I. Stylohyoid chain ossification: A discussion of etiology

A. J. Camarda, DDS, MSc,* C. Deschamps, MSc, DMD,** and D. Forest, DSD, MSF, Montréal and Dorval, Québec, Canada

UNIVERSITÉ DE MONTRÉAL

Not all patients with cervicopharyngeal pain related to the stylohyoid apparatus can be diagnosed as having Eagle’s syndrome. An attempt is therefore made to classify these patients according to etiology. First, a diagnosis of Eagle’s syndrome is applied to symptomatic patients in whom elongated, ossified styloid processes develop within a period of time posttraumatically. Second, a diagnosis of stylohyoid syndrome is applied to patients in whom elongated styloid processes and/or stylohyoid chain ossification develop early in life as an anatomic anomaly, and in whom symptoms later develop. Last, a diagnosis of pseudostylohyoid syndrome is applied to patients in whom, because of aging, a tendinosis at the junction of the stylohyoid ligament and the lesser horn of the hyoid bone develops. It is hoped that such a simplified classification may lead to a global understanding of the causes of such symptomatology, a more practical approach to treatment, and less confusion among the professions in regard to terminology.

(ORAL SURG ORAL MED ORAL PATHOL 1989;67:508-14)

As a result of the documented association between ossification of the stylohyoid apparatus and cervicopharyngeal pain, a number of syndromes have been described: Eagle’s syndrome,1-6 styloid syndrome,5-7 stylohyoid syndrome,8 and styloid-stylohyoid syndrome.9 Although Eagle’s syndrome persists as the preferred clinical diagnosis, there remains much confusion as to the differences between these syndromes, necessitating their explanation and a simplification of such a classification. Moreover, the mere presence of cervicopharyngeal pain clinically and stylohyoid chain ossification radiographically does not lead to, in all cases, a diagnosis of Eagle’s syndrome2 for the following reasons.

First, a certain number of patients with ossified stylohyoid chains are young and asymptomatic.10 Second, there does not appear to be a correlation between the severity of pain and the extent of the stylohyoid chain ossification in older, symptomatic patients.10-13 Third, the majority of symptomatic patients have had no recent history of tonsillectomy or other cervicopharyngeal trauma.8,10,13 We must therefore develop an alternative diagnosis.

REVIEW OF THE LITERATURE

Gossman and Tarsitano1 have divided the literature concerning stylohyoid chain ossification into three distinct historical periods. The first period evolved during the seventeenth century,1 at which time anatomists recognized anomalies of these structures postmortem, but were obviously not involved with their clinical presentation and, therefore, these anomalies were considered variants of normal rather than pathogenic.

The second period took place during the early twentieth century with the advent of diagnostic radiology, which could detail these “abnormalities” antemortem. These occurred in patients who had neck radiographs taken for complaints such as dysphagia, otalgia, cephalgia, TMJ pain, and pain during mastication.

At this time, W.W. Eagle,1 an otorhinolaryngologist, reported several cases of cervicopharyngeal symptoms associated with a radiographic diagnosis of an elongated, ossified styloid process occurring a few months posttonsillectomy. Eagle postulated that such a traumatic event occurring in the pharyngeal region could stimulate the styloid process, resulting in its overgrowth and subsequent pharyngeal symptoms. This became known as Eagle’s syndrome.
The third and final period occurred during the mid-twentieth century with the advent of panoramic radiology, which allowed practicing dental surgeons to retrospectively question patients presenting with such radiographic evidence of stylohyoid anomalies. Therefore, although Eagle's syndrome has become the globally accepted diagnosis for such cases, it was now possible to show that such trauma was not always responsible for ossification of the stylohyoid apparatus.

Following such panoramic interpretation, Stafne and Hollinshead concluded that, in most cases of true stylohyoid chain ossification, such ossification often occurred in persons younger than 31 years of age. Moreover, it had also been estimated that between 2% and 4% of the general population presents radiographic evidence of an ossified portion of the stylohyoid chain. Studies have also shown the following interesting points. First, the majority of these 2% to 4% of patients are asymptomatic. Second, that when symptoms do exist, there is little correlation between the extent of the symptoms and the severity of the ossification. Third, those few patients who are symptomatic are usually over 40 years of age. Fourth, the majority of symptomatic patients have had no recent history of tonsillectomy or other cervicopharyngeal trauma.

Of recent added interest has been the increase in the number of patients with reported non-MPDS (myofascial pain dysfunction syndrome) who manifest symptoms of the classic Eagle's syndrome in the absence of both previous neck trauma and radiographic evidence of ossified stylohyoid chain segments. Steinman suggests that some of these patients could have a tendinosis at the junction of the stylohyoid ligament and the lesser horn of the hyoid bone as a result of aging.

From the above results, one may conclude that patients who manifest similar, localized symptoms fall into different diagnostic groups based on differing etiologic factors. This becomes extremely important when one attempts to treat these patients in the most reversible yet appropriate modality.

THEORIES OF OSSIFICATION

In man, the ceratohyal element of the second branchial arch degenerates in time. However, its fibrous sheath persists as the stylohyoid ligament, which contains cartilaginous and osseous potential. Since the stylohyoid process usually ossifies 5 to 8 years after birth, variation in ossification and fusion of the four elements of the second branchial arch (under the appropriate stimulation) can lead to a marked variation in radiographic appearance of the whole stylohyoid chain at a young age. At this point, we must state that the term calcification—albeit popular—is erroneous, since histologically there is a hyperplasia of the styloid process or a metaplasia of the stylohyoid fibrocartilaginous tissue into osseous tissue. For this reason, the term ossification is presently applied.

Steinman has proposed three theories that could explain such ossification. The “Theory of Reactive Hyperplasia” proposes that if the styloid process is appropriately stimulated, as in pharyngeal trauma, ossification would occur at the terminal end of the process, and at the expense of the stylohyoid ligament (Fig. 1). This could occur during the posttraumatic healing period and could lead to symptoms in
Fig. 2. Note the elongated styloid process and the ossified stylohyoid apparatus on the right. There seems to be an articulation or joint along this stylohyoid chain.

Fig. 3. A, Note the fully ossified stylohyoid apparatus bilaterally in a 22-year-old woman with no history of cervicopharyngeal trauma. This patient’s report of dysphagia subjectively improved with reassurance only. B, Lateral neck view of patient in Fig. 3. A. Note the right ossified stylohyoid chain as it articulates with the hyoid bone (arrowhead).

The area. This elongated process would then impinge on nearby structures (primarily the carotid arterial system) and thereby give rise to symptoms of arterial occlusion with lateral head movement.

The second theory, the “Theory of Reactive Metaplasia,” also involves a traumatic stimulus that would induce certain ligamentous sections of the stylohyoid apparatus to undergo metaplastic changes and thereby become intermittently ossified (Figs. 2 and 3). This metaplasia would be feasible due to the presence of osseous centers within these fibrous jointed sections. If stimulated, such osseous centers, within the ligamentous portions in question, could induce these segments to become osseous links, thereby joining the stylohyoid apparatus into a stiff, inelastic cord to the styloid process. This event could also occur during the posttraumatic healing period and lead to developing symptomatology. However, in this case, the symptoms would occur in the region of the preexisting stylohyoid ligament, which is now
ossified. Such ossification would impinge upon nearby pharyngeal soft tissues and give rise to strictly dysfunctional symptomatology, as in pain during deglutition. These two theories could indeed explain the marked ossification of the stylohyoid apparatus secondary to traumatic events, such as tonsillectomy in any age group.

The third theory, called the “Theory of Anatomic Variance,” involves the stylohyoid process and/or the stylohyoid ligament as ossified structures that develop in the early, formative years after birth (Fig. 4). This theory could apply to those cases in which there are early radiographic findings of such ossification in children and young adolescents in the absence of antecedent cervicopharyngeal trauma (as an inductive stimulus).

DISCUSSION

Origin of symptoms

The three aforementioned theories of developmental ossification are all certainly plausible. However, cervicopharyngeal symptomatology seems to be the exception rather than the rule in such cases.8, 10

With this in mind, and having reviewed the recent literature, it would further seem that of the recently documented cases of Eagle's syndrome, fewer still can be actually ascribed to this syndrome.8, 10, 16, 17, 21, 22 W.W. Eagle originally intended this syndrome to be applied to patients who exhibit such radiographic and clinical findings within a few months after tonsillectomy. Thus one must avoid applying this syndrome as a diagnosis based solely on the radiographic findings of such ossification, especially in the absence of a traumatic etiology. This is crucial when one considers the classic, irreversible treatment protocol for Eagle's syndrome, which primarily involves surgical excision.

There is no doubt that a true Eagle's syndrome due to either the Theory of Reactive Hyperplasia or the Theory of Reactive Metaplasia presents a definitive diagnosis, the treatment of which is surgical excision of the ossified segment(s), if one is to definitely treat the patient. However, if symptoms occur as a result of a longstanding anatomic variant of the stylohyoid chain (i.e., elongation or ossification) in the absence of a traumatic stimulus, as described by the Theory of Anatomic Variance, then this cannot be diagnosed as a true Eagle's syndrome. We postulate that a diagnosis of "stylohyoid syndrome" be applied, since the styloid process may be elongated, and/or any or all of the stylohyoid apparatus may be ossified. Treatment in such cases would involve an initial conservative phase followed, if needed, by the surgical excision of the ossification.

A third and final category of patients is occasionally encountered. These patients manifest symptoms identical to those of the stylohyoid syndrome group, but in the absence of radiographic evidence of an ossified stylohyoid chain. To explain the origin of these symptoms, we postulate a “Theory of Aging Developmental Anomaly.” That is to say, with aging there is an increased inelastic property of soft tissues, whereby a tendinosis would tend to develop at the junction of the stylohyoid ligament and the lesser horn of the hyoid bone, secondary to the increased ligament resistance to joint movement (between ligament and bone), in certain older patients. Such reactive inflammation could lead to localized symptoms mimicking a stylohyoid syndrome. In order to avoid confusion in the diagnosis and treatment of
Table I.

<table>
<thead>
<tr>
<th>Syndrome</th>
<th>Age group</th>
<th>Differentiating features</th>
</tr>
</thead>
<tbody>
<tr>
<td>Eagle's syndrome</td>
<td>Any</td>
<td>1. Recent history of neck trauma (e.g., tonsillectomy)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>2. Radiographic evidence of elongated styloid process or stylohyoid chain ossification</td>
</tr>
<tr>
<td></td>
<td></td>
<td>3. Clinical palpation of such elongation or ossification</td>
</tr>
<tr>
<td></td>
<td></td>
<td>4. No pretraumatic clinical and/or radiographic evidence of styloid process elongation</td>
</tr>
<tr>
<td></td>
<td></td>
<td>and/or stylohyoid chain ossification</td>
</tr>
<tr>
<td>Stylohyoid syndrome</td>
<td>Any, but usually over 40 years of age</td>
<td>1. No history of previous neck trauma</td>
</tr>
<tr>
<td></td>
<td></td>
<td>2. Radiographic evidence of stylohyoid chain ossification (in part or whole) at a young age</td>
</tr>
<tr>
<td></td>
<td></td>
<td>3. Possible clinical palpation of such ossification</td>
</tr>
<tr>
<td></td>
<td></td>
<td>4. Theory of Developmental Anomaly</td>
</tr>
<tr>
<td>Pseudostylohyoid syndrome</td>
<td>40 years of age and older</td>
<td>1. No history of previous trauma</td>
</tr>
<tr>
<td></td>
<td></td>
<td>2. No radiographic evidence of stylohyoid chain ossification (in part or whole)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>3. No clinical palpation of such ossification</td>
</tr>
<tr>
<td></td>
<td></td>
<td>4. Theory of Aging Development Anomaly</td>
</tr>
</tbody>
</table>

such patients, we suggest that they be diagnosed as “pseudostylohyoid syndrome” patients.

Having presented these three diagnoses, we must now address the fact that the majority of all symptomatic patients with completely or partially ossified stylohyoid chains are 40 years of age and older. We have presented two possible explanations. One, the Theory of Aging Developmental Anomaly supposes that with aging a tenodnosis develops at the junction of the stylohyoid ligament and the hyoid bone. Symptoms occur in the absence of radiographically demonstrable ligament ossification. This remains a possibility, but, as stated previously, these patients constitute only a small fraction of those who present for treatment. As a result, the pseudostylohyoid syndrome is less often encountered in this age group. Moreover, a majority of these symptomatic patients have radiographic evidence of stylohyoid ossification, but with no history of antecedent pharyngeal trauma. Therefore, some other reason must have been the cause of such ossification. We believe that the origin is probably related to early developmental anomalies of the stylohyoid chain, as proposed by our second theory, the Theory of Developmental Anomaly.

We propose that although such ossification appears early in life, symptoms rarely occur at this age, because of the “adaptability” and elasticity of the regional soft tissues. With age, however, regional ligaments and soft tissues become less elastic and offer more resistance to surrounding hard tissue structures; this results in pain and/or discomfort with movement (as in deglutition, for example). Such symptomatic ossification without evidence of previous cervicopharyngeal trauma (such as tonsillectomy) would lead to a diagnosis of stylohyoid syndrome. This could possibly explain how a significant number of asymptomatic young patients with anatomically ossified stylohyoid chains—and with no history of previous gross cervicopharyngeal trauma—could exhibit worsening clinical symptoms after many years of increasing ossification. Such patients would thus form the majority of these symptomatic older patients and thereby negate the often used diagnosis of Eagle's syndrome.

**Treatment of symptoms**

The normal styloid process is too short to be palpable. However, in cases of an abnormal stylohyoid apparatus, palpation of the tonsillar fossa should reveal an elongated styloid process and/or an ossified stylohyoid chain. Historically, therefore, treatment in cases of true stylohyoid chain ossification caused by trauma (Eagle's syndrome) is immediate surgical excision. The first such recorded surgical procedure is credited to Weinlechner in 1872. To date, the most reliable long-term treatment in such cases involves the intraoral approach, with complete surgical excision of the styloid process and/or ossified stylohyoid ligament. It is imperative to remove the whole process as closely as possible to the cranial base in order to avoid renewed ossification distally. The extraoral lateral neck approach is only indicated when the intraoral approach is not possible—as in limited jaw opening.

However, in light of our hypothesis that most symptomatic patients suffer from stylohyoid syndrome rather than from Eagle's or pseudostylohyoid syndromes, we propose that the initial treatment be...
less irreversible. In order to lend support to this proposal, we should mention that some patients who refuse surgical intervention initially improve! Can such patients possibly adapt to their condition? As Gossman states, "..."(ultimately) as in the TMJ patient, it is not the physical findings but the ability by the patient to tolerate or accept these findings that constitutes (in part) the morbid condition.\textsuperscript{10} We propose, therefore, that treatment of such symptomatic patients be divided into two phases. The first involves reassurance and local injection of longacting local and steroidal solutions regionally to confirm the diagnosis and to offer relief. In the pseudostylohyoid syndrome, this is all that is necessary. In the patients with stylohyoid syndrome, who are refractive to such initial treatments and who cannot obviously adapt to their developing symptoms with time, would then be required to undergo a surgical intervention. In the patients with proven Eagle's syndrome, however, surgical treatment is the initial treatment of choice because of the severity of the rapidly occurring ossification and symptomatology, to which the patient has no time to ever really adapt. Therefore, an accurate medical history is essential in establishment of the correct etiology and diagnosis, and in initiation of the most appropriate yet conservative treatment modality.

SUMMARY

A detailed review of the literature has been presented in regard to the association of developing cervicopharyngeal pain with stylohyoid ossification. Four interesting findings are presented. First, the majority of symptomatic patients with a true stylohyoid chain ossification are 40 years of age and older. Second, very few of these patients give detailed histories of tonsillectomy or other recent neck trauma. Third, the majority of asymptomatic patients with radiographically-confirmed stylohyoid chain ossification are young. Fourth, some of these older symptomatic patients may tolerate their symptoms well, in spite of not having had surgery performed for removal of the ossification(s).

The evidence presented seems to indicate that anatomically abnormal and ossified stylohyoid chains develop in a significant number of young, healthy individuals during their early formative years, and that many years may pass before symptoms occur. These symptoms of dysphagia and/or cervicopharyngeal pain occur because of the loss of elasticity of surrounding soft tissue structures, rendering these same tissues less adaptable to pressures caused by the slowly-worsening abnormally-ossified stylohyoid chain. According to our proposed Theory of Anatomic Anomaly, it is believed that a number of these young asymptomatic patients go on to acquire such symptoms with age. Therefore, a diagnosis of stylohyoid syndrome would be appropriate.

Therefore, an indiscriminate diagnosis of Eagle's syndrome in the absence of an antecedent history of trauma is inappropriate. That is to say, a diagnosis of Eagle's syndrome is applied to such symptomatic patients only when: they have no radiographic evidence of such ossification pretraumatically; such ossification develops within a few months after trauma, with accompanying symptoms; or they demonstrate such ossification radiographically and clinically (on palpation) posttraumatically, due to either the Theory of Reactive Hyperplasia or Reactive Metaplasia. It may be summarized from the review of literature, however, that very few of the majority of symptomatic patients actually demonstrate findings (including positive palpation) that lead to a diagnosis of Eagle's syndrome.

We have also presented evidence showing that a less significant number of older patients demonstrate symptoms identical to patients with stylohyoid syndrome, but who have no radiographic or clinical (on palpation) findings or evidence of such stylohyoid ossification. These patients would probably have a tendinosis at the junction the stylohyoid ligament and the lesser horn of the hyoid bone, and would be diagnosed as having a pseudostylohyoid syndrome.

It is hoped that a simpler, more accurate diagnostic scheme of such cervicopharyngeal symptoms and a better understanding of the differing etiology of such patient reports would lead to a more appropriate treatment, based on accurate determination of causative factors. The global application of the term "Eagle's syndrome" to patients in the older age groups who manifest radiographic evidence of stylohyoid chain ossification should be avoided.

We are presently undertaking a radiographic evaluation of a significant number of pedodontic patients in an attempt to discover the percentage of children or young adolescents who actually demonstrate asymptomatic, ossified stylohyoid chains as a developmental anomaly. If such is the case in a statistically significant number of patients, and in the absence of previous cervicopharyngeal trauma, then we believe that a statistically significant number of patients from this group possibly go on to acquire symptoms over a number of years and, therefore, a diagnosis of stylohyoid syndrome is made. We believe, in fact, that it is this diagnostic syndrome that greatly predominates over the other two in frequency of occurrence. Our findings will be published in Part II of this series.
REFERENCES


Reprint requests to:
Dr. A. J. Camarda
Université de Montréal, Faculté de Médecine dentaire
Département de Stomatologie, C.P. 6128, "SUCC A"
Montréal, Québec, Canada H3C 3J7

May 1989