

The Burden of Surgery for Tenosynovial Giant Cell Tumor: A Targeted Review

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OBJECTIVE

- A targeted literature review was conducted to characterize the burden of tenosynovial giant cell tumor (TGCT) in the global contemporary treatment landscape, with a focus on surgery-related burden

BACKGROUND

- TGCT is a locally aggressive neoplasm arising from the synovium of joints, bursae, and tendon sheaths¹
- Patients with TGCT experience substantial pain and stiffness, impaired physical function, and limited range of motion; if left untreated, TGCT may cause significant disability^{2,3}
- TGCT can be classified into two subtypes:
 - Localized (nodular) TGCT (L-TGCT) is identified by well-defined margins that may envelop tendons
 - Diffuse TGCT (D-TGCT) lacks distinct boundaries and can infiltrate tendon sheaths and extra-articular structures^{1,4}
- Surgery is the standard of care treatment for TGCT, although systemic therapies may be used in patients whose disease recurs or is not amenable to surgery⁴

METHODS

- Embase, MEDLINE, and selected conference proceedings were searched in August 2023
- Eligible studies were published between 2013 and 2023, in English, and included ≥ 20 patients with TGCT (≥ 40 for humanistic burden studies), and reported epidemiology, humanistic burden, treatment patterns, or economic burden
- For treatment patterns and economic outcomes, studies that collected data from 2008 and later were included
- For studies with multiple publications, those with non-identical populations were considered unique and included
- The titles and abstracts of identified records were screened against the eligibility criteria by one reviewer, with all included records and 10% of excluded records screened by a second, independent reviewer
 - The full texts of potentially relevant records were subsequently screened by the same method

RESULTS

- Of 1,171 records screened, 48 publications reporting on 36 studies were included
 - A list of all included studies is provided in [Supplementary Table 1](#), available through the ISPOR App
 - Of these, 33 of 36 reported surgery-related outcomes
- Most patients were young and female ([Figure 1](#))
- The proportion of patients who received ≥ 2 surgeries ranged from 9% of 933 patients with L-TGCT in Mastboom 2017 to 60% of 210 patients with D-TGCT in Mastboom 2018 ([Figure 2](#))
 - In Lin 2023, a large claims database study, 29% of patients who received one surgery received a second surgery over a 3-year follow-up period
- Three studies found that arthroplasty was performed more frequently for subsequent surgeries (3%–28%) compared with initial surgeries (0%–15%)
 - Li 2023 reported greater healthcare resource use for arthroplasty compared with arthroscopy due to longer operative durations, longer hospital stays, and higher complication rates requiring revisions
- Post-operative recurrence rates were up to 67% ([Figure 3](#))
- Eight studies reported surgery-related resource use, including repeated hospitalizations, imaging, specialist visits, and supplemental care, such as physical therapy
- Two studies reported healthcare costs
 - Surgery-related hospitalizations comprised $>70\%$ of total healthcare costs in TOPP 2021, a large, multinational European study of patients with D-TGCT
 - In Lin 2023, inpatient costs were \$2,654–\$5,045 per patient per year (20%–26% of total costs), although the proportion of admissions related to surgery was not reported
- Assessments for humanistic burden outcomes were heterogeneous across studies included in the review ([Figure 4](#))
- Patients frequently reported pain and stiffness, and TGCT negatively impacted health state utility values (HSUV), health-related quality of life (HRQoL), and physical function
 - These assessment scores improved after surgery in most studies, but were worse for patients who received multiple surgeries compared with those who received one surgery

Figure 1. Patient characteristics

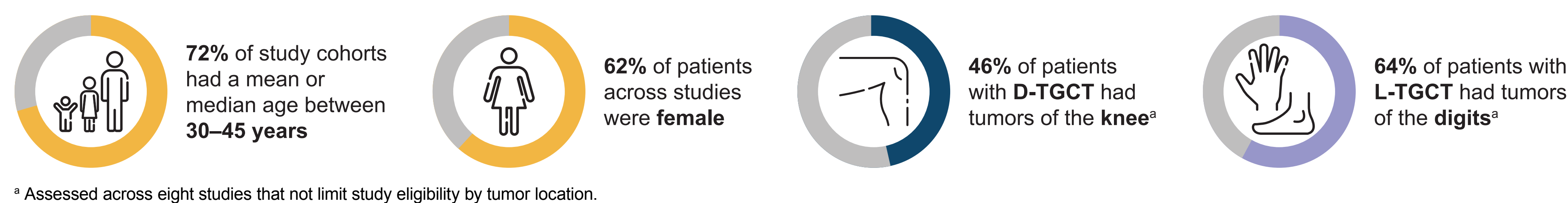
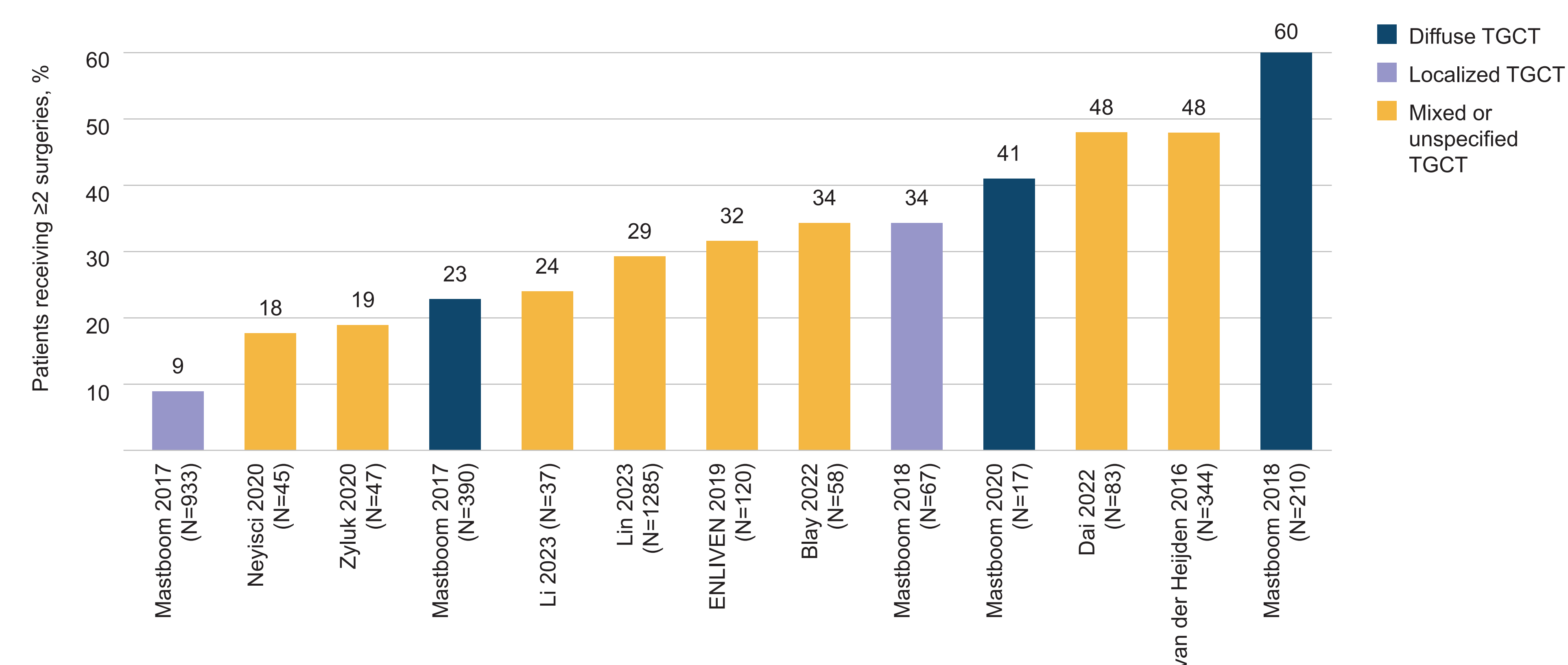
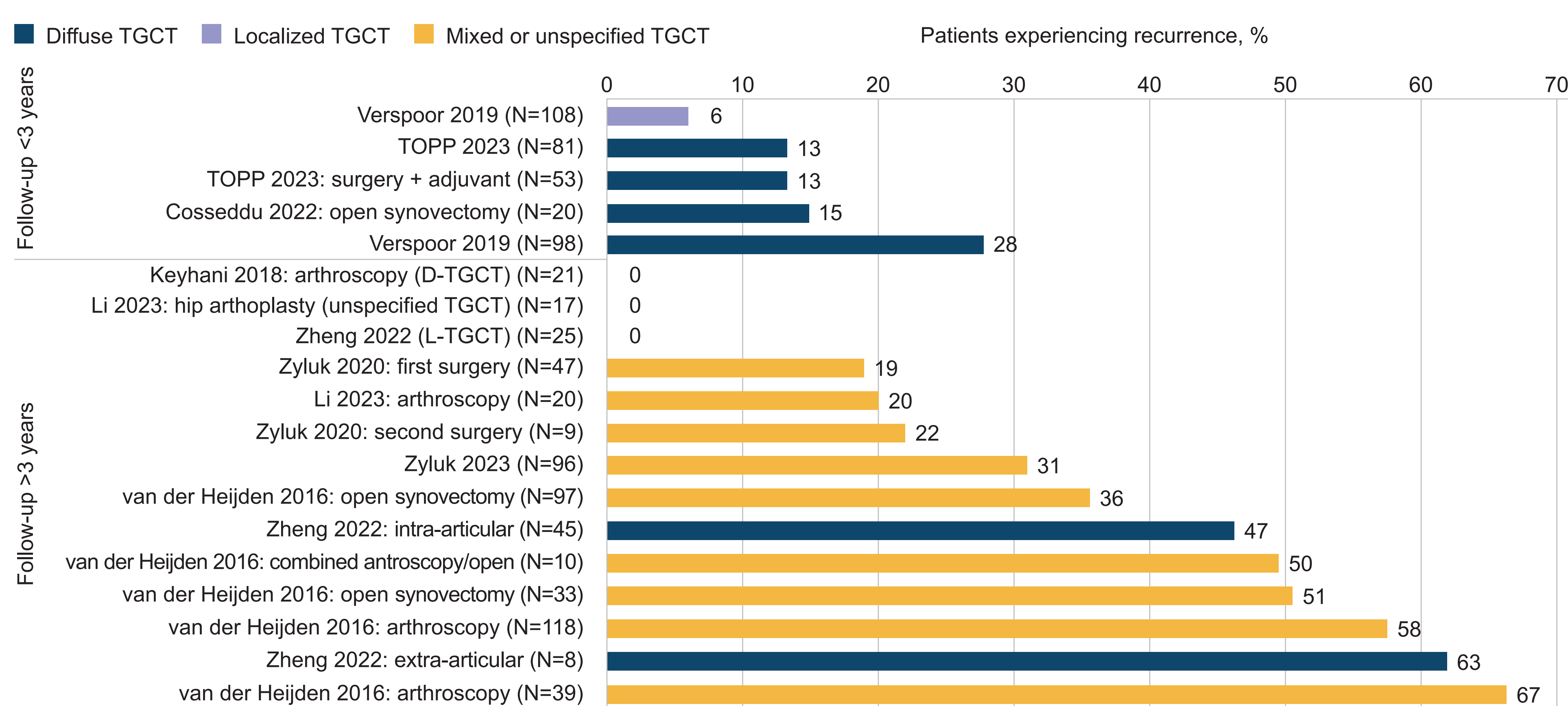


Figure 2. Proportion of patients who received ≥ 2 surgeries



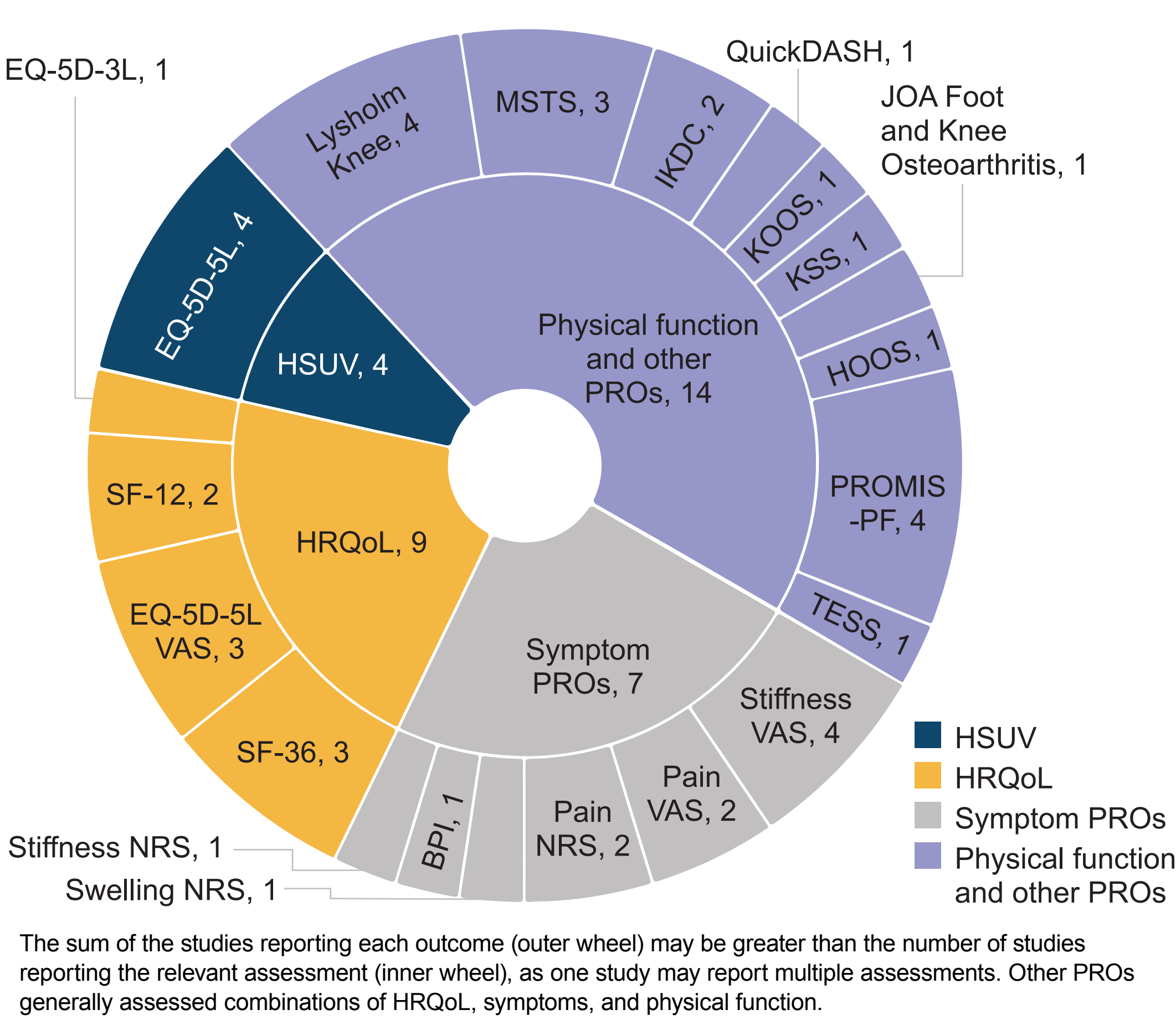
Assessment timeframe was only reported for two studies (Neyisci 2020, 5 years; Zyluk 2020, mean follow-up 4 years). All other data were reported for unspecified assessment timeframes either prior to study enrollment or during study follow-up.

Figure 3. Post-operative recurrence rates



Only studies that reported follow-up durations are included in this figure. Various surgery types were considered by the studies unless otherwise specified.

Figure 4. Number of studies reporting each humanistic burden outcome category and assessment



CONCLUSIONS

- Reported post-operative recurrence rates varied but were over 50% in multiple studies
 - Recurrence rates may have been underestimated due to the short follow-up durations and single center designs of many included studies, as patients who experience recurrence may seek subsequent surgeries at different institutions
- TGCT symptoms negatively impact HRQoL and physical functioning, which may be exacerbated by multiple surgeries
 - Understanding the humanistic impact of both the disease itself and surgical treatment is complicated by heterogeneity in assessment methodologies
- Surgical treatment is associated with high costs and healthcare resource use, although the full economic burden of TGCT, especially the impact on employment, has been understudied to date
- The risk/benefit profile of surgery, particularly repeat surgeries, should be considered alongside other non-invasive treatment options

Abbreviations

BPI, Brief Pain Inventory; DASH, Disabilities of the Arm, Shoulder and Hand; D-TGCT, diffuse TGCT; HOOS, Hip Disability and Osteoarthritis Outcome Score; HRQoL, health-related quality of life; HSUV, health state utility values; IKDC, International Knee Documentation Committee; JOA, Japanese Orthopaedic Association; KOOS, Knee Injury and Osteoarthritis Outcome Score; KSS, Knee Society Score; L-TGCT, localized (nodular) TGCT; MSTS, Musculoskeletal Tumor Society; NRS, numeric rating scale; PRO, patient-reported outcome; PROMIS-PF, PROMIS Measurement Information System - Physical Function; SF, Short Form; TESS, Toronto Extremity Salvage Score; TGCT, tenosynovial giant cell tumor; VAS, visual analog scale.

References

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Disclosures

BH and NZ report employment with Deciphera Pharmaceuticals, LLC. ML and EK were employees of Costello Medical, Inc. at the time this study was conducted, which received fees from Deciphera Pharmaceuticals, LLC for conducting this work.



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