European Journal of Surgical Oncology 45 (2019) 1520–1526

Contents lists available at ScienceDirect

European Journal of Surgical Oncology

journal homepage: www.ejso.com

Review

Myoepithelioma of the soft tissue: A systematic review of clinical reports

Marco Rastrelli [a, 1], Paolo Del Fiore [a, *, 1], Giovanni Battista Damiani [c, 1], Simone Mocellin [a, b], Saveria Tropea [a, b], Romina Spina [a], Alessandra Costa [a], Francesco Cavallin [d], Carlo Riccardo Rossi [a, b]

[a] Surgical Oncology Unit, Veneto Institute of Oncology IOV – IRCCS, Italy
[b] Department of Surgery, Oncology and Gastroenterology (DISCOG), University of Padova, Italy
[c] Department of Medicine (DIMED), University of Padova, Italy
[d] Independent Statistician, Solagna, Italy

ARTICLE INFO

Article history:
Received 6 March 2019
Received in revised form
26 April 2019
Accepted 2 May 2019
Available online 6 May 2019

Keywords:
Myoepithelioma of soft tissues
Soft tissue sarcoma
Systematic-review

ABSTRACT

Background: Myoepithelioma of the soft tissue (MES) is a rare neoplasm, and available literature includes few studies with small sample sizes, thus limiting the comprehension of this disease. Hence, a systematic review was performed to summarize the available information on MES patients evaluated for surgical treatment.

Methods: The MEDLINE/PubMed, EMBASE and SCOPUS databases were reviewed for eligible studies in January 2019. Inclusion criteria were: patients with MES; indication for wide resection or amputation; observational studies; articles published from 1997 to 2018; English language; reporting of outcome measures such as overall survival (OS), disease-free survival (DFS), recurrence, metastases. Studies not including humans were excluded. Quality appraisal was performed using the Methodological Index for Non-Randomized Studies (MINORS) tool. A narrative synthesis of included studies was conducted, a formal meta-analysis being unfeasible.

Results: Overall, 10 eligible studies including 233 MES patients were identified. The most frequent tumor sites were the limbs and trunk. Most patients underwent surgical treatment, with R0 resection margin rates ranging from 24% to 78%. During follow-up, local recurrence rates ranged from 17% to 50, and distant metastasis rates varied 8%–48%. At 5 years, OS rates ranged from 8% to 94% and DFS rates from 11% to 40%. The average MINORS score was 9.1 (range: 6–11).

Conclusions: Surgical resection was often sub-optimal. The long-term prognosis was poor, with a non-negligible rate of disease recurrence and metastasis. On average, the quality of available information is moderate. The centralization of patient information in large international registries is warranted to provide a better understanding of MES biology and ultimately improve patient outcomes.

Introduction

Myoepithelioma is a rare neoplasm that usually occurs in the salivary glands [1]. The World Health Organization lists myoepithelioma in the category of "tumor of uncertain differentiation", along with myoepithelial carcinoma and mixed tumors [2].

Myoepithelioma of the soft tissue (MES) is an even more rare entity which may occur in several anatomic locations, the most common being the trunk, upper limb and legs [3,4]. In 1997, Kilpatrick et al. characterized MES for the first time in a case series

* Corresponding author. Surgical Oncology Unit, Veneto Institute of Oncology IOV – IRCCS, Via Gattamelata 64, 35128, Padua, Italy.
E-mail addresses: marco.rastrelli@iov.veneto.it (M. Rastrelli), paolo.delfiore@iov.veneto.it (P. Del Fiore), damianigv@libero.it (G.B. Damiani), simone.mocellin@unipd.it (S. Mocellin), saveria.tropea@iov.veneto.it (S. Tropea), romina.spina@iov.veneto.it (R. Spina), alessandra.costa@iov.veneto.it (A. Costa), cescocava@libero.it (F. Cavallin), carlo.rossi@unipd.it (C.R. Rossi).
[1] These authors contributed equally to the work.

https://doi.org/10.1016/j.ejso.2019.05.003
0748-7983/

study [3]. In 2003, Hornick et al. reported clinico-pathologic and immuno-histochemical characteristics of 101 cases [4]. This neoplasm is clinically heterogeneous; the tumors with severe cytological atypia or high grade are classified as myoephitelial carcinomas, while the tumors with low cytological atypia are classified as myoephitelioma [4]. Current approach to MES usually includes surgical resection (i.e. wide excision or amputation), while the role of chemotherapy remains unclear [5].

So far, available literature on surgical treatment and outcome in cohorts of MES patients includes single-center retrospective case series [3,4,6–13]. In addition, most studies include only few patients, therefore summarizing the available data may provide useful and more comprehensive information on MES to researchers and clinicians.

This systematic review aimed to overview available information on characteristics, treatment and outcome of MES patients evaluated for surgical treatment.

Materials and methods

Study design

This is a systematic review of observational studies on MES patients receiving evaluation for surgical treatment. The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) recommendations were followed in this review [14].

Search strategy

To identify relevant studies, we systematically searched MEDLINE/PubMed, EMBASE and SCOPUS in January 2019. Two researchers (PDF and GDB) independently reviewed search results, screened titles/abstracts, and excluded duplicate articles (n = 69). Two researchers (MR and SM) resolved any inconsistency. We obtained the full texts of all potentially eligible studies. In PubMed, the following search strategy was used: “myoepithelioma” AND “soft tissue“. This search strategy was adapted to suit the other electronic sources. Only clinical studies in English were considered. The reference lists of included articles were hand-searched to identify additional studies of interest.

Criteria for considering studies for this review

Observational studies reporting data on MES patients were considered eligible for inclusion in this review. Inclusion criteria were:

- subjects: patients with myoepithelioma of soft tissue (both adult and pediatric patients);
- interventions: indications for wide resection or amputation;
- outcomes measures: overall survival (OS), disease-free survival (DFS), disease recurrence, distant metastases;
- type of study: observational studies (including retrospective case series);
- language: English;
- publication year: 1997–2018.

Studies not including humans were excluded

Data extraction

Two researchers (GBD and AC) independently extracted key data from included studies. A third researcher (PDF) checked the extracted data. For each study, extracted data included: study characteristics (study design, follow-up periods), patient characteristics (median age, sex distribution), tumor characteristics (tumor site, tumor depth, tumor size, malignancy grade, cytological atypia, necrosis, mitoses), treatment (neo-adjuvant therapy, type of surgery and surgical margin), clinical outcomes (recurrence, metastases) and survival (overall survival, disease-free survival). Contacts with authors in order to retrieve additional data was not necessary.

Quality assessment

Two researchers (PDF, GBD) completed the quality appraisal with the Methodological Index for Non-Randomized Studies (MINORS) tool [15]. The MINORS tool includes eight items for non-comparative studies and four additional items for comparative studies. Items are scored as 0 (not reported), 1 (reported but inadequate) and 2 (reported and adequate), with a total score of 16 for non-comparative studies and 24 for comparative studies. A third researcher (FC) resolved any inconsistency.

Data synthesis

A narrative synthesis of included studies was conducted, because some aspects limited the feasibility of a meaningful meta-analysis. Such aspects included the very small sample size (less than 20 patients) of some studies, the inclusion of both adults and children, and the heterogeneous intent in the surgical approach (i.e. some patients might have not been properly recognized as MES, thus receiving low radical surgery).

Results

Study selection

Overall, the searches yielded 436 non-duplicated articles; 361 articles were excluded based on title and/or abstract, while 75 articles were retrieved for full text review. Sixty-five case reports were excluded. Finally, ten observational studies were included in the qualitative synthesis (Fig. 1) [3,4,6–13].

Study and patient characteristics

Nine studies were retrospective observational studies [3,4,6–8,10–13] and one was a case series [9]. Seven studies [7–13] were published within the last 12 years (2007–2018). Studies included from 4 to 101 patients, with a total of 233 patients (229 from retrospective studies and 4 from the case series) included in this review. Patient characteristics of the included studies are reported in Table 1. The median age was 43 years and 54% of patients were males. Overall, 53% of patients were pediatrics, with one study including only pediatric patients [7].

Tumor characteristics

Tumor characteristics are reported in Table 1. The most frequent site was the limbs (upper and lower) and the trunk. MES was superficial and localized in the subcutaneous layer in eight studies [3,4,6,7,9,11–13], while it was predominantly in the intermediate but supra-axial layers in one study [10] and in the deep fascial intramuscular layers in one study [8]. The median tumor size in cm varies from 1 to 7 cm. Only three studies reported the degree of

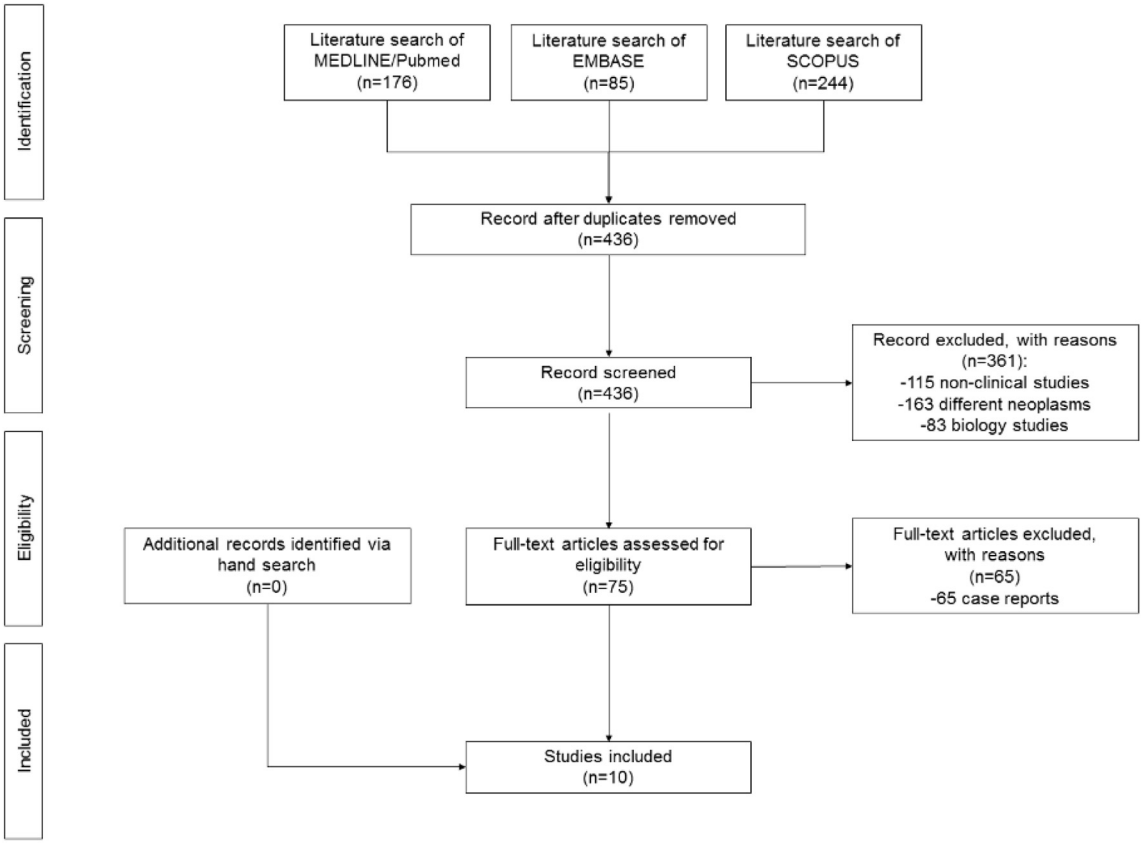

Fig. 1. Flowchart of study inclusion.

malignancy [12–14]. The percentage of severe atypia was above 50% in five studies [7,9–12] (not reported in one [13]), while the percentage of necrosis ranged from 10% to 50% in five studies [3,7,9,10,12] and was present in two [6,8] (not reported in three [4,11,13]). Median number of mitoses ranged from 1 to 11.25 in eight studies and was not detectable in two studies [11,13].

Treatment

Treatment approach is reported in Table 2. The vast majority of patients underwent surgical treatment, while 11 patients underwent surgical evaluation but did not receive surgical resection due to severe comorbidities. Wide excision was performed in almost all surgical approaches, while four patients underwent limb amputation [7,10] and 10 patients had other surgical treatments (i.e. biopsy, lymph node dissection). The percentage of patients with R0 resection margin ranged from 24% to 78%. In Gleason et al. three patients received chemotherapy and one was treated with isolated limb perfusion (ILP) (one study [7]) with no subsequent surgical resection.

Outcomes

Oncologic and survival outcomes are reported in Table 3. In these studies the outcomes were described for the entire cohort and no separate data for potentially interesting subgroups (such as R0 vs R1 vs R2) were available. Overall, follow-up ranged from 12 to 84 months in nine studies, while it was not reported in one [11]. Follow-up for all patients was available only in three studies [6,12,13]. During follow-up, the proportion of patients with recurrence ranged from 17% to 50% (10 studies), and that with metastasis from 8% to 48% (eight studies). The most frequent metastatic site was the lung.

Nine studies reported OS data, while only five studies reported DFS data. However, the timing of reporting (1, 3 and/or 5 years) was heterogeneous among included studies, thus limiting any attempt to summarize the data (Table 3). OS ranged from 21% to 100% at 1 year (seven studies), from 11% to 44% at 3 years (five studies) and from 8% to 94% at 5 years (eight studies). DFS ranged from 22% to 100% at 1 year (two studies), from 11% to 71% at 3 years (three studies) and from 11% to 40% at 5 years (three studies).

Quality assessment

The MINORS scores for the methodological quality appraisal (range 0–16) of included articles is shown in Supplementary Table 1. All included articles were non-comparative prospective or retrospective cohort studies with an average MINORS score of 9.1 (range 6–11). All studies scored 0 in the following items: unbiased assessment of the study endpoint and prospective calculation of the study size.

Discussion

MES is a rare neoplasm with a heterogeneous manifestation, and

Table 1
Patient and Tumor characteristics.

									Tumor site				Tumor depth[a]			Tumor size	Malignancy grade				
Author	Year	Country	N of pts	Median age, years	Females, n	Males, n	Adult, n	Pediatric, n	Head or neck, %	Trunk, %	Lower limb, %	Upper limb, %	Superficial, %	Intermediate, %	Deep, %	Median, cm	Low Grade, %	High Grade, %	Severe atypia, %	Necrosis, %	Mitoses, median
Kilpatrick [3]	1997	USA	19	30	7	12	14	5	5	21	32	37	73	27	0	2	–	–	47	10	0.84
Michal [6]	1999	Czech Republic	12	58	3	9	12	0	25	33	33	8	10	1	1	2.75	–	–	42	0	0.83
Hornick [4]	2003	USA	101	–	48	53	85	16	15	10	35	41	53	22	25	1	52	48	39	–	8.35
Gleason [7]	2007	USA	29	9	15	14	0	29	13	28	38	21	73	27	0	4.7	–	–	52	31	1
Hallor [8]	2008	Sweden	6	58	4	2	5	1	16.6	16.6	50	16.6	0	0	100	7	–	–	33	0	2.16
Meenakshi [9]	2009	Northern Ireland	4	43	4	0	4	0	0	100	0	0	100	0	0	2.25	–	–	100	50	11.25
Flucke [10]	2012	Netherlands	10	42	5	5	10	0	12	33	22	33	22	77	1	4	–	–	100	20	2.7
Rekhi [11]	2012	India	14	–	2	12	14	0	0	71	8	21	5	–	–	–	–	–	64	–	–
Yoshida [12]	2015	Japan	9	41	9	0	9	0	0	100	0	0	100	0	0	3.25	33	77	100	44	5.88
Domingo Musibay [13]	2016	USA	20	53	11	9	20	0	0	50	25	25	80	0	0	2.4	65	35	–	–	–

[a] The distinction of MES in superficial, intermediate, and deep is defined by the distribution of these in the anatomical planes, respectively subcutaneous fascial and intramuscular.

Table 2
Treatment.

						Surgical approach			Resection[b]		
Author	CT, n	RT, n	ILP, n	Surgery, n	Surgery, %	Wide excision	Amputation	Other[a]	R0, %	R1, %	R2, %
Kilpatrick [3]	0	1	0	19	100	19	0	0	78	22	0
Michal [6]	0	0	0	12	100	12	0	0	66	25	9
Hornick [4]	0	6	0	100	99	99	0	0	57	36	7
Gleason [7]	3	4	1	23	79	17	3	3	24	60	16
Hallor [8]	0	0	0	6	100	5	0	0	67	33	0
Meenakshi [9]	0	0	0	4	100	4	0	0	75	25	0
Flucke [10]	0	0	1	10	100	7	1	0	70	10	20
Rekhi [11]	0	0	0	11	79	11	0	0	56	22	22
Yoshida [12]	0	0	0	9	100	6	0	3	67	23	10
Domingo Musibay [13]	0	4	0	19	95	19	0	0	63	37	0

CT: chemotherapy. ILP: isolated limb perfusion. RT: radiation therapy.
[a] Debulking and marginal excision.
[b] The margin is categorized either as grossly positive (R2), microscopically positive (R1), or microscopically negative (R0).

Table 3
Clinical outcomes.

Author	Pts with FU n	Pts with recurrence		Pts with metastases		Site of metastases (not cumulative)			FU	Overall survival			Disease-free survival		
		N	%	N	%	Meta Inf, %	Meta liver, %	Meta Lung, %	Median follow-up, months	1 y, %	3 ys, %	5 ys, %	1 y, %	3 ys, %	5 ys, %
Kilpatrick [3]	10	2	20	2	20	50	0	50	29	21	16	16	–	–	–
Michal [6]	12	2	17	1	8	0	0	100	61	33	25	25	–	27	–
Hornick [4]	64	19	30	10	16	50	0	50	49	–	–	–	–	–	–
Gleason [7]	25	12	48	12	48	42	8	75	18	23	12	8	–	–	–
Hallor [8]	5	2	40	1	20	0	0	100	80	–	–	60	–	–	40
Meenakshi [9]	2	1	50	–	–	–	–	–	12	100	–	–	100	–	–
Flucke [10]	9	2	22	3	33	22	0	100	36	66	44	33	–	–	–
Rekhi [11]	9	3	33	3	33	33	0	67	–	22	11	11	22	11	11
Yoshida [12]	9	3	33	–	–	–	–	–	67	56	–	44	–	–	–
Domingo	20	7	35	4	20	–	–	–	84	–	–	94	–	71	36
Musibay [13]															

FU: follow-up.

radical wide surgical excision is an important part of the therapeutic process, as the early local recurrence rate is not negligible [4]. However, available literature includes few studies with limited sample size, thus limiting comprehension of the disease. In the present work, we summarize available information on clinico-pathological characteristics, treatment and clinical outcomes of MES patients receiving surgical evaluation, in order to provide useful and more comprehensive information on MES to researchers and clinicians.

Our synthesis showed that MES occurred equally in both males and females. As regards onset age, the majority of patients were adults; however, we cannot rule out that some authors have intentionally excluded pediatric cases, which prevents us from defining the actual disease burden in children [3,4,6,8–13].

MES was mainly superficial and localized in the subcutaneous tissue, and the most frequent tumor site were the limbs (upper and lower), followed by trunk and head/neck [3,4,6–8,10–13]. It is noteworthy that some clinical presentations of MES in the limbs resemble other dermatologic/cutaneous lesions [16], such as vulgaris verruca or cutaneous fibroma, thus increasing the likelihood of inappropriate identification and inadequate treatment.

Immuno-histochemical and bio-molecular patterns have been investigated in non-clinical studies (i.e. laboratory and pathology studies) that are outside the scope of this systematic review. However, our review confirmed that MES usually showed a marked cytologic atypia, necrosis and high number of mitoses [3,4,6,8–10,12], as previously suggested in non-clinical studies [17,18]. Interestingly, malignancy grade was reported only in three studies [4,12,13]. Given the inclusion of other cellular data (atypia, necrosis, mitoses) from histopathologic report, we think that the underreporting of malignancy grade may be an author's choice in drafting the manuscript.

In the included studies, the vast majority of patients received wide excision with curative intent [3,4,6–13].

Only few patients received neo-adjuvant radiotherapy [3,4,7,13], and very few patients received chemotherapy with palliative intent for metastatic disease [7]. However, we think the high rate of R1 and/or R2 resection margins in seven studies [4,6–8,10,11,13] might suggest inadequate knowledge of the potential malignant behavior of MES leading to non-surgically radical excision. Therefore, increasing awareness and recognition of MES at diagnosis stage may help to referring the patient to a physician specialized in soft tissue sarcoma care.

Available information in literature suggested a poor long-term prognosis for MES patients, with a recurrence rate ranging from 17% to 50% and a metastasis rate from 8% to 48% [3,4,6–12].

These studies show a potential association between local recurrence and the high rate of R1 and/or R2 resection margins [3,4,6–12]; however, the correlation of non-radical surgery with distant metastatic recurrence needs additional evidence to be proved. Moreover, the rarity of the disease and the scarce knowledge of its biological behavior might lead to underestimate its malignant potential and explain the relatively high rates of sub-optimal surgical resection, which in turn could at least in part be responsible for the reported averagely poor prognosis. Unfortunately, the small sample size in most studies and the heterogeneous timing of reporting of survival data prevented any meaningful summarizing analysis. Interestingly, one study reported a very good long-term survival despite the non-negligible rate of recurrence and metastases occurring during follow-up [13]. This result might have been associated with patient care in a referral center with specialized staff (including a soft tissue pathologist) and dedicated follow-up.

The quality of the studies included in this review was limited,

since it was penalized by the retrospective nature of all studies, the un-blinded assessment of the study endpoints, and the loss of patients during follow-up. These weak points are common in studies on rare diseases, which are often investigated with a retrospective review of hospital medical records. Given the rarity of these tumors and the generally poor outcomes likely due to variability in surgical treatment, patients with suspected MES should be addressed to a sarcoma center guarantee a correct pathological assessment as well as an adequate surgical treatment by a team of physicians dedicated to sarcoma management.

In addition, the centralization of patient information in a large international registry may provide a better understanding on epidemiological, pathogenetic, prognostic and therapeutic aspects regarding rare diseases such as MES. To this aim, we intend to build a web-based platform to enable worldwide physicians to share clinico-pathological data on patients affected with MES.

Overall, the present work is the first systematic review summarizing the available information on MES patients receiving evaluation for surgical treatment. There is a growing interest on this topic as shown by the increasing number of reports in the last 10 years [19–21]. Given the rarity of the neoplasm, patient care can benefit from the summary of available information on this disease. However, the findings of our review should be interpreted with caution due to its limitations. First, the quality of included studies was limited as above mentioned. Second, seven out of ten studies had a small sample size (fewer than 20 patients). Third, some aspects of the included studies (i.e. some very small sample sizes, inclusion of both adults and children, and heterogeneous intent in the surgical approach) prevented us from conducting a meaningful meta-analysis. Finally, only studies in English were evaluated, which might have led to miss some information reported in non-English language articles.

Conclusion

The vast majority of MES patients received wide excision with curative intent, but the surgical resection was often sub-optimal. The long-term prognosis was poor, with a non-negligible rate of local recurrence and distant metastasis. Available information is currently provided by few small retrospective studies with limited quality. This is why an international effort to collect data in an individual patient data repository is eagerly awaited.

Declaration of authorship and conflict of interest

All authors, *Paolo Del Fiore, Marco Rastrelli, Giovanni Battista Damiani, Simone Mocellin, Saveria Tropea, Romina Spina, Alessandra Costa, Francesco Cavallin, Carlo Riccardo Rossi* have seen and approved the final version of the manuscript (Myoepithelioma of the soft tissue: a systematic review of clinical reports) study being submitted. They warrant that the article is the authors' original work, has not received prior publication and is not under consideration for publication elsewhere.

Conflict of interest

All the authors declare no conflict of interest.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ejso.2019.05.003.

Supplementary Table 1
Critical appraisal of included studies using the MINORS tool (for non-comparative studies)

Author	A clearly stated aim	Inclusion of consecutive patients	Prospective collection of data	Endpoints appropriate to the aim of the study	Unbiased assessment of the study endpoint	Follow-up period appropriate to the aim of the study	Loss to follow up less than 5%	Prospective calculation of the study size	Total score *
Kilpatrick [3]	2	2	1	2	0	2	0	0	9
Michal [6]	2	2	1	2	0	2	0	0	9
Hornick [4]	2	2	1	2	0	2	0	0	9
Gleason [7]	2	2	1	2	0	2	0	0	9
Hallor [8]	2	2	1	2	0	2	2	0	11
Meenakshi [9]	2	0	1	2	0	1	0	0	6
Flucke [10]	2	2	1	2	0	2	0	0	9
Rekhi [11]	2	2	1	2	0	2	2	0	11
Yoshida [12]	2	2	1	2	0	2	0	0	9
Domingo Musibay [13]	2	2	1	2	0	2	0	0	9

* Items are scored as 0 (not reported), 1 (reported but inadequate) and 2 (reported and adequate), with a total score of 16 for non-comparative studies.

References

[1] Jo VY, Fletcher CD. Myoepithelial neoplasms of soft tissue: an updated review of the clinicopathologic, immunophenotypic, and genetic features. Head Neck Pathol 2015;9:32–8.

[2] Fletcher CDM, Bridge JA, Hogendoorn P, Mertens F. WHO classification of tumours of soft tissue and bone. fourth ed., vol. 5. Lyon: IARC Press; 2013.

[3] Kilpatrick SE, Hitchcock MG, Kraus MD, Calonje E, Fletcher CD. Mixed tumors and myoepitheliomas of soft tissue: a clinicopathologic study of 19 cases with a unifying concept. Am J Surg Pathol 1997;21:13–22.

[4] Hornick JL, Fletcher CD. Myoepithelial tumors of soft tissue: a clinicopathologic and immunohistochemical study of 101 cases with evaluation of prognostic parameters. Am J Surg Pathol 2003;27:1183–96.

[5] Kabarriti R, Quinn TJ, Ewart MR, Mehta KJ, Lomita C, Geller DS, et al. Neoadjuvant radiation therapy for the management of myoepithelial carcinoma of the upper extremity. Int J Cancer 2018;142:854–62.

[6] Michal M, Miettinen M. Myoepitheliomas of the skin and soft tissues. Report of 12 cases. Virchows Arch: An International Journal of Pathology 1999;434:393–400.

[7] Gleason Briana C, Fletcher Christopher DM. Myoepithelial carcinoma of soft tissue in children: an aggressive neoplasm analyzed in a series of 29 cases. Am J Surg Pathol 2007;31(12):1813–24.

[8] Hallor Karolin H, Manuel R, Teixeira Christopher, Fletcher DM, Bizarro Susana, Staaf Johan, Domanski Henryk A, , Fredrik Vult von Steyern, Panagopoulos Ioannis, Mandahl Nils, Mertens Fredrik. Heterogeneous genetic profiles in soft tissue myoepitheliomas. Mod Pathol: An Official Journal of the United States and Canadian Academy of Pathology, Inc 2008;21(11):1311–9.

[9] Meenakshi Meenakshi, Glenn McCluggage W. Myoepithelial neoplasms involving the vulva and vagina: report of 4 cases. Hum Pathol 2009;40(12):1747–53.

[10] Flucke Uta, Bastiaan BJ, Tops Marian, A.J. Verdijk, Patricia JH, van Cleef, et al. NR4A3 rearrangement reliably distinguishes between the clinicopathologically overlapping entities myoepithelial carcinoma of soft tissue and cellular extraskeletal myxoid chondrosarcoma. Virchows Arch 2012;460(6):621–8.

[11] Rekhi Bharat, Sable Mukund, Nirmala A, Jambhekar. Histopathological, immunohistochemical and molecular spectrum of myoepithelial tumours of soft tissues. Virchows Arch: An International Journal of Pathology 2012;461(6):687–97.

[12] Yoshida, Akihiko, Hiroshi Yoshida, Masayuki Yoshida, Taisuke Mori, Eisuke Kobayashi, et al, 2015. Myoepithelioma-like tumors of the vulvar region: a distinctive group of SMARCB1-deficient neoplasms. Am J Surg Pathol 39(8):1102–13.

[13] Domingo-Musibay, Evidio Andre M Oliveira, Okuno Scott H, Petersen Ivy A, Peter S. Rose, Robinson Steven I. Myoepithelioma of soft tissues: a single institution retrospective case series. Am J Clin Oncol 2018;41(4):357–61.

[14] Systematic Reviews. In: CRD's guidance for undertaking reviews in Health care. York, UK: Centre for Reviews and Dissemination, University of York; 2009. .Hutton, B.; Salanti, G.; Caldwell, D.M.; Chaimani, A.; Schmid, C.H.; Cameron, C.; Ioannidis, J.P.A.; Straus, S.;Thorlund, K.; Jansen, J.P.; et al. The PRISMA Extension Statement for Reporting of Systematic Reviews Incorporating Network Meta-analyses of Health Care Interventions: Checklist and Explanations. Ann. Intern.Med. 2015, 162, 777.

[15] Slim K, Nini E, Forestier D, Kwiatkowski F, Panis Y, Chipponi J. Methodological index for non-randomized studies (minors): development and validation of a new instrument. ANZ J Surg 2003;73:712–71.

[16] Rastrelli M, Passuello N, Cecchin D, Basso U, Tosi AL, Rossi CR. Metastatic malignant soft tissue myoepithelioma: a case report showing complete response after locoregional and systemic therapy. J Surg Case Rep 2013;2013(12).

[17] Rekhi B, Sable M, Jambhekar NA. Histopathological, immunohistochemical and molecular spectrum of myoepithelial tumours of soft tissues. Virchows Arch 2012;461:687–97.

[18] Fletcher CD. Recently characterized soft tissue tumors that bring biologic insight. Mod Pathol 2014;27(Suppl 1):S98–112.

[19] Guo L, Zhou F, Zhang N, Dai H, Zeng X, Gong H. Myoepithelioma of the lateral abdominal wall: a case report. Medicine (Baltim) 2018;97:e11209.

[20] Kabarriti R, Quinn TJ, Ewart MR, Mehta KJ, Lomita C, Geller DS, et al. Neoadjuvant radiation therapy for the management of myoepithelial carcinoma of the upper extremity. Int J Cancer 2018;142:854–62.

[21] LeBlanc RE, Taube J. Myofibroma, myopericytoma, myoepithelioma, and myofibroblastoma of skin and soft tissue. Surg Pathol Clin 2011;4:745–59.