INTRODUCTION

Due to advances in chemotherapy, medical imaging and surgical techniques, Limb salvage surgery has become the preferred procedure for Musculoskeletal tumours, if surgical margins are not compromised. However, in certain instances including involvement of neurovascular bundle, active infection, involvement of the joint and patient preference, ablative surgery is performed. Tumours involving the shoulder girdle are not uncommon (Figure-1). Tumours largely restrict themselves to same bone, but on occasion can involve the joint. In cases where they involve the shoulder joint, neurovascular bundle or majority of shoulder girdle muscles and in cases where tumours are non-responsive to chemo and or radiotherapy or in case of fungating tumours, Forequarter amputation (FQA) is a viable surgical treatment. Forequarter amputation involves removal of entire upper limb and shoulder girdle, scapula and a portion of clavicle. However, FQA may not always be required and limb salvage may still be possible, by performing the extra articular resections and reconstruction, if only the joint is involved and neurovascular bundle is spared.

Forequarter amputation can be performed both in a curative and palliative setting and can be a disfiguring procedure imparting physical and psychological trauma to the patient, hence necessitating thorough pre-operative counselling. Ralph Cumming was the first to describe the forequarter amputation in 1808, but at the time that was reported for trauma. However, it was Dixie Crosby in 1836 who reported and described its use in oncological cases. The procedure can be performed by using either the anterior approach of Berger or the posterior approach of Littlewood. Ferrario et al described a more convenient combined anterior and posterior approach. This approach may be preferred because of the excellent surgical exposure and early ligation of subclavian vessels at the thoracic inlet. The purpose of our study was to evaluate the outcomes of combined anteroposterior approach for forequarter amputation for tumours involving the shoulder girdle.

MATERIAL AND METHODS

Data for this single surgeon series was collected retrospectively from a prospectively held database at the authors primary institution. All the patients who underwent FQA in either a curative or palliative setting...
from January 2016 to August 2019 were included in this study. All the patients underwent staging studies and a biopsy for histological confirmation of diagnosis followed by discussion in a tumour board meeting (Figure-2). Chemo sensitive tumours, underwent neoadjuvant treatment where appropriate, followed by surgery. All surgeries were performed under general anaesthesia by a single surgeon, in a standard fashion, with standard antibiotic cover. Patients were placed in a floppy lateral position, stabilised with side supports and tapes with the affected upper extremity on the top (Figure-3). A combined anteroposterior approach was used in all the patients with the apex of incision at the junction of medial third of clavicle with middle third. Osteotomy of the clavicle was performed in all cases at the skin incision level with a gigli saw and care was taken not to disturb the attachment of sternocleidomastoid muscle. Careful soft tissue dissection was followed by identification of subclavian vessels which were clamped, transected and ligated twice at the root of the stump. Cords of the brachial plexus were then identified and transected with a sharp blade and allowed to retract medially to avoid neuromas (Fig.4). The position of posterior limb of incision was variable and depended on extent of involvement of the scapula or periscapular muscles and was also adapted to size of posterior flap required to close the wound. This however, was usually lateral to the medial border of scapula. Once all the muscles were lifted from the scapula, careful assessment was made of any involvement of chest wall by the tumour, by inserting the hand examining the axillary space. If chest wall is involved, this needs to be removed with the tumour (not required in any of our cases). Once the scapula was freed, the whole shoulder girdle fell free and was wrapped and sent for histological analysis. After careful haemostasis, the wound was closed in layers over a suction drain.

RESULTS
A total of 13 patients were identified and included in the study including 8 male patients. Six patients underwent surgery with curative intent. Mean age of patients at surgery was 20 years (Range 10–53 years). Six patients had primary osteosarcoma and 4 had Ewing’s sarcoma (Table-1). Two patients presented with fungating masses while the rest presented with large painful lumps. All patients except 2 received neoadjuvant chemotherapy. Two patients had undergone previous surgeries, with one having debulking surgery for osteosarcoma and one had 3 prior surgeries for spindle cell sarcoma. Seven patients had Enneking stage 3 disease and had surgery performed for intractable pain (palliative procedure).

No major per operatives complication were encountered in any of the cases with a mean blood loss of 350 ml and mean duration of surgery being 75 minutes. One patient had post-operative wound problems which required debridement under local anaesthesia with no further complications. No immediate (within 6 weeks) mortality was reported post operatively. All patients had clear margins, except 1 patient (planned positive margins resection) who had palliative amputation and had 3 prior surgeries before presentation (patient 4 in table-1).

At last, follow up only 6 patients were alive, with 2 patients alive with disease (Metastasis) and undergoing palliative treatment. None of our patients had local recurrence. Mean survivorship for the whole cohort was 9.2 months (Range 3–18 months) with a mean survivorship for the deceased group was 7.1 months (Range 3–16 months) and mean survivorship for alive patients was 11.6 months (range 9–18 months). All the alive patients had phantom limb sensations and only one had a prosthesis in place only for cosmetic reasons, at last follow up. None of the patients reported any significant problems with neck movement or balancing issues (Figure-5).
Figure 3: Floppy lateral position for Forequarter amputation in a near fungating lesion (a). Skin marking for incision for combined anteroposterior approach (b). Tension free primary wound closure over a suction drain in line of incision (c).

Figure 4: Osteotomy of the clavicle using giggly saw, identification, transection and ligation of subclavian vessels (b). Identification of cords of brachial plexus before resection.

Figure 5: Post op clinical photographs from front (a), back (b) and side (c) of a patient at 2 months following Forequarter amputation.
DISCUSSION

Forequarter amputation is an uncommon operation but a viable treatment option in selected cases of bone and soft tissue sarcomas, when limb salvage procedures are not possible.\(^9,14\) Malignant tumors of the upper extremity can remain a challenging problem for the patient and surgeon, however, FQA is an option for patients who have failed to respond to other treatments and is a salvage procedure performed only for narrow indications. Most of the patients in our study had received chemo and/or radiotherapy and few already had undergone some form of surgery from non-sarcoma surgeons (mostly debulking). Forequarter amputation is also an extensive and disfiguring procedure and can create initial refusal on the part of the patient, and surgeons too can feel hesitant in performing the surgery. This results in few such procedures being performed.\(^15\)

There are however, clear indications for this procedure and it gives the opportunity of achieving clear margins in high grade and difficult tumour presentations.\(^9,16\) In our study the most common malignancy was locally advanced bony sarcomas (Osteosarcoma followed by Ewing’s sarcoma). Only two patients had soft tissue sarcoma with fungating masses (Table-1). In almost all our cases there was neurovascular involvement and involvement of the joint.

Besides the poor cosmesis, the common reported complications associated with FQA are failure to attain clear margins, blood loss, wound problems and phantom limb pain.\(^2,17,18\) In a study by Kumar et al using anterior approach, the average time of surgery was 62 minutes, blood loss ranged from 400 to 750 ml and 1 patient out of 11 had superficial wound infection, which is comparable to our results.\(^19\) Elsner et al, reported mean operation time of 119 min, 1 major and five minor complications and local recurrence in 4 out of 30 patients treated with forequarter amputation.\(^20\) In contrast, none of our patients had local recurrence till last follow up. The prognosis of the patients undergoing FQA depends on the underlying disease and stage. The mean duration of survival was 5.8 months in a study conducted by Faisham et al, using posterior approach, while in our study the mean duration of survival was 9.2 months.\(^21\) The mean estimated blood loss reported in this study was 900 ml and the mean 8 duration of surgery was 3.8 hours. In comparison our average blood loss and mean operating time was far less than reported in this study. Patients having high grade lesions of the shoulder girdle in whom FQA is performed have grim outcomes because of size of the tumour, stage of disease and grade of the tumour. Another study reported better outcomes when amputation was done primarily in comparison to those patients who had amputation for local recurrence after previous excision.\(^22\) Those requiring amputation of a limb with systemic disease at presentation have a very poor prognosis, as is also seen in our series.\(^23\) In a study performed by Bhagia et al, the 5-year survival rate was 21 percent and 30 percent for palliative and curative cases respectively.\(^2\) They also reported that none of their patients used artificial prosthesis. In our study, out of 6 patients with stage 2 disease who underwent FQA, 5 were alive, while in 7 patients with stage 3 disease only 1 was alive (Table-1). This shows good prognosis of the procedure when performed earlier in the course of the disease with curative intent, however our follow up was short.

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As FQA is a mutilating procedure, there is considerable psychological stress associated with the amputation (Figure-5). Phantom limb is another commonly reported psychological factor experienced by patients after amputation with numerous remedies mentioned in literature with mixed results.\(^24\) In our study almost, all patients had phantom limb sensation. We used Gabapentin for all our patients, although with no significant help and cognitive behavioural therapy where available. Pre-operative counselling is necessary to avoid such events and should include meeting with other patients who have had a similar procedure. Majority of the patients reported reluctance to the use of artificial limb, possibly because of absence of proprioception and lack of fine movements. In our study only one patient had prosthesis in placed for cosmetic reasons. Currently the options of a cosmetically and functionally accepted prosthesis are very limited and also because of the difficulty in maintaining a prosthesis.

Table-1: Status of patients on last follow up

<table>
<thead>
<tr>
<th>Age</th>
<th>Gender</th>
<th>Diagnosis</th>
<th>Stage</th>
<th>Status</th>
<th>Survivorship (Months)</th>
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<tbody>
<tr>
<td>10</td>
<td>M</td>
<td>Ewing’s sarcoma</td>
<td>2a</td>
<td>Deceased</td>
<td>8</td>
</tr>
<tr>
<td>16</td>
<td>M</td>
<td>Osteosarcoma</td>
<td>3</td>
<td>Deceased</td>
<td>4</td>
</tr>
<tr>
<td>26</td>
<td>F</td>
<td>Ewing’s sarcoma</td>
<td>2b</td>
<td>Deceased</td>
<td>16</td>
</tr>
<tr>
<td>53</td>
<td>M</td>
<td>Spindle cell sarcoma</td>
<td>2</td>
<td>Deceased</td>
<td>7</td>
</tr>
<tr>
<td>14</td>
<td>M</td>
<td>Osteosarcoma</td>
<td>2b</td>
<td>NED</td>
<td>11</td>
</tr>
<tr>
<td>12</td>
<td>M</td>
<td>Osteosarcoma</td>
<td>3</td>
<td>AWD</td>
<td>10</td>
</tr>
<tr>
<td>22</td>
<td>F</td>
<td>GCT</td>
<td>2b</td>
<td>NED</td>
<td>18</td>
</tr>
<tr>
<td>12</td>
<td>F</td>
<td>Osteosarcoma</td>
<td>3</td>
<td>Deceased</td>
<td>6</td>
</tr>
<tr>
<td>14</td>
<td>M</td>
<td>Rec Osteosarcoma</td>
<td>3</td>
<td>Deceased</td>
<td>5</td>
</tr>
<tr>
<td>16</td>
<td>F</td>
<td>Ewing’s sarcoma</td>
<td>3</td>
<td>Deceased</td>
<td>6</td>
</tr>
<tr>
<td>27</td>
<td>M</td>
<td>Spindle cell sarcoma</td>
<td>2b</td>
<td>NED</td>
<td>12</td>
</tr>
<tr>
<td>17</td>
<td>M</td>
<td>Ewing’s sarcoma</td>
<td>2b</td>
<td>AWD</td>
<td>9</td>
</tr>
<tr>
<td>11</td>
<td>F</td>
<td>Osteosarcoma</td>
<td>2b</td>
<td>NED</td>
<td>10</td>
</tr>
</tbody>
</table>

(NED= No evidence of disease, AWD= Alive with disease (Metastasis), M= Male, F= Female, GCT= Giant cell tumour)
in position. To maintain the shoulder, contour a shoulder pad is advised to almost all patients for ease of wearing clothes.25,26 However, Myoelectric and body powered prostheses are the predominant options for person with upper limb amputation but the high cost, maintenance and training are challenges incurred by the users.27

CONCLUSION

Although FQA is a significant physically disfiguring procedure, but in large tumours of shoulder girdle, this is a viable option for local control of the disease. Using anteroposterior approach, our results suggest a dismal survivorship after surgery but this is related mainly to the stage of disease at presentation and the size of the tumour. However, even in palliative settings patients report significant pain relief and improvement in quality of life. We did not encounter any significant complications during surgery utilising the anteroposterior approach with reasonable duration of surgery and would recommend this approach for these large tumours.

AUTHORS’ CONTRIBUTION

IK: Literature, study design, data collection, write-up, data interpretation, follow-up. ZK: Study design, data analysis, proof reading. IA: Data analysis, proof reading. AK: Study design, proof reading. MS: Literature search, data collection.

REFERENCES

11. Crosby A. The first operation on record for removal of the entire arm, scapula, and three-fourths of the clavicle. 1875.