Dear Sir,

Referred or secondary otalgia is the complaint of ear pain arising from pathology of non-otologic locations with which the ear shares common neural pathways. The pain can present in various degrees of intensity and characteristics while severity is not proportional to the seriousness of the underlying cause\textsuperscript{1,2}. It is a relatively common encounter in everyday practice while the aetiologies related to the extensive and well-documented aural nerve supply are numerous and well-recognised\textsuperscript{2}. As a result several attempts have been made at addressing a structured evidenced-based approach to the management of the complaint but none have applied the actual frequency of causes to their respective protocols\textsuperscript{1,3}. The Ten Ts of referred otalgia as an aide memoire when addressing a patient with secondary otalgia has been proposed previously\textsuperscript{4}.

The frequency of causes in referrals to a general secondary ENT department remains contentious where several authors report that 50\% of referred otalgia has a dental origin whereas two studies have demonstrated that the majority are due to musculoskeletal sources\textsuperscript{1-2,5-8}.

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Our objective was to review the accuracy of the 10 Ts axiom of referred otalgia as described by applying each category to the recognised causes of non-otogenic ear pain using a literature review\textsuperscript{4}. Our subsequent aims were to present our experience in the frequency of diagnosis of the primary sources of referred otalgia in a cohort of adult patients, to shape the results in order to provide a loose but guided approach in the management of this condition and to apply our findings to an evaluation of the usefulness of this longstanding but under-utilised aide memoire.

Materials and methods

Discretionary ethical approval from the Chairman of the local Ethics committee at the University Hospital Limerick was obtained.

A documenting of known causes of referred otalgia was performed using a literature review of the search terms ear pain, otalgia and referred using online search engines Medline and Google scholar, The identified causes of referred otalgia from the literature were assessed and subsequently refined to fit loosely into convenient groupings each commencing with the letter T.

A retrospective assessment of a consecutive series of data collected prospectively on all adult patients 18 years or more with otalgia as a primary symptom that presented to the routine out-patients service of the senior author (JEF) during a 10-year period from 2003 to 2012. Patients without an active disease process in the ear and who were subsequently followed up until a definitive or presumptive diagnosis was made were included in the study. The ultimate diagnosis was allocated to the relevant category of T group.

Results

A review of the literature produced an extensive list of causes and these were collated into loose anatomical categories beginning with the letter T. Thirteen convenient groupings (Table 1) were produced, 9 of which were part of the original ten Ts and 4 more categories revealed by our study. One of the subsets described by Harvey was attributed to the tympanic membrane but was omitted as it did not fit
the defined diagnosis\textsuperscript{4}. A degree of latitude was required for the cervical cohort including muscular or cervical spine causes and it was decided to apply the term Torticollis or 'Tender Neck' to this grouping.

A total of 226 patients, comprising 64 males and 162 females were included in the study. The age range (in years) for males was 26 to 84 with a mean age of 38.4 years, while the age range for females was 18 to 94 with a mean age of 47.6 years. The population ratio of males to females presenting with otalgia to our service was 2:5. Females presented earlier than males and younger patients presented earlier and had more severe symptoms. Mild to moderate pain score at 4-6 in severity related more to otalgia of cervicogenic origin while severe 8-9/10 tended to be from temporomandibular joint disorders. Among patients presenting with severe otalgia (n=35), the males were younger than females (mean age 39.7 years versus 47.1 years). Record of duration of otalgia was available in 125 cases revealing an interstice ranging from 1 to 72 months with a mean duration of 10.3 months.

The majority of referred otalgia in adults (Table 2) was musculoskeletal from jaw and neck (66%), out of which 51% were attributable to cervical muscles or spine and 15% from the temporomandibular joint. Specific diagnoses are presented in Table 3 although 10 cases (4.4%) in the throat and thyroid groups remained undiagnosed. More than one aetiological condition was reported in 33 (14.6%) patients. Throat pathology accounted for 17% of causes while dental causes were identified in 5.75%. Thirteen cases (5.75%) were found to be due to underlying sinister pathologies: these included 8 cases of laryngopharyngeal SCC, 4 cases of tonsil/tongue base malignancy and an oesophageal carcinoma was found to be the cause of otalgia in one patient presenting with associated dysphagia.

Discussion
The wide-spread shared innervation can potentially transmit pain to the ear from structures as distant as the thorax\textsuperscript{3}. It is one of the few
diagnoses in our area that encompasses the entire spectrum of our specialty and involves many allied disciplines. The approach to the assessment of patients with this condition is therefore quite a broad canvas demanding a structured application that is relatively easy to remember and perform.

Synopsis of key/new findings;
The study produced a list of causes presented in Table 1 which is comprehensive but not exhaustive and the relatively flexible categorisation should allow for the inclusion of new or omitted conditions. Harvey introduced the concept involving the 10Ts of referred otalgia, however the tympanic membrane was included as one category, but otalgia from tympanic membrane pathology should be classified as primary rather than referred. Furthermore some authors have defined referred otalgia as the presence of ear pain with normal otoscopy but it quite obviously can occur in patients with abnormal but quiescent changes. In addition to excluding tympanic membrane in this study, we have described 4 more sources of referred otalgia, each of which we have assigned nomenclatures beginning with ‘T’ to retain a similar scaffold to the relevant aide memoire, resulting in the evolution of 13Ts. Our experience indicates that the majority of adult referred otalgia in such a setting will be caused by mechanical disorders of neck and jaw (66%), the teeth consisting only 5.7% and importantly 5% will arise from sinister pathologies. A further misconception identified by our survey is the number of authors that have based their assertions that dental causes are the most common aetiology of secondary pain on reviews that either quote other reviews, cite an abstract or report what ultimately appears to be an opinion.

Strengths of the study;
All patients attended to and followed up by senior author (JEF)

Comparisons with other studies;
In accordance with Jaber et al and Al-Sheikhli, this study reveals that most cases of non-otogenic otalgia that are referred to routine secondary otolaryngology centres are musculoskeletal and not of
primary dental origin unlike the majority of published reports\textsuperscript{1-2,5-8}. Al-Sheikhli's findings in his 1980 study that 80% of cases are due to three conditions including neck, jaw joint and teeth mirror our results except that pharyngeal causes have replaced dental aetiology which may reflect changes in referral patterns and improvements in dental services\textsuperscript{8}. The age profile of our musculoskeletal group was lower than Jaber et al but the breakdown of that particular cohort in our study was muscular rather than degenerative disease of the cervical spine \textsuperscript{7}.

Clinical applicability of the study;
The neck and jaw including a frequently subtle tender point at the upper insertion of sternocleidomastoid into the mastoid process should be assessed after otoscopy reaching a probable diagnosis clinically in almost 70% of cases. The importance of history taking and complete clinical examination including cranial nerves and nasopharyngoscopy in addition to the advocated use of digital tonsil and tongue base palpation cannot be over-emphasised. Associated features on history-taking are paramount and the red flag combination of throat symptoms and ear pain in a smoker should be foremost in any clinician's mind. It has been shown that 5\% of malignant skull base tumours diagnosed at a tertiary referral neuro-otologic centre presented with referred otalgia\textsuperscript{9}. Our rate of approximately 5\% of patients with upper aero-digestive malignancy reflects this incidence of mitotic disease in a cohort of patients with secondary otalgia and all of our patients had associated symptoms or signs. Ear pain as an isolated presenting symptom is unusual in pharyngo-laryngeal cancer but can be the only complaint in the less frequent cases of skull base or temporal bone malignancy\textsuperscript{9-10}. Special attention is crucial in high risk patients mindful of the younger non-smoking patient with potential HPV-related tumours, particularly where persistent ear pain is associated with any throat symptoms\textsuperscript{10}. Investigations should be targeted at any of the 13 Ts dependent on the suspected diagnosis but also including those with idiopathic or un-responding persistent pain. Admittedly this is a relatively small uncontrolled study but it does provide guidance for further research into this common complaint.
Keypoints

“Thirteen Ts” is a useful, memorable and straightforward aide-memoire in the approach to referred otalgia. Two-thirds of secondary otalgia in adults presenting to a regional or secondary centre is of musculo-skeletal origin, either neck or TMJ. If Pharyngeal causes (Throat Tonsil Tube) are included more than 85% of conditions will be identified. The definition should not include normal otoscopy. Dental causes excluding temporomandibular abnormalities are less than 6% of cases.

References
10. Tikka T, Pracy P, Paleri V. Refining the head and neck cancer...

Table 1  Aetiology of referred otalgia presented as 13 Ts.

1. Torticollis/Tender neck (muscles & Cervical spine)
   Arthritis, muscle spasms, trauma, cervical radiculopathy, lymphadenitis, neck abscesses, carotidynia

2. Temporomandibular Joint (TMJ)
   TMJ dysfunction, Syndrome, Arthritis, malocclusion, dentures, bruxism

3. Thyroid (& Salivary)
   Thyroiditis- Hashimoto, De Quirvan’s, thyroid cyst haemorrhage, sialadenitis, salivary tumours

4. Teeth
   Dental abscesses, caries, pulpitis, impacted tooth

5. Tongue
   Glossitis, carcinomas, trauma

6. Tonsil (oropharynx)
   Tonsillitis, tonsiloliths, peritonsillar abscess, malignancy, Eagle's Syndrome, Glossopharyngeal neuralgia

7. Throat (hypopharynx/larynx)
   Neoplasms, Retropharyngeal or parapharyngeal abscess, laryngitis, vocal cord tension dysphonia

8. Trigeminal
   Trigeminal neuralgia, Mid-facial Segment Pain

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9. Turbinates
Nasal conditions: Sinusitis, rhinosinusitis, neoplasms, Sphenopalatine (Sluder's) Neuralgia, rhinogenic contact point otalgia

10. Tube (nasopharynx)
Nasopharyngeal malignancy, Nasopharyngitis.

11. Temporal bone
Myositis, Temporal arteritis, petrous apicitis, Tympanic or Intermediate nerve neuralgia, CPA/Temporal bone tumours (primary & metastatic)

12. Trachea
Tracheitis, tracheobronchitis, chondritis, neoplasms,

13. Thorax
Oesophagitis/Gastroesophageal reflux, carcinomas, angina/myocardial infarct, aneurysmal disease of aorta and innominate
Table 2  Frequency of Diagnosis in 226 patients with referred otalgia applied to 13 Ts

(> 1 condition counted for each category see Table 3)

<table>
<thead>
<tr>
<th>T</th>
<th>No.</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Tender neck / Torticollis</td>
<td>116</td>
<td>51%</td>
</tr>
<tr>
<td>Throat</td>
<td>38</td>
<td>17%</td>
</tr>
<tr>
<td>TMJ dysfunction</td>
<td>34</td>
<td>15%</td>
</tr>
<tr>
<td>Teeth</td>
<td>13</td>
<td>5.75%</td>
</tr>
<tr>
<td>Tonsil</td>
<td>7</td>
<td>3%</td>
</tr>
<tr>
<td>Thyroid</td>
<td>7</td>
<td>3%</td>
</tr>
<tr>
<td>Turbinate</td>
<td>6</td>
<td>2.6%</td>
</tr>
<tr>
<td>Tongue</td>
<td>4</td>
<td>1.76%</td>
</tr>
<tr>
<td>Tube</td>
<td>2</td>
<td>1.3%</td>
</tr>
<tr>
<td>Temporal</td>
<td>1</td>
<td>0.4%</td>
</tr>
<tr>
<td>Trigeminal Neuralgia</td>
<td>1</td>
<td>0.4%</td>
</tr>
<tr>
<td>Thorax</td>
<td>1</td>
<td>0.4%</td>
</tr>
<tr>
<td>Trachea</td>
<td>0</td>
<td>0%</td>
</tr>
</tbody>
</table>
Specific Diagnoses related to 13 ts

**Torticollis/Tender Neck**
- SCM/Muscle Neck Spasm 86
- Combined Muscle Spasm + TMJ Arthritis 13
- C-Spine Arthritis 12
- Muscle Spasm + C-spine and TMJ Arthritis 2
- Tender Neck + Toothache 1
- Laryngopharyngeal reflux (LPR) + neck spasm 1
- Neck muscle and tonsillitis 1

**Throat**
- Pharyngitis nonspecific 10
- LPR 6
  - Laryngopharyngeal SCC 5
  - LPR + Neck Muscle Spasm 4
  - Supraglottic SCC 2
  - LPR + TMJ Dysfunction 1
  - Tonsillitis + Neck muscle 1
  - Pyriform Sinus SCC 1
  - Vocal Cord Tension Dysphonia 1
  - Other unspecified throat pain 7

**Temporomandibular Joint (TMJ)**
- TMJ Arthritis/Dysfunction 19
- TMJ Arthritis + Neck Spasm 13
- TMJ Arthritis + Dental 2
- TMJ + LPR 1

**Thyroid (& Salivary)**
- Autoimmune 1
- Left Thyroid Nodule 1
- Thyroid unspecified tenderness 3
Submandibular Sialadenitis
Parotid Tumours

**Teeth**
Dental Caries/ Toothache
Teeth + TMJ Dysfunction
Teeth + Neck Pain

**Tonsil**
Tonsillitis/Tonsil stones
Tonsillitis + neck spasm

**Turbinates**
Septal Spurs contact point
Chronic Rhinosinusitis/nasal polyps

**Tongue/Tongue base**
Tongue base SCC

**Tube**
Nasopharyngitis

**Temporal**
Temporalis Fibromyalgia/myositis

**Thorax**
Oesophageal SCC

**Trigeminal Neuralgia**

**Trachea**