malunion and osteochondroma is less well reported.4-6

Various techniques for fusion have been described using cerclage wires, rush pins, plate fixation, allograft and autografts.1,2,4,5,7,12-14 Complications include pulmonary and neurovascular injury as well as failure with painful nonunion.6,15,16 There is little information on the risk factors for nonunion after STF and the need for revision surgery and there have been no comparative studies between patients with dystrophic conditions that are progressive and non-dystrophic conditions that are not. Therefore the aim of this study was to report the differences in clinical and functional outcome of STF for dystrophic and non-dystrophic conditions and analyse the risk factors for nonunion and failure.

Patients and Methods

In our tertiary referral shoulder unit between 2001 and 2010, 42 consecutive STFs were
performed in 34 patients using a modified multifilament transscapular, subcostal cable technique supported by a dorsal one-third semi-tubular plate and autologous bone graft. There were 18 females and 16 males with a mean age of 30 years (15 to 75) and a mean follow-up of 5.0 years (2 to 10.6). All patients were followed to failure or pain-free fusion. A total of eight patients had staged bilateral procedures. Surgery was performed on the right or pain-free fusion. A total of eight patients had staged bilateral procedures. Surgery was performed on the right or pain-free fusion. A total of eight patients had staged bilateral procedures.

Table I. Patient diagnoses

<table>
<thead>
<tr>
<th>Diagnosis*</th>
<th>Number of patients (n = 34)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Muscular dystrophies (n = 15)</td>
<td></td>
</tr>
<tr>
<td>FHSD</td>
<td>13</td>
</tr>
<tr>
<td>Myotonic dystrophy</td>
<td>1</td>
</tr>
<tr>
<td>Limb girdle muscular atrophy</td>
<td>1</td>
</tr>
<tr>
<td>Non-dystrophic pathology (n = 19)</td>
<td></td>
</tr>
<tr>
<td>Isolated serratus anterior palsy</td>
<td>6</td>
</tr>
<tr>
<td>Isolated spinal accessory nerve and trapezius palsy</td>
<td>2</td>
</tr>
<tr>
<td>Brachial plexus injury</td>
<td>3</td>
</tr>
<tr>
<td>Poland syndrome with scapula dyskinesia</td>
<td>1</td>
</tr>
<tr>
<td>Neuromuscular scoliosis and spinal dysraphism</td>
<td>2</td>
</tr>
<tr>
<td>Hereditary multiple exostosis with previous excisions and scapula dyskinesia</td>
<td>2</td>
</tr>
<tr>
<td>Loss of scapula stabilisers secondary to central nervous system pyramidal tract injury (CVA, head injury, bulbar palsy)</td>
<td>3</td>
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* FHSD, facioscapulohumeral dystrophy; CVA, cerebrovascular accident

Radiological union was defined as the presence of bridging trabeculae between the scapula and ≥ two ribs on at least three cortices visible on two orthogonal radiographs (anteroposterior and lateral scapular-Y view). Clinical union was defined as no tenderness or movement at the fusion site with no pain on functional loading. A failure analysis was performed. Failure of the procedure was defined as: 1) the need for revision or exploration of the fusion whatever the cause (nonunion, persistent pain, fracture of fusion mass etc); 2) failure to achieve improvement in pain and/or range of movement more than two years post-operatively; or 3) subjective dissatisfaction (very satisfied, satisfied, or not sure).

All procedures were performed by the senior authors (DSH, MF, SML, IB). The patient is positioned prone on a well-padded mattress with the head supported in the neutral position and both hands in symmetrical external rotation next to the head. The skin is draped from C6 to the sacrum, permitting access to the ipsilateral posterior iliac crest. The approach is through an incision placed medial to the medial border of the scapula at 20° to the midline in line with the posterior angle of the third to seventh ribs (Fig. 1a). The trapezius muscle, rhomboid major and minor and levator scapulae are partially detached medially and retracted to expose the medial border of the scapula. A medial band of supraspinatus and infraspinatus 2 cm to 3 cm wide is elevated from the dorsal aspect of the scapula to expose the dorso-medial border. The subscapularis and serratus anterior are elevated from the deep surface of the scapula antero-medially and partially excised. Care is taken not to elevate the subscapularis beyond the midline in order to maintain the inferior and lateral attachments and prevent denervation. The cortical surface of the anteromedial scapula is roughened with a curette. The dorsomedial end of the spine of the scapula is contoured with a burr to permit seating of a supporting one-third semi-tubular plate. The posterior angles of the second/third to sixth/seventh ribs are exposed sub-periosteally and partially decorticated to punctate bleeding with a burr.
All redundant soft tissue between the scapula and the ribcage is removed to maximise bony contact. The pleura is carefully protected with blunt retractors and four or five 12G stainless steel multifilament cables (Atlas, Medtronic, Minneapolis) are passed subcostally around the third to sixth/seventh ribs and through the scapula supported on its posterior surface by a nine- or ten-hole contoured stainless steel one-third semi-tubular plate (Synthes, Waldenberg, Switzerland) (Fig. 1b). The scapula is brought to the desired position and the cables provisionally tensioned. The scapula should be placed with the caudal tip over the sixth rib with the medial border making a 15° to 20° angle with the spinous processes of the thoracic spine distally. This makes the glenoid face superiorly, which is the position obtained when the arm is at the side at rest.

Stabilisation of the scapula in excessive adduction limits function of the glenohumeral joint. External rotation of the scapula improves the mechanical advantage of the supraspinatus and deltoid; however, excessive rotation will cause the shoulder to appear abnormally high. In patients with progressive neuromuscular disorders we aimed to place the scapula subtending the angle of 10° to 15° with respect to the thoracic spine distally to enable pain-free adduction of the arm at rest without impingement, in order to prevent progressive weakness limiting future movement. There is a tendency for the scapula to displace laterally and care should be taken to place it on the curvature of the posterior ribs, as when it is placed too far laterally the shoulder protrudes. Autogenous cancellous bone graft is harvested from the posterior iliac crest and packed under the medial border of the scapula and the cables are definitively tensioned to achieve stability (Fig. 1c). The operative site is filled with crystalloid solution and the anaesthetist generates a Valsalva manoeuvre.
in the patient to detect any tears of the pleura. The rotator cuff muscles and rhomboids are reattached and the wound closed in layers with a suction drain. In cases performed after 2007 silicated calcium phosphate putty (Actifuse ABX, Apatch, Elstree) was used to augment the fusion.

A thoracobrachial orthosis is used for three months with the shoulder in neutral rotation, 15° flexion, and 30° abduction with the elbow in neutral rotation and 70° flexion. A post-operative chest radiograph is taken to assess scapular and implant position (Fig. 2), and to detect pulmonary complications. Patients with muscular dystrophy are given salbutamol to maintain muscle tonicity while the arm is immobilised. Physical therapy begins at three months with active movements and muscle strengthening exercises.

Radiological review was undertaken at three, six, and 12 months until union and annually thereafter.

**Statistical analysis.** This was performed using Stata/IC version 11.2, (StataCorp, College Station, Texas). Odds ratios (OR) with 95% confidence intervals (CI) using Fisher’s exact test were calculated to analyse risk factors for nonunion. The unpaired Student’s t-test was used to compare quantitative measures. A p-value of < 0.05 was considered significant.

**Results**

Pre-operatively all 34 patients (42 scapulae) had painful winging and significant functional difficulty using the arm. Primary clinical and radiological union was achieved in 36 scapulae (86%; 28 patients) at a mean of seven months (4 to 15). Failure of the procedure occurred in 11 STFs (26%; 11 patients).

Subjectively 23 patients (67%) were very satisfied with their result, five (15%) were satisfied, three (9%) were unsure and three (9%) were dissatisfied. Improvement in pain was reported by 29 patients (85%). Failure of the procedure occurred in 11 STFs (26%; 11 patients).

One patient with FSHD gradually lost all the gains achieved after STF and was left with worse movement than before the operation; and one patient developed a painful nonunion that required revision. This latter patient had originally undergone STF for painful winging secondary to a stroke and had ipsilateral upper motor neuron weakness in the arm.
The mean OSS improved from 20 (4 to 39) pre-operatively to 31 (4 to 48) at final follow-up. And the mean range of active forward elevation and abduction improved from $59^\circ$ (20° to 90°) and $51^\circ$ (10° to 90°), respectively, to $97^\circ$ (30° to 150°) and $83^\circ$ (30° to 130°), respectively. Patients with non-dystrophic conditions, despite having lower pre- and post-operative OSS, achieved greater functional improvements following STF at final follow-up than the dystrophic patients, although this was not significant (mean difference in pre- and post-operative OSS between groups = 4 (95% confidence interval (CI) -2.2 to 11.7) (p = 0.173, unpaired t-test). Subgroup analyses are shown in Table III. In the dystrophic group three patients gradually lost all their initial improvement because their disease progressed and at last follow-up they had less movement than before the operation. One non-dystrophic patient had less movement due to persistent pain and scapulothoracic dysfunction.

A total of 24 patients (70%) reported an improvement in cosmetic appearance. This was primarily due to an improvement in shoulder symmetry. Six (18%) reported no difference and four (12%) were dissatisfied with their cosmetic result. In one this was due to a prominent scar, and in the other three it was because of asymmetrical prominence of the shoulder that was made more noticeable by the muscular atrophy of the shoulder girdle. In the 15 patients who underwent fusion for muscular dystrophy, seven reported an improvement in the posture of their lumbar spine and in their gait. This was not reported by any patient in the non-dystrophic group.

Revision was required in five shoulders (12%, five patients) for nonunion at a mean of 17 months (7 to 31) and in two (5%) for fracture of the fusion mass, which may have been due to previous falls, at a mean of 72 months (34 to 110) (Fig. 3). In one patient (patient 3) a second revision was required at 32 months for persistent nonunion; union was finally achieved at 38 months. Patients who underwent revision STF achieved clinical and radiological union at a mean time of 11 months (6 to 38). In one patient with FSHD (patient 8) a nonunion developed but revision was not undertaken as he reported an improvement in pain and was able to return to his job as a manual labourer; however, there was loss of mobility as the pre-operative forward elevation was 80°, which reduced to 45° at seven years. Removal of metalwork was performed in 13 shoulders (31%, 13 patients) and one patient required excision of a prominent supero-medial pole of the scapula for persistent pain. A

<table>
<thead>
<tr>
<th>Variable</th>
<th>Pre-operative</th>
<th>Post-operative</th>
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<tbody>
<tr>
<td>All fusions (n = 42)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Forward flexion (°)</td>
<td>59 (20 to 90)</td>
<td>97 (30 to 150)</td>
</tr>
<tr>
<td>Abduction (°)</td>
<td>51 (10 to 90)</td>
<td>83 (30 to 130)</td>
</tr>
<tr>
<td>Oxford shoulder score</td>
<td>20 (4 to 39)</td>
<td>31 (4 to 48)</td>
</tr>
<tr>
<td>Muscular dystrophy fusions (n = 22)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Forward flexion (°)</td>
<td>62 (30 to 90)</td>
<td>103 (70 to 130)</td>
</tr>
<tr>
<td>Abduction (°)</td>
<td>51 (30 to 90)</td>
<td>88 (45 to 120)</td>
</tr>
<tr>
<td>Oxford shoulder score</td>
<td>25 (4 to 34)</td>
<td>34 (4 to 48)</td>
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<td>Non-dystrophic fusions (n = 20)</td>
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<tr>
<td>Forward flexion (°)</td>
<td>53 (20 to 90)</td>
<td>88.5 (30 to 150)</td>
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<tr>
<td>Abduction (°)</td>
<td>49 (10 to 90)</td>
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<tr>
<td>Oxford shoulder score</td>
<td>15 (4 to 39)</td>
<td>28 (7 to 47)</td>
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</table>

Fig. 3a: Post-operative a) chest and b) right lateral scapular radiographs at eight years showing broken wires and fragmentation of the fusion. The scapula has lifted off the thoracic cage and migrated superiorly. The patient underwent successful revision (c) using a compression plate with screw fixation into the solid intercostal fusion that was present.

Fig. 3b

Fig. 3c
pneumothorax developed in three patients that resolved without intervention in a week. Rib fractures developed at the site of the wires in two patients, one of whom was dissatisfied and required a T3-T7 intercostal nerve block for allodynia and costal pain. An ipsilateral reactive pulmonary effusion arose in three patients but resolved without intervention.

Patients who smoked were at a higher risk of nonunion than non-smokers (OR 12.4 (95% CI 1.3 to 156.6); p = 0.0135). Patients ≥ 35 years of age at the time of surgery were at a higher risk of nonunion (OR 10 (95% CI 0.9 to 492); p = 0.032), as were patients who had undergone previous shoulder girdle surgery (OR 11 (95% CI 0.9 to 118); p = 0.029). We did not find a statistically increased risk of nonunion when independently analysing the number of ribs involved in the STF, the side, the comorbidities or the primary diagnosis (all p > 0.05).

Discussion
Winging of the scapula, which is the most common cause of symptomatic scapulohumoral dysfunction, may be caused by a number of traumatic or atraumatic conditions that result in loss of the scapular suspensory mechanism, weakness of the scapular stabilising muscles, serratus anterior or trapezius weakness, scapulothoracic pain or glenohumeral instability. Initial treatment should involve physiotherapy to strengthen the scapular stabilisers, which are the thora-cohumeral and scapulohumeral muscles, alongside selective use of cortisone injections for pain relief. Many patients respond to these non-operative measures, but there are those who have complex instability and require surgical stabilisation. Pectoralis major muscle transfer was first described in 1904 and remains the most common and successful surgical treatment for winging of the scapula, especially for winging caused by isolated serratus anterior palsy. This success has enabled the evolution of other muscle transfers such as the levator scapula and rhomboids in patients with trapezius palsy. However, these muscles may not be available in patients who have traumatic damage, and transfers are unsuitable for patients with progressive muscular dystrophy. Scapulopexy using strips of fascia lata or synthetic graft to attach the scapula to the ribs does not produce rigid fixation and is limited by subsequent stretching of the fascial slings and functional deterioration. STF is a salvage procedure that has been reported to optimise movement and alleviate pain for patients with painful winging of the scapula; however, there is little information on the causes of failure, nonunion and the requirement for revision surgery.

We performed a retrospective review of 42 STFs with a mean follow-up time of five years and found high patient satisfaction levels in 82% of patients, but there was a 26% failure rate of the procedure. There were six nonunions (14%) in six patients, five of whom underwent revision, and two patients were revised for fracture of the fusion (5%). We found smoking, age and previous surgery of the shoulder girdle were the main risk factors for nonunion.

Rates of nonunion between 0% and 30% have been reported following STF with rates of union being higher in muscular dystrophy than in non-dystrophic conditions, but no directly comparative studies have been undertaken. Diab, Darras and Shapiro reported rates of union of 100% in eight patients with FSHD and Letournel et al reported only one asymptomatic pseudarthrosis in nine patients with FSHD. In contrast Bizot et al reported a 30% rate of nonunion in ten patients with serratus anterior palsy, two of whom underwent revision. In the current study nonunion occurred in two patients with dystrophic and four patients with non-dystrophic conditions. Variability in rates of union reported in previous studies may reflect confounding from factors such as age, as patients with FSHD tend to be a younger cohort.

In the current study, patients who underwent revision STF achieved clinical and radiological union at a mean time of 11 months (6 to 38); however, one patient required a further revision for persistent nonunion. In our experience, an intercostal fusion between the ribs is a consistent finding during revision surgery for failed STF. The periostium of the ribs is a rich source of stem cells and the intercostal space is a stable mechanical environment that probably explains why the ribs fuse together. High shearing and torsional forces on the scapula with narrow fusion surfaces and consequent lack of stability, coupled with failure of biology to achieve fusion in an adequate time, are the probable primary causes for nonunion following STF. This was our rationale for immobilisation in a thoracobrachial orthosis for 12 weeks post-operatively. This is a long time for patients to tolerate wearing a bulky orthosis, although we did not identify a problem with compliance during this study. Pre-operative counselling is essential to ensure patients understand the rehabilitation programme. During revision STF, the intercostal fusion is a stable bony platform upon which to obtain either screw or wire fixation. We aimed to achieve further fixation with wires unless we considered the risk of iatrogenic pleural injury to be too high because of difficulty obtaining a circumferential passage of wires around the area of fusion. We also routinely used autologous cancellous bone graft and BMP-7.

Patient satisfaction was high in both the dystrophic and non-dystrophic groups. The primary reason for this was improvement in pain and ability to perform activities of daily living. The mean pre- and post-operative functional scores were higher in the dystrophic group, which correlated with a greater range of movement of the shoulder. However, the mean gain in function was smaller in the muscular dystrophy group which may reflect progression of their disease. FSHD is an autosomal dominant muscular dystrophy with variable expressivity that is characterised by progressive weakness of the facial and shoulder girdle muscles with relative sparing of the deltoid and rotator cuff. The natural history is slowly progressive and may involve the lower limbs with eventual wheelchair dependence in 19% of patients. It has been hypothesised that the pathology is related to calcium regulation in muscle cells and that calcium antagonists such as
diltiazem may be effective in delaying progression. Early studies analysing the effect of prednisolone on halting or retarding muscle weakness were unsuccessful. Modest benefit was seen with the administration of salbutamol, a beta-2-adrenergic agonist with a myotrophic action. The use of beta-2-adrenergic agonists has been advocated for patients with muscular disease who undergo prolonged inactivity, in order to preserve the competence of unaffected muscle groups. We have started to use salbutamol routinely post-operatively in our dystrophic group since 2008 but have not noticed significant differences in functional outcome.

STF has been associated with complications including haemorhax, pneumothorax, pleural effusion, brachial plexopathy and vascular injury. Krishnan et al reported a pulmonary complication rate of 46% using a wire technique. They advocated deflation of the ipsilateral lung during passage of cerclage wires around the ribs and prophylactic insertion of a thoracic drain retained for one to two days post-operatively. Pulmonary complications occurred in 14% of fusions in the current study. We did not deflate the lung prior to the passage of cerclage wires, which may explain the smaller number of reactive pleural effusions experienced, without a concomitant increase in the incidence of pneumothorax. There is a theoretical risk of reduced respiratory function in patients who undergo bilateral STF due to reduced chest expansion. One study demonstrated a reduction of forced vital capacity by 21%, however, other studies have shown little or no change in respiratory function. We did not formally assess respiratory function in this study but none of the patients with bilateral STF reported limitation of respiratory function at final view. We experienced two cases of fracture of the fusion mass. Both these patients had relatively lateralised and posteriorly prominent scapulae that may have predisposed to this complication. Hardware removal was the most common cause of re-operation.

This study has several limitations. It is a retrospective review with no true control group and no randomisation, making it prone to confounding and measurement bias. Selection bias was a problem and was the cause of differences in baseline functional scores. Relatively small patient numbers and inadequate power risk type 2 error and were the cause of large confidence intervals. STF is a rare procedure and there have been no reports comparing the outcome of STF for dystrophic and non-dystrophic pathologies or analysing risk factors for nonunion and failure.

Supplementary material
A table detailing the pre-operative demographics and risk factors for patients who developed nonunion and those who underwent revision is available with the electronic version of this article on our website www.bjj.boneandjoint.org.uk

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References