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Burden of Illness Associated with Tenosynovial Giant Cell Tumors

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ABSTRACT

Purpose: Little is known about the burden of illness in patients with tenosynovial giant cell tumors (TGCT), which are rare, typically benign, lesions of the synovial tissue including giant cell tumor of the tendon sheath (GCT-TS) and pigmented villonodular synovitis (PVNS). The objective of this study was to describe health care resource use and costs for patients with GCT-TS and PVNS, which are rare and typically benign TGCT.

Methods: A retrospective cohort study design was used to analyze administrative claims for adult commercial and Medicare Advantage health plan enrollees with evidence of GCT-TS and PVNS from January 1, 2006 through March 31, 2015. Participants were continuously enrolled for 12 months before (pre-index period) and 12 months after (post-index period) the date of the first tenosynovial giant cell tumor (TGCT) claim (index date). Preindex and postindex measures were compared using the McNemar test and Wilcoxon signed-rank test. Results were stratified by TGCT type.

Findings: The study identified 4664 patients with TGCT, 284 with GCT-TS, and 4380 with PVNS. Mean age (GCT-TS group: 50 years; PVNS group: 51 years) and sex distributions (GCT-TS group: 60.2% female; PVNS group: 59.5% female) were similar for each group. Most patients with GCT-TS (78.2%) had at least one postindex surgery, compared with 38.7% of patients with PVNS. Mean total health care costs increased from $8943 in the preindex period to $14,880 in the postindex period ($P < 0.001$) for GCT-TS and from $13,221 in the preindex period to $17,728 in the postindex period ($P < 0.001$) for PVNS. Preindex to postindex ambulatory costs increased nearly 120% for patients with GCT-TS ($4340 to $9570, $P < 0.001$) and 50% for patients with PVNS ($6782 to $10,278, $P < 0.001$), and physical therapy use increased significantly during the same period (GCT-TS: 18% to 40%, $P < 0.001$; PVNS: 38% to 60%, $P < 0.001$).

Implications: Costs increased substantially 1 year after the first TGCT claim, with more than half the costs covering ambulatory care. These results suggest a high health care burden once TGCT is identified. (*Clin Ther*. 2018;▮:▮▮▮–▮▮▮) © 2018 Elsevier HS Journals, Inc. All rights reserved.

Key words: tenosynovial giant cell tumors, giant cell tumor of the tendon sheath, pigmented villonodular synovitis, retrospective claims study, healthcare costs.

INTRODUCTION

Tenosynovial giant cell tumors (TGCTs) comprise a group of rare, usually benign but locally aggressive, soft tissue lesions characterized by inflammation of the synovial lining among joints and tendons. These neoplasms are thought to arise via overexpression of colony-stimulating factor 1 receptor.[1] The giant cell tumor of the tendon sheath (GCT-TS) and pigmented villonodular synovitis (PVNS) represent the localized and diffuse types of tumors, respectively. The localized type is most often found in hands and feet, whereas diffuse lesions include diffuse-type giant cell tumor

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and PVNS, which affect lower limb joints[2] and tend to be more aggressive.[3]

TGCTs are rare, with an annual US incidence of approximately 1.8 cases per million for PVNS and 9.2 cases per million for GCT-TS reported in 1980. A recent study in the Netherlands estimated worldwide annual incidence rates of 29, 10, and 4 per million for TGCTs localized to digits, localized to extremities, and diffuse presentation, respectively.[4,5] TGCTs are rarely lethal, but they can be disabling, affecting a patient's quality of life via tumor-related limitations and surgical sequelae. The current standard of care for localized tumor is complete surgical resection of the tumor, aimed at reducing symptoms, improving function, and minimizing risk of recurrence.[6] However, surgical excision or amputation is commonly required in diffuse tumor cases.[7] When the tumor infiltrates surrounding soft tissues and because relapse rates are high,[8,9] radiotherapy has been used.[10] However, no approved drug treatment targeting the colony-stimulating factor 1 receptor is currently available, although treatment with tyrosine kinase inhibitors has been studied.[11,12]

As a rare type of tumor, TGCTs are likely underreported and underdiagnosed, and there is a paucity of literature describing patterns of care and burden of illness among affected patients. The purpose of this study was to advance the understanding of the patient characteristics, treatment patterns, and health care resource use and costs associated with TGCTs. These data will clarify the context in which varying treatment options may be pursued and developed for these patients, thus better informing researchers, clinicians, and payers.

METHODS

Study Design and Data Source

This was a retrospective cohort analysis using administrative claims from the Optum Research Database, which contains data regarding health care enrollment, and medical service use and costs covering the period from 1993 to the present. For 2014, the available data related to approximately 12.3 million individuals with commercial medical and pharmacy benefit coverage and 2.1 million with Medicare Advantage plus Prescription Drug coverage, representing a geographically diverse sample of the US population. Claims for pharmacy services are typically submitted electronically by the pharmacy at the time prescriptions are filled. Medical claims are collected from all available health care sites for virtually all types of provided services, coded to conform with insurance industry standards. This study was conducted in accordance with the principles of the Declaration of Helsinki. Institutional review board approval was not required because no identifiable protected health information was extracted or accessed.

Study Sample

The study sample included adult (18 years or older) commercial and Medicare Advantage plus Prescription Drug health plan enrollees with evidence of TGCT from January 1, 2006, through March 31, 2015 (identification period). Individuals were required to have ≥2 nondiagnostic or 1 nondiagnostic and 1 diagnostic claim at least 30 days apart for (1) GCT-TS (*International Classification of Diseases, Ninth Revision, Clinical Modification* [ICD-9-CM] code 727.02) or (2) PVNS (ICD-9-CM code 719.2x). The index date was defined as the date of the first claim for either GCT-TS or PVNS during the identification period. Individuals were also required to be continuously enrolled in the health plan with both medical and pharmacy benefits for 12 months before (preindex period) and 12 months after (postindex period) the index date. Individuals were excluded if they had claims for a TGCT or a malignant tumor (ICD-9-CM codes 140.xx-209.36, 209.70-209.79) in the primary position during the preindex period. Participants were assigned to the GCT-TS cohort or PVNS cohort, depending on their index TGCT claim. Individuals with both GCT-TS and PVNS codes on the index date were excluded from the analysis.

Measures

From enrollment data, demographic characteristics of the participants were obtained, including age at the index date, sex, insurance type, and US geographic region. On the basis of ICD-9-CM diagnosis codes, a preindex Quan-Charlson comorbidity score was calculated, and the top 5 most prevalent comorbid conditions were identified by the Clinical Classifications Software managed by the Agency for Health care Research and Quality (AHRQ).[13,14] In addition, the following preindex clinical information was obtained from medical claims: PVNS tumor location