SYSTEMATIC REVIEW

Surgical management of upper limb lipoma arborescens: a systematic review

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Abstract

Background: Lipoma arborescens (LA) is a rare benign synovial tumour characterized by the proliferation of mature adipocytes within the synovial cells. Given its rarity, current evidence is mainly based on case reports and case series, and no guidelines are available. The present study investigated the current surgical management and related outcomes of LA in the upper limb.

Methods: This systematic review was conducted following the PRISMA guidelines. PubMed, Scopus, and Virtual Health Library were accessed in September 2021. Clinical studies evaluating patients with LA undergoing surgical treatment were considered eligible for this systematic review. Only studies which reported data on LA located in the upper limb with histopathological confirmation were considered. Articles that reported data from nonsurgical management were not considered.

Results: A total of 21 studies reporting 22 lesions in 21 patients were assessed. The mean age of the patients was 48.48 years (range 22–77). Most studies evaluated the restoration of range of motion and symptom resolution for the functional outcome assessment. Open or arthroscopic excision and synovectomy were the most common surgical procedures for LA. The concomitant lesions were treated in a single-stage procedure. All patients had satisfactory outcomes after open or arthroscopic excision and synovectomy without recurrence at a mean follow-up of 21.14 months (range 2–60). One patient developed postoperative cellulitis (4.55%).

Conclusion: Open and arthroscopic excision combined with synovectomy should be considered the standard treatment option of upper limb LA. Concomitant pathologies can be addressed in a one-stage procedure. Although LA was recognized as a clinical entity decades ago, there is a lack of evidence based guidelines and long term outcome data are unavailable.

Introduction

Lipoma arborescens (LA) is a rare benign synovial tumour characterized by the proliferation of mature adipocytes within the synovial cells [1–5]. Clinical manifestations of LA are nonspecific and frequently resemble osteoarthritis, inflammatory arthritis, or infection [4, 6]. Monoarticular swelling or pain of insidious onset, intermittent joint effusion episodes or a slowly growing subcutaneous mass are common in patients with LA [1, 7]. Magnetic resonance imaging (MRI), using fat suppression or short tau inversion recovery (STIR) sequences point to the diagnosis in most patients with LA [8]. Although its etiology remains unknown [1], it has been hypothesized that LA may result from reactive differentiation of synovial cells towards adipocytes [9]. Two aetiological types have been described. The primary type is considered idiopathic and is mainly observed in younger population [7, 10, 11]. The secondary type is more common in the elderlies, and is associated with pathological...
conditions or lesions causing chronic irritation [7, 12]. The knee is the most frequent location of LA [1–3]; however, lesions of the wrist, elbow, shoulder, ankle, and hip joints have been reported [2, 10, 13–16]. For LA in the knee, arthroscopic synovectomy demonstrated excellent short-term results and a low rate of recurrence [15]. To the best of our knowledge, no review is available concerning the management of LA located in the upper limb. Given its rarity, current evidence is mainly based on case reports and case series, and no guidelines are available. The present study investigated the current surgical management and related outcomes of LA in the upper limb.

Methods
Search strategy
This systematic review was conducted following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. Two investigators (G.K., TMF) independently performed the database search. PubMed, Scopus, and Virtual Health Library were accessed in September 2021. The terms “lipoma arborescens” AND/OR “synovial lipomatosis” AND/OR “villous lipomatous” were used alone and in combination (Additional file 1).

Eligibility criteria
Clinical studies evaluating patients with LA undergoing surgical treatment were considered eligible for this systematic review. Given the authors’ language capabilities, articles published in English or Spanish were eligible. Only studies which reported data on LA located in the upper limb with histopathological confirmation were considered. Screening of the bibliographies of the potentially eligible articles was also performed. Articles that did not clearly state the length of the follow-up were excluded, as were those that did not report quantitative data. Articles that reported data from nonsurgical management were not considered.

Data extraction and outcomes of interest
Two investigators (G.K., TMF) independently reviewed the included studies, and data were extracted to a predefined Excel spreadsheet with the following variables: author, year, type of study, number of women and mean age, history of inflammatory disease and trauma, number and location of the lesions, imaging studies, surgical procedures, length of the follow-up, recurrence, postoperative outcomes.

Methodological quality assessment
The quantitative content assessment was performed using Murad’s tool for evaluating the methodological quality of case reports and case series, which is a modified version of the Newcastle–Ottawa Scale [17]. This scale has been used recently in systematic reviews of case reports and case series [18–21]. The tool has five questions with dichotomic answers. A good assessment has to have five points, moderate four, and low less than three points.

Statistical analysis
Data was presented in tables using absolute values from individual studies. Pooled data were presented as means with standard deviations and percentages. Statistical analysis was performed using SPSS V.19 and Microsoft Excel 2016 (Microsoft®, USA).

Results
Search results
The literature search identified 488 potentially relevant records after the exclusion of duplicates (N=188). Titles and abstracts were screened and 35 articles were retrieved for full-text evaluation. No additional study was identified after citation screening. After full text assessment 14 studies were excluded due to insufficient data regarding follow-up. Twenty-one studies met the predetermined eligibility criteria (Fig. 1).

Methodological quality assessment
The quality assessment was moderate for eight studies and low for 13. No single study was scored as good according to the modification of Murad et al. [17] (Table 1).

Synthesis of results
A total of 21 studies reporting 22 lesions in 21 patients were assessed. The patient demographics is summarized in Table 2. Twelve patients (57.14%) were men and 11 (42.86%) women. The mean age of the patients was 48.48 ± 15.98 years (range 22–77). Fourteen lesions were right-sided, three patients had a history of inflammatory disease, and three had a history of previous trauma.

Imaging findings and surgical treatment outcomes are summarized in Table 3. All patients had single lesion; one has a bilateral presentation [26]. Eleven lesions (50%) were located in the shoulder [1–3, 22, 24, 28, 30, 32, 33, 35, 36], seven (31.82%) in the elbow [13, 23, 25, 26, 29, 34], and four (18.18%) in the wrist [14, 27, 31, 37]. All patients but one had preoperative MRI scans during the diagnostic assessment [27]. Concomitant rotator cuff tears were reported in five patients [1, 3, 24, 35, 38]. Similarly, a labral tear [33], a long head biceps tendon fraying [32], and a distal biceps pathology [34] were concomitant lesions to the LA. Most studies evaluated the restoration of range of motion and symptom resolution for the functional outcome assessment. In one study [13], the
Mayo Elbow Performance Score and Single Assessment Numeric Evaluation were employed. Open or arthroscopic excision and synovectomy were the most common surgical procedures for LA. The concomitant lesions were treated in a single-stage procedure. All patients had satisfactory outcomes after open or arthroscopic excision and synovectomy without recurrence at a mean follow-up of 21.14 ± 18.38 months (range 2–60). One patient developed postoperative cellulitis (4.55%) [37].

**Discussion**

According to the main finding of the present systematic review, patients undergoing surgical excision and synovectomy for LA of the upper limb evidenced satisfactory outcomes regardless of the surgical technique used, with low complication rate and no recurrences at approximately 2 years follow-up.

The aetiology of LA is still controversial. The present systematic review findings did not show a relevant correlation with either inflammatory disease or trauma history. Oni et al. [39, 40] suggested that LA may result from chronic synovitis, and questioned the lesion’s pathognomonic findings found on MRI. On the other hand, Ragab et al. [41] suggested that LA may cause joint inflammatory synovitis, mimicking undifferentiated inflammatory arthritis. The authors highlighted the importance of diagnostic tools such as MRI that led to better decision-making and avoidance of unnecessary disease-modifying
Table 1  Outcomes of Murad’s tool for methodological qualities assessment of case reports and case series [(1) Did the patient(s) represent the whole case(s) of the medical center? (2) Was the diagnosis correctly made? (3) Were other important diagnosis excluded? (4) Were all important data cited in the report? (5) Was the outcome correctly ascertained?]

| Studies                      | 1 | 2 | 3 | 4 | 5 | Assessment |
|------------------------------|---|---|---|---|---|------------|
| Nisolle et al. [22]          | Yes | Yes | Yes | Yes | No | Moderate   |
| Levadoux et al. [23]         | Yes | Yes | Yes | Yes | No | Moderate   |
| Kaneko et al. [24]           | Yes | Yes | No  | Yes | No | Low        |
| Doyle et al. [25]            | Yes | Yes | Yes | Yes | No | Moderate   |
| Dinauer et al. [26]          | Yes | Yes | No  | Yes | No | Low        |
| Yildiz et al. [27]           | Yes | Yes | No  | No  | No | Low        |
| In et al. [28]               | Yes | Yes | No  | Yes | No | Low        |
| Mayayo Sinués et al. [29]    | Yes | Yes | No  | No  | No | Low        |
| Chae et al. [30]             | Yes | Yes | Yes | Yes | No | Moderate   |
| Hill et al. [37]             | Yes | Yes | No  | Yes | No | Low        |
| Benegas et al. [1]           | Yes | Yes | Yes | Yes | No | Moderate   |
| Silva et al. [31]            | Yes | Yes | Yes | Yes | No | Moderate   |
| White et al. [32]            | Yes | Yes | No  | Yes | No | Moderate   |
| Kim et al. [33]              | Yes | Yes | No  | Yes | No | Low        |
| Stepan et al. [14]           | Yes | Yes | No  | Yes | No | Low        |
| Mohammad et al. [34]         | Yes | Yes | Yes | Yes | No | Moderate   |
| Beyth and Safran [2]         | Yes | Yes | No  | Yes | No | Low        |
| Lim et al. [35]              | Yes | Yes | No  | Yes | No | Low        |
| Paccaud and Cunningham [13]  | Yes | Yes | No  | Yes | Yes| Moderate   |
| Kawashima et al. [3]         | Yes | Yes | No  | Yes | No | Low        |
| Elamin et al. [36]           | Yes | Yes | No  | Yes | No | Low        |

Table 2  Patients demographics

| Study                        | Sex | Age | Side | History of inflammatory disease | History of trauma |
|------------------------------|-----|-----|------|---------------------------------|-------------------|
| Elamin et al. [36]           | M   | 55  | L    | No                              | No                |
| Kawashima et al. [3]         | M   | 67  | L    | No                              | No                |
| Paccaud and Cunningham [13]  | M   | 54  | R    | Rheumatoid arthritis            | Not disclosed     |
| Lim et al. [35]              | F   | 38  | R    | No                              | Yes               |
| Beyth and Safran [2]         | M   | 44  | R    | Not disclosed                   | No                |
| Mohammad et al. [34]         | F   | 68  | R    | No disclosed                    | Not disclosed     |
| Kim et al. [33]              | F   | 43  | R    | Not disclosed                   | No                |
| Stepan et al. [14]           | F   | 24  | R    | No                              | Not disclosed     |
| White et al. [32]            | M   | 64  | L    | No                              | Not disclosed     |
| Benegas et al. [1]           | M   | 65  | R    | No                              | No                |
| Hill et al. [37]             | M   | 41  | R    | Not disclosed                   | Yes               |
| Silva et al. [31]            | M   | 45  | L    | No                              | Not disclosed     |
| Chae et al. [30]             | M   | 37  | R    | No                              | No                |
| Mayayo Sinués et al. [29]    | F   | 44  | L    | No                              | No                |
| In et al. [28]               | M   | 22  | L    | No                              | No                |
| Yildiz et al. [27]           | M   | 23  | R    | Not disclosed                   | No                |
| Dinauer et al. [26]          | M   | 37  | R    | No                              | Not disclosed     |
| Doyle et al. [25]            | F   | 50  | L    | Psoriatic arthritis             | Yes               |
| Kaneko et al. [24]           | F   | 77  | L    | No                              | No                |
| Levadoux et al. [23]         | M   | 44  | R    | Psoriatic arthritis             | No                |
| Nisolle et al. [22]          | M   | 44  | R    | No                              | No                |
| Study                          | Number of lesions | Location                      | Imaging studies                                                                                                                                   | Procedure                                                                                          | Follow-up | Recurrence | Postoperative outcomes                                      |
|-------------------------------|-------------------|-------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------|----------------------------------------------------------------------------------------------------|-----------|------------|-------------------------------------------------------------|
| Elamin et al. [36]            | 1                 | Shoulder (subacromial)       | Xray: no, MRI: supraspinatus tendinopathy with a partial tear. Soft tissue mass in the subacromial space measuring 2.5 × 10 × 0.5 cm            | Arthroscopic excision                                                                               | 60 No     | No         | Full active ROM and normal RC strength                     |
| Kawashima et al. [3]          | 1                 | Shoulder (subdeltoid)        | Xray: normal, MRI: subdeltoid fluid villous projections, full-thickness supraspinatus tear                                                      | Arthroscopic synovectomy and RC repair                                                            | 9 No      | No         | Occasional aching, good function                            |
| Paccaud and Cunningham [13]   | 1                 | Elbow (intraarticular)       | Xray: no, MRI: large intra-articular multilobulated pseudo-tumoral mass causing posterior humeroulnar impingement with mixed components including lipomatous and synovial fringes | Arthroscopic synovectomy and posterior humeroulnar decompression                                    | 14 No     | No         | Full ROM, Asymptomatic                                     |
| Lim et al. [35]               | 1                 | Shoulder (subacromial, subdeltoid) | Xray: bony spurs in the acromion and greater tuberosity, MRI: Partial-thickness bursal tear of the supraspinatus tendon, subacromial-subdeltoid bursa fluid-distended fat like nodular projections, greater tuberosity, and lateral acromion osteophytes | Arthroscopic bursectomy, lipoma excision, acromioplasty, and RC repair                              | 5 No      | No         | Asymptomatic                                               |
| Beyth and Safran [2]          | 1                 | Shoulder (intraarticular)    | Xray: Hill Sachs, MRI: joint effusion and synovial hyperplasia                                                                              | Arthroscopic synovectomy and lipoma excision                                                       | 12 No     | No         | Full ROM, Asymptomatic                                     |
| Mohammad et al. [34]          | 1                 | Elbow (antecubital fossa)    | Xray: reactive changes in the radial tuberosity                                                                                            | Open bicipitoradial bursectomy, lipoma excision, and biceps debridement                              | 6 No      | No         | Occasional aching, no calcifications                        |
| Study                | Number of lesions | Location                                      | Imaging studies                                                                                                                                                                                                                                                                                                                                 | Procedure                                                                                                  | Follow-up | Recurrence | Postoperative outcomes                                      |
|---------------------|-------------------|-----------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----------------------------------------------------------------------------------------------------------|-----------|------------|-------------------------------------------------------------|
| Kim et al. [33]     | 1                 | Shoulder (subdeltoid, subacromial)            | MRI: cystic swelling in the right bicipitoradial bursa with peripheral frond-like and ovoid fatty components. Thickening of the distal biceps tendon insertion and hypertrophy of the bicipital radial tuberosity with some associated edema and chronic bicipitoradial bursitis. MRI: paralabral cyst which extends into suprascapular and spinoglenoid notch after a posterior labral tear, SLAP, lipoma in front of the anterolateral cortex of the humeral head, encapsulated mass between infraspinatus and deltoid muscle, villous projections (lipoma arborescens) within the mass with osteochondral metaplasia. Xray: multiple calcifications, enthesophyte at greater tuberosity. | Open lipoma excision, lipoma arborescens excision, and arthroscopic posterior labrum repair.            | 36        | No         | High satisfaction and no limitations                        |
| Stepan et al. [14]  | 1                 | Wrist (dorsal-extensor retinaculum)           | Xray: mass dorsal to the carpus, soft tissue, and fat attenuation. MRI: proliferative tenosynovitis distending the fourth dorsal compartment, containing extensive areas of thick, enhancing tenosynovium as well as macroscopic lobules of subsynovial fat encircling extensor digitorum communis and extensor indicis tendons. | Open tenosynovectomy of the fourth dorsal compartment and fatty mass excision.                         | 3         | No         | Pain-free full shoulder function                            |
| White et al. [32]   | 1                 | Shoulder (bicipital groove)                   | Xray: normal                                                                                                                                                                                                                                                                                                                               | Open synovectomy, lipoma excision, tenodesis, diagnostic arthroscopy.                                    | 6         | No         | Pain-free with full shoulder function and rotation          |
### Table 3 (continued)

| Study                     | Number of lesions | Location                              | Imaging studies                                                                 | Procedure                                      | Follow-up | Recurrence | Postoperative outcomes           |
|---------------------------|-------------------|---------------------------------------|--------------------------------------------------------------------------------|-----------------------------------------------|-----------|------------|----------------------------------|
| Benegas et al. [1]        | 1                 | Shoulder (intraarticular, subacromial) | MRI: frond-like tissue extending from the synovium, which followed the signal intensity of subcutaneous fat on all sequences. The synovium of the glenohumeral joint had no evidence of involvement by this process. | Arthroscopic and open synovectomy, lipoma excision, and RC repair | 4         | No         | Asymptomatic                     |
| Hill et al. [37]          | 1                 | Wrist (dorsal-extensor retinaculum)   | Xray: increased soft tissue. Simple radiography did not show any abnormalities, except for increased soft-tissue volume. MRI: full-thickness tear of the anterior portion of the supraspinatus tendon and significant glenohumeral and subacromial synovitis, with signs of fatty metaplasia | Open lipoma excisional biopsy                  | 2         | No         | Significant improvement. Complication: minor postoperative cellulitis |
| Silva et al. [31]         | 1                 | Wrist (dorsal-extensor retinaculum, also in the knee and ankle) | Xray: soft tissue mass                                                                                             | Open excision                                | 48        | No         | Asymptomatic                     |
| Chae et al. [30]          | 1                 | Shoulder (intraarticular)             | Xray: humeral head erosion                                                                                   | Open synovectomy and lipoma excision          | 12        | No         | Favorable outcome                |
| Mayayo Sinues et al. [29] | 1                 | Elbow (antecubital fossa)             | MRI: well-capsulated, mass-like projections were encircling the right glenohumeral joint and containing a villonodular fat component | Open partial synovectomy                       | 48        | No         | Full ROM                         |
| Study          | Number of lesions | Location                  | Imaging studies                                                                                                                                  | Procedure                                    | Follow-up | Recurrence | Postoperative outcomes |
|---------------|-------------------|---------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------|----------------------------------------------|-----------|------------|------------------------|
| In et al. [28] | 1                 | Shoulder (intraarticular) | MRI: circumscribed mass along the bicipitoradial bursa enveloped the biceps tendon, with a heterogeneous signal bursal effusion and fat tissue deposits similar to small polypoid lesions from the wall to the interior of the mass. MRI: intra-articular frond-like or villous nodules of high signal intensity represent fat. Bone erosion was present at the superior aspect of the humerus. | Arthroscopic synovectomy                      | 12        | No         | Uneventful             |
| Yildiz et al. [27] | 1                     | Wrist (dorsal-extensor retinaculum) | MRI: intra-articular frond-like or villous nodules of high signal intensity represent fat. Bone erosion was present at the superior aspect of the humerus. | Open excision                                | 24        | No         | Asymptomatic           |
| Dinauer et al. [26] | 2 (asynchronous bilateral lesion) | Elbow (bicipitoradial bursa) | MRI: diffuse frond-like, fat-containing lesion involving the bicipitoradial bursa, lipoma arborescens arising from the bicipitoradial bursa was offered. | Open excisional biopsy                       | a) 46; b) 6 No | Good function |                        |
| Doyle et al. [25] | 1                 | Elbow (antecubital fossa) | MRI: lobulated mostly fatty mass anterior to the elbow joint and wrapping around the distal biceps tendon. | Open excisional biopsy                      | 12        | No         | Diminished pain        |
| Kaneko et al. [24] | 1                     | Shoulder (subdeltoid) | MRI: villous mass with surrounding synovial fluid in sub-deltoid bursa FT tear ST. Enormous bursa | Open excisional biopsy and supraspinatus tear open repair | 40        | No         | Full ROM, Residual pain |
| Lebadoux et al. [23] | 1                     | Elbow (anterolateral mass) | MRI: circumscribed mass along the bicipitoradial bursa enveloped the biceps tendon, with a heterogeneous signal bursal effusion and fat tissue deposits similar to small polypoid lesions from the wall to the interior of the mass. MRI: intra-articular frond-like or villous nodules of high signal intensity represent fat. Bone erosion was present at the superior aspect of the humerus. | Open excisional biopsy                      | 48        | No         | Full ROM, Asymptomatic |
| Study          | Number of Lesions | Location                        | Imaging studies                                                                 | Procedure                                      | Follow-up | Recurrence | Postoperative outcomes                                |
|---------------|-------------------|---------------------------------|---------------------------------------------------------------------------------|------------------------------------------------|-----------|------------|-----------------------------------------------------|
| Nisolle et al. [22] | 1                 | Shoulder (subdeltoid, subacromial) | MRI: joint effusion and synovial-based soft tissue mass. Numerous frond-like projections | X-ray: soft tissue swelling Open bursectomy and RC repair | 12 No     | Full ROM, Diminished pain                           |
anti-rheumatic drug prescription [41]. Both theories regarding the aetiopathology of LA concluded that the lesion is closely related to or affected by inflammatory condition. Combining this chronic inflammation with mechanical irritation from the LA mass may predispose patients to other local concomitant lesions.

LA is characterized by typical pathognomonic MRI features. Frond-like architecture synovial mass with fat signal intensity in all sequences and suppression in short tau inversion recovery sequencing or spin-echo, associated with effusion, chemical-shift artifacts at the fat fluid interface without haemosiderin magnetic susceptibility effects, or intravenous contrast enhancement point toward LA. Specific features of the LA may provide useful information and may lead to better management [42, 43]. The included studies in the present systematic review suggested that LA may be present in combination with other concomitant pathological conditions, highlighting the importance of MRI for diagnosis and preoperative planning.

In common with other rare clinical entities, the management of LA lacks evidence-based guidelines. Being a benign lesion, theoretically, if asymptomatic, surgical intervention may not be mandatory [5]. However, to the best of our knowledge, there is no long-term follow-up study observing and examining the natural history of LA. Excision and synovectomy of the affected joint have been proposed as a treatment option. Both open and arthroscopic techniques have been reported, leading to good short-term functional results without recurrences [5, 15]. According to this systematic review, one-stage open or arthroscopic procedures address both LA and potential concurrent pathologies, such as rotator cuff or labral tears, and should be considered as standard treatment option.

This study has several limitations. The limited number of studies included for analysis and related sample size did not allow to infer solid conclusion. The length of the follow-up was limited in all the included studies. Moreover, there was a lack of validated tools in the outcome assessment. Finally, all of the studies included reported no recurrences, mainly based only on symptom regression. The limited length of the follow-up and the absence of imaging at the time of the final evaluation may have under-reported possible recurrences. Given the limited data available for inclusion, comparisons between open and arthroscopic management were not possible to evaluate. However, it is unclear whether lesion size and location may play a role in determining specific approaches. A systematic review on the arthroscopic treatment of LA of the knee revealed a satisfactory short-term outcome [15]. The present study supports similar findings: patients may benefit from less invasive arthroscopic procedures when feasible, as arthroscopic treatment of shoulder [2, 3, 28, 35, 36] and elbow lesions [13] led to promising short-term outcomes. Although LA was recognized as a clinical entity decades ago, the evidence is scarce and long term outcome data are unavailable.

Conclusion
Open and arthroscopic excision combined with synovectomy should be considered the standard treatment option of upper limb LA. Concomitant pathologies can be addressed in a one-stage procedure. Although LA was recognized as a clinical entity decades ago, there is a lack of evidence based guidelines and long term outcome data are unavailable.

Abbreviations
LA: Lipoma arborescens; MRI: Magnetic resonance imaging; STIR: Short tau inversion recovery.

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Authors’ contributions
GK and TMF conceptualization, literature search, data collection. JMH methodological quality assessment. TMF and NS data interpretation and synthesis. SV and MH supervision. FM and NM draft, revision, validation. All authors approved the final version of the manuscript.

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