**Supplementary Appendix Content**

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# Table S1: JBI Critical Appraisal Checklist for Case Reports.

|  |  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Checklist** | **Sacco et al. 1996** | **Davies et al. 2000** | **Thatayatikom et al. 2005** | **Matsubara et al. 2008** | **Boujaoude et al. 2013** | **Vitale et al. 2015** | **Jolles et al. 2016** | **Tashtoush et al. 2016** | **Pathria et al. 2016** | **Hasegawa et al. 2017** |
| 1. Were patient’s demographic characteristics clearly described? | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes |
| 2. Was the patient’s history clearly described and presented as a timeline? | Yes | Yes | No | Yes | no | Yes | Yes | Yes | Yes | Yes |
| 3. Was the current clinical condition of the patient on presentation clearly described? | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes |
| 4. Were diagnostic tests or assessment methods and the results clearly described? | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes |
| 5. Was the intervention(s) or treatment procedure(s) clearly described? | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes |
| 6. Was the post-intervention clinical condition clearly described? | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes |
| 7. Were adverse events (harms) or unanticipated events identified and described? | Yes | Yes | No | Yes | NA | NA | NA | NA | NA | Yes |
| 8. Does the case report provide takeaway lessons? | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes |
| **Total score** | 8 | 8 | 6 | 8 | 6 | 7 | 7 | 7 | 7 | 8 |

UC, unclear; NA, not applicable.

**Table S1: Continued.**

|  |  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Checklist** | **Zdziarski et al. 2017** | **Routes et al. 2017** | **Arraya et al. 2018** | **Shah et al. 2018** | **Sood et al. 2018** | **Tillman et al. 2019** | **Tessarin et al. 2019** | **Cowen et al. 2020** | **Pattanaik et al. 2021** | **Nishimura et al. 2022** |
| 1. Were patient’s demographic characteristics clearly described? | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes |
| 2. Was the patient’s history clearly described and presented as a timeline? | Yes | Yes | Yes | No | Yes | Yes | Yes | Yes | Yes | Yes |
| 3. Was the current clinical condition of the patient on presentation clearly described? | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes |
| 4. Were diagnostic tests or assessment methods and the results clearly described? | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes |
| 5. Was the intervention(s) or treatment procedure(s) clearly described? | NA | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes |
| 6. Was the post-intervention clinical condition clearly described? | NA | Yes | Yes | NO | Yes | Yes | Yes | Yes | Yes | Yes |
| 7. Were adverse events (harms) or unanticipated events identified and described? | NA | Na | NA | NA | Yes | NA | NA | Yes | Yes | NA |
| 8. Does the case report provide takeaway lessons? | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes |
| **Total score** | 5 | 7 | 7 | 5 | 8 | 7 | 7 | 8 | 8 | 7 |

UC, unclear; NA, not applicable.

# Table S2: JBI Critical Appraisal Checklist for Case series.

|  |  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Checklist** | **Franxman et al. 2014** | **Rao et al. 2015** | **Ng et al. 2019** | **Beaton et al. 2020** | **Strunz et al. 2021** | **Ruiz-Alcaraz et al. 2021** | **Perlman et al. 2021** | **Bucciol et al. 2017** | **Deyà‐Martínez et al. 2018** | **Pac et al. 2020** |
| 1. Were there clear criteria for inclusion in the case series? | Yes | Yes | Yes | No | Yes | Yes | No | NO | Yes | Yes |
| 2. Was the condition measured in a standard, reliable way for all participants included in the case series? | Yes | Yes | Yes | Yes | Yes | Yes | Yes | No | Yes | No |
| 3. Were valid methods used for identification of the condition for all participants included in the case series? | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes |
| 4. Did the case series have consecutive inclusion of participants? | NO | No | No | UC | Yes | Yes | No | Yes | No | Yes |
| 5. Did the case series have complete inclusion of participants? | No | No | No | UC | Yes | Yes | No | No | No | Yes |
| 6. Was there clear reporting of the demographics of the participants in the study? | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes |
| 7. Was there clear reporting of clinical information of the participants? | Yes | Yes | Yes | Yes | UC | Yes | Yes | Yes | Yes | Yes |
| 8. Were the outcomes or follow up results of cases clearly reported? | Yes | Na | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes |
| 9. Was there clear reporting of the presenting site(s)/clinic(s) demographic information? | Yes | Yes | UC | Yes | Yes | Yes | No | NA | Yes | Yes |
| 10. Was statistical analysis appropriate? | Na | Yes | NA | NA | NA | NA | NA | NA | Na | NA |
| **Total score** | 7 | 7 | 6 | 6 | 8 | 9 | 5 | 5 | 7 | 8 |

UC, unclear; NA, not applicable.

# Table S3: Quality of included cohort studies (Newcastle-Ottawa Scale, NOS).

|  |  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Author Year** | **Selection** | | | | **Comparability of cohorts on the basis of the design or analysis** | **Outcome** | | |  | |
|  | **Representativeness of the exposed** | **Selection of the non-exposed** | **Ascertainment of exposure** | **Outcome not present at start** | **Assessment of outcome** | **follow-up length** | **Adequacy of follow up of** | **Total Quality Score** |
| Bates et al. 2004 | 0 | \* | \* | 0 | \* | \* | \* | \* | 6 |
| Torigian et al. 2008 | 0 | 0 | \* | 0 | \* | \* | \* | \* | 5 |
| Van de ven et al. 2011 | \* | 0 | \* | 0 | \* | \* | \* | \* | 6 |
| Chase et al. 2013 | \* | 0 | \* | 0 | \* | \* | \* | \* | 6 |
| Kollert et al. 2014 | 0 | 0 | \* | 0 | \* | \* | 0 | 0 | 3 |
| Maglione et al. 2014 | \* | 0 | \* | 0 | \* | \* | \* | \* | 6 |
| Maglione et al.2015 | \* | 0 | \* | 0 | \* | \* | \* | \* | 6 |
| Maglione et al. 2019 | 0 | 0 | \* | 0 | \* | \* | \* | \* | 5 |
| Verbsky et al. 2020 | \* | \* | \* | 0 | \* | \* | \* | \* | 7 |
| Fraz et al. 2020 | \* | 0 | \* | 0 | \* | \* | 0 | 0 | 4 |
| Lopez et al. 2020 | \* | 0 | \* | 0 | \* | \* | \* | \* | 6 |
| Meerburg et al. 2020 | 0 | 0 | \* | \* | \* | \* | \* | \* | 6 |
| Szczawinska et al. 2021 | 0 | 0 | \* | 0 | \* | \* | \* | 0 | 4 |
| Lopes et al. 2021 | \* | 0 | \* | 0 | \* | \* | \* | \* | 6 |
| Fraz et al. 2022 | \* | \* | \* | 0 | \* | \* | 0 | 0 | 5 |
| Bintalib et al. 2022 | 0 | 0 | \* | 0 | \* | \* | \* | \* | 5 |
| Smits et al. 2022 | 0 | \* | \* | \* | \* | \* | \* | \* | 7 |

# Table S4: Quality of included case-control studies (NOS).

|  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
|  | **Selection** | | | | **Comparability of Cases and Controls on the Basis of the Design or Analysis** | **Outcome** | | |  |
| **Author Year** | **Is the Case Definition Adequate?** | **Representativeness of the Cases** | **Selection of Controls** | **Definition of Controls** | **Ascertainment of Exposure** | **Same method of ascertainment for cases and controls** | **Non-Response Rate** | **Total Quality Score** |
| Bouvry et al. 2013 | \* | \* | 0 | \* | \* | \* | \* | 0 | 6 |
| Mannina et al. 2016 | \* | \* | \* | \* | \* | \* | \* | 0 | 7 |
| Hartono et al. 2017 | \* | \* | \* | \* | \* | \* | \* | 0 | 7 |
| Friedmann et al. 2021 | \* | \* | 0 | 0 | \* | \* | \* | 0 | 5 |

# Table S5: Quality of included cross sectional studies (NOS- adapted for cross sectional studies).

|  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- |
|  | **Selection** | | | | **Comparability of cohorts on the basis of the design or analysis** | **Outcome** | |  |
| **Author Year** | **Representativeness of the sample:** | **Sample size** | **Non-respondents** | **Ascertainment of the exposure** | **Assessment of outcome** | **Statistical test** | **Total Quality Score** |
| Maarschalk-Ellerbroek et al. 2014 | 0 | 0 | 0 | \* | \* | \*\* | \* | 5 |
| Hanitsch et al. 2019 | \* | 0 | 0 | \* | \* | \*\* | 0 | 4 |
| Cinetto et al. 2021 | \* | \* | 0 | \* | \* | \* | \* | 6 |
| Cabanero et al. 2022 | \* |  | 0 | \* | \* | \* | \* | 5 |

The star rating according to the Newcastle-Ottawa Scale indicate; **Good quality**: 3 or 4 stars in selection domain AND 1 or 2 stars in comparability domain AND 2 or 3 stars in outcome/exposure domain. **Fair quality:** 2 stars in selection domain AND 1 or 2 stars in comparability domain AND 2 or 3 stars in outcome/exposure domain. **Poor quality:** 0 or 1 star in selection domain OR 0 stars in comparability domain OR 0 or 1 stars in outcome/exposure domain.

# Table S6: References for table 2 Diagnostic tests in the work-up of suspected GLILD.

|  |  |
| --- | --- |
| Tests | Articles |
| Radiology |  |
| Chest X-ray | (1-16) |
| CT | (1-11, 16-35) (13, 17, 36-46) |
| PET | (9, 20, 24, 25, 27, 29) (40) (47) |
| Pulmonary function tests | (2, 4-7, 9-14, 17, 18, 20-26, 28-35, 39, 42, 44, 45, 48-55) |
| Bronchoalveolar lavage |  |
| BAL- culture | (1, 2, 5-8, 10, 11, 16, 21, 22, 25, 27, 31, 32, 34, 56) |
| BAL- differential cell count | (4-6, 11, 14, 16, 25, 32, 34, 48, 49, 54-57) |
| Biopsy | (1-11, 16-35) (12, 14, 15, 45, 48-53, 57) |
| TBB | (5-7, 11, 20, 21, 24, 28, 32, 34, 40, 48, 51, 53-55, 57) |
| TBLC | (34) |
| VATS | (4, 5, 10, 16, 23, 24, 26, 29-31, 35, 37, 49, 51-53, 55) |
| Thoracotomy (open surgery) | (1-3, 7, 18, 19, 32, 52, 53) |
| Lung but not specified | (8, 12, 14, 15, 17, 19, 22, 33, 34, 39, 48, 58, 59) |
| Other site | (21, 25, 29, 34, 39, 51, 53) |
| Genetic | (13, 15, 18, 22, 29, 31, 33-35, 38, 40, 49-51, 53-55) |

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