Preliminary results on the international multicenter retrospective Tenosynovial Giant Cell Tumour Database

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Introduction

Tenosynovial Giant Cell Tumour (TGCT), previously named Pigmented Villonodular Synovitis (PVNS), is a rare, locally aggressive neoplasm. The lesion can either present as a single nodule (nodular-type), or as multiple nodules (diffuse-type) along a synovial layer or tendon sheath. Current literature primarily consists of several relatively small cohort studies containing generally inhomogeneous data. A multicenter-pooled database of individual patient data is therefore essential in order to evaluate current treatment protocols and their clinical results, as well as risk factors for progressive disease and local recurrence. Our goal is to set up a retrospective multicenter cohort with histologically proven TGCT, treated between 1990 and 2014 with a minimum follow-up of two years.

Methods

(Un)published data of individual patients from five tertiary orthopedic oncology centers are the base of this international multicenter database. 407 (239 female, median age at operation 34.7 years) cases with TGCT are included: 190 of 276 affected knee-joints are diffuse-type, 86% of these primarily treated with open-resection; 86 nodular-type, 85% primarily treated with arthroscopic-resection. 131 other joints are affected of which 84(64%) are diffuse-type. TGCT of fingers and toes are excluded.

The median follow-up time overall is 6.39 (95%CI 5.19-7.59) years. To assess the effect of risk factors on first recurrence, a multivariate cox-regression model with risk factors: gender (male/female), age at first operation (years), surgical treatment (arthroscopic/open), affected joint (knee/other) and TGCT-type (diffuse/nodular) is estimated. The results are reported as hazard ratios (HR) and their corresponding 95% confidence intervals (95%CI).

Local recurrence free survival at 2 and 5 years is calculated from the time of first surgical resection to first recurrence.

Results

Total number of first recurrence is 157(39%): located about the knee, diffuse-type 103(54%) and nodular-type 13(15%); other affected joints, diffuse-type 30(35%) and nodular 7(15%). Mean time to local recurrence is 11.68 (95%CI 9.57-13.80) years.

The HR for gender (0=male), age at operation, surgical treatment (0=open-resection), joint (0=knee) and TGCT-type (0=diffuse) is 1.43 (95%CI 1.04-1.96), 0.99 (95%CI 0.98-1.00), 1.07 (95%CI 0.83-1.37), 1.39 (95%CI 0.96-2.00) and 2.40 (95%CI 1.64-3.52) respectively. Recurrences occurred significantly more frequent in male patients (p=0.018) and in diffuse-type (p=0.0001).

The median time to recurrence: in male is 5.16 (95%CI 3.10-7.20) years and in female 16.05 (95%CI 4.82-27.36) years (p=0.017); after arthroscopy 5.00 (95%CI 2.36-7.64) years and after open-resection 10.56 (95%CI 7.52-13.53) years (p=0.089).

Local recurrence free survival at 2 and 5 years is 0.74 (95%CI 0.69-0.80) and 0.59 (95%CI 0.54-0.65) respectively. At final follow-up 343 patients (84%) show no evidence of disease (49 alive with disease, 5 death of other disease, 10 lost).

Conclusion

Preliminary results on the international multicenter TGCT database show a high risk of first local recurrence, especially with diffuse-type. Nodular-types recur less, but still remarkably often. Interestingly recurrences are diagnosed significant earlier in men. To prolong time to recurrence, open resection is advocated, especially in young patients with TGCT about the knee and in diffuse or recurrent cases.

Further investigation of risk factors for recurrence of TGCT is essential for proper treatment planning in an era of new systemic and (neo)adjuvant treatment possibilities. In order to get more reliable information about this orphan disease and expose possible risk factors for local recurrence, multidisciplinary but foremost international multicenter collaboration is of utmost importance.

Keywords : Tenosynovial Giant Cell Tumour (TGCT), Pigmented Villonodular Synovitis (PVNS),
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