REVIEW

Eagle's Syndrome: A Review of the Literature

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Eagle's syndrome represents symptoms brought on by compression of regional structures by elongation of the styloid process or ossification of the stylohyoid or stylomandibular ligaments. Watt Eagle described it for the first time in 1937, dividing it into two subtypes: the ''classic syndrome'' and the ''stylo-carotid artery syndrome.'' Many theories have been put forth regarding its pathogenesis. Depending on the underlying pathogenetic mechanism and the anatomical structures compressed or irritated by the styloid process, symptoms vary greatly, ranging from cervicofacial pain to cerebral ischemia. The syndrome generally follows tonsillectomy or trauma. Diagnosis is confirmed by radiological findings. Palpation of the styloid process in the tonsillar fossa and infiltration with anesthesia are also used in making the diagnosis. The treatment is primarily surgical; however, some conservative treatments have also been used. The current literature on Eagle's syndrome is reviewed, highlighting its often underestimated frequency and its clinical importance. Clin. Anat. 22:545–558, 2009. © 2009 Wiley-Liss, Inc.

Key words: ossification; ligament; temporal bone; syndrome

INTRODUCTION

The terms ''Eagle's syndrome'' or ''Stylohyoid syndrome'' describe a series of symptoms caused by an elongated styloid process and/or the mineralization (ossification or calcification) of part or the entire stylohyoid ligament (Winkler et al., 1981; Catelani and Cudia, 1989; Babad, 1995; Feldman, 2003).

Anatomists have been observing elongated styloid processes since antiquity. Dr Watt Eagle estimated the normal length of the styloid process to be 2.5 cm (Eagle, 1949). Further research proved it to vary from 1.52 to 6 cm (average length from 2 to 3.2 cm) (Yetiser et al., 1997; Gozil et al., 2001) and Yavuz et al. (2008) determined the length of the styloid process as 3.5 to 8 cm (mean length 5 cm on the right side and 5.2 cm on the left side) in a Turkish patient group. The variation in size found by Lang (1983) was: 0.1–0.9 cm. in 18 cases; 1–1.9 cm in 40 cases; 2.0–2.9 cm in 19 cases; 3.0–3.9 cm in 14 cases; and 4.0–4.2 cm in 1 case (Bergman et al., 1988).

Generally, if the length of the styloid process is more than 3 cm, it is considered to be elongated (see Fig. 1) (Palesy et al., 2000). A short styloid process may be completely hidden by the vaginal process (see Fig. 2) and an elongated styloid process may reach the hyoid bone (Chandler, 1977).

Such an anatomical variation was first reported by Pietro Marchetti of Padua in 1652 who described the ossification of the stylohyoid ligament (Shamrani, 1991). Demanchetis described a calcified stylohyoid ligament in 1852 (Rechtweg and Wax, 1998). The related painful syndrome was first described in 1870 by Lücke and in 1872, in Vienna. Weinlechner surgically excised the styloid process in order to treat symptoms (Moffat et al., 1977; Balbuena et al., 1997). Several sporadic reports followed, however, it was Eagle who provided a comprehensive description of the syndrome for the first time in 1937 (Eagle,

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Fig. 1. Elongated styloid process on the left side of a male skull (3.9 cm). [Color figure can be viewed in the online issue, which is available at www.interscience.wiley.com.]

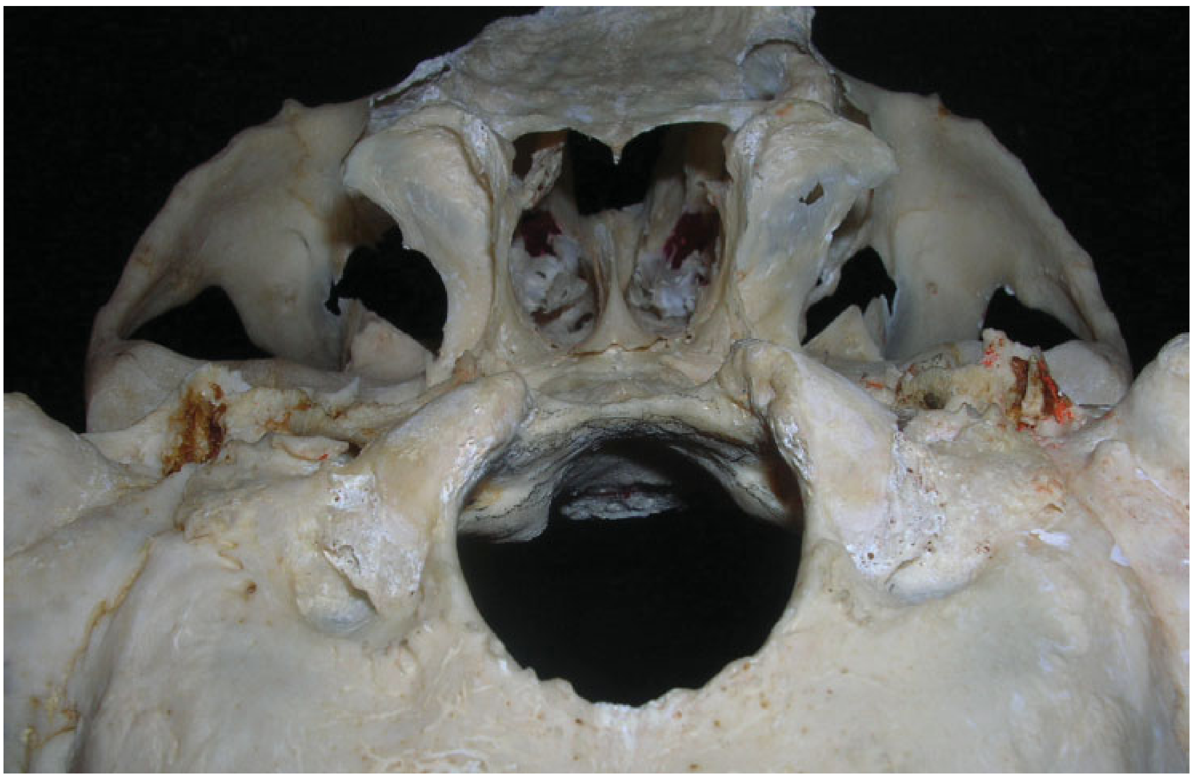

Fig. 2. Short styloid process (bilaterally) may be completely hidden by the vaginal process. [Color figure can be viewed in the online issue, which is available at www.interscience.wiley.com.]

TABLE 1. The Frequency of the Elongation or Calcification of the Styloid Process According to Different Researchers

Date	Authors	Type of cases	Number of cases	Frequency (%)
1958	Eagle	Clinical	>200	4
1970	Kaufman	Clinical	484	28
1970	Barclay and Donaldson	Panoramic radiographs	100	1
1977	Gossman and Tarsitano	Panoramic radiographs	4,200	1,4
1979	Correll et al.	Panoramic radiographs	1,771	18,2
1984	O'Carroll	Clinical and radiological	479	78,5
1986	Keur et al.	Clinical and radiological	1,135	31
1986	Benazzi et al.	Radiological	1,000	31
1986	Monsour and Young	Panoramic radiographs	670	21,1
1988	Lengele and Dhem	Skulls	246	29
1990	Ferrario et al.	Panoramic radiographs	286	84,4
1991	Rath et al.	Skulls	232	0,4
1997	Bozkir et al.	Clinical	200	4
2005	Rizzati-Barbosa et al.	Panoramic radiographs	2,252	20
2005	Ilguy et al.	Panoramic radiographs	860	3,7

1937) and identified two forms of the syndrome (Eagle, 1948, 1949).

Eagle described the symptoms of the classic stylohyoid syndrome as persistent unilateral pharyngeal pain, aggravated by swallowing and frequently reverberating in the ear (Dolan et al., 1984; Bafaqeeh, 2000; Sandev and Sokler, 2000). Eagle attributed these symptoms to the development of scar tissue around the tip of the styloid process shortly after tonsillectomy.

The vascular form of the syndrome was the ''stylo-carotid artery syndrome'' (Eagle, 1949), which is not related to tonsillectomy (Buttura da Prato et al., 2004) and is attributed to impingement of the internal carotid artery, extracranially by the styloid process (Mendelsohn et al., 2006; Chuang et al., 2007). This can cause a compression when turning the head or in dissection of the carotid artery resulting in a transient ischemic accident or stroke (Infante-Cossio et al., 2004; Farhat et al., 2009). Ruwanpura et al. (2008) found after the autopsy examination of a 39-year-old female, that the elongation of the styloid processes (in the form of fibrous membranes) compressed both carotid sinuses.

EPIDEMIOLOGICAL FACTS

The incidence of Eagle's syndrome is very controversial (Rogers and Chang, 2007). Eagle estimated the incidence of an elongated styloid process in the general population to be 4%, of which only 4% display symptoms (Fritz, 1940; Eagle, 1949, 1958, 1962). In another clinical study, Kaufman et al. (1970) reported elongated styloid processes in 28% of their patients, while Correll et al. (1979) examined the largest number of radiographs (1,771 panoramic views) and estimated the incidence to be 18.2%, 93% of which exhibited bilateral elongation. Only a relatively small number of patients (8 out of 1,771) however, exhibited symptoms related to Eagle's syndrome, most of which were unilateral (Correll et al., 1979) (Table 1). Although the researchers found different frequencies of elongation or calcification of the styloid process, most of them agreed that only a small number of cases with elongation present with symptoms. Styloid process elongation, in most cases, appears bilaterally. However, in most of the symptomatic cases, the symptoms are unilateral, even in cases with bilateral elongation. Bilateral elongation is believed to occur frequently, although bilateral symptoms are quite rare (see Fig. 3) (Harma, 1967; Jan, 1989).

Styloid process elongation also appears to be slightly more common in women (Woolery, 1990; Liu et al., 2005). Some studies have found that women with such elongation may be more symptomatic than men (Strauss et al., 1985; Keur et al., 1986; Liu et al., 2005; Yavuz et al., 2008) and some have revealed a greater incidence in elderly women, likely due to menopause (Rizzatti-Barbosa et al., 2003, 2005).

There is little correlation between severity of symptoms and the severity of the ossification. Even though ossification occurs in individuals under the age of 31, very few become symptomatic (1–5%) (Gossman and Tarsitano, 1977; Van der Westhuijzen et al., 1999). Patients exhibiting symptoms are typically over the age of 40 (Balasubramanian, 1964; Manson-Hing, 1969; Barclay and Donaldson, 1970; Kaufman et al., 1970; Gossman and Tarsitano, 1977; Van der Westhuijzen et al., 1999).

Elongated styloid processes occur more frequently in older patients and result in the deposition of calcium salts into the ligaments and processes (Unlu et al., 2008). Styloid process elongation might be a predictor of peak bone mass (Sisman et al., in press). Ectopic calcification might have a role in the elongation of the styloid process. Abnormal calcium, phosphorus, and vitamin D metabolism is very common in patients with end-stage renal disease, and this calcification due to such levels is commonly associated with this disorder (Gokce et al., 2008). There are no data on the prevalence of an elongated styloid process in children (Holloway et al., 1991; Quereshy et al., 2001). The incidence of an enlarged styloid process has been found to be

Fig. 3. Posterior view of the elongated process bilaterally (4 cm). [Color figure can be viewed in the online issue, which is available at www.interscience.wiley.com.]

greater in rural Indian populations (see Fig. 4) (Thot et al., 2000; Yadav et al., 2001). This may possibly be explained dietary habits (Rath and Anand, 1991).

Direction and angulation of the elongated process may be responsible for the irritation of a number of anatomical structures traversing the parapharyngeal space (Strauss et al., 1985). *Lateral deviation* may cause its tip to impinge upon the external carotid artery, at its bifurcation into the maxillary and superficial temporal arteries. *Posterior deviation* may trap the last four cranial nerves, internal carotid artery and the internal jugular vein between the elongated styloid and the transverse process of the atlas (Ghosh and Dubey, 1999). *Medial deviation* may impinge the tonsillar fossa (Ghosh and Dubey, 1999). *Anterior angulation* may result in mucosal irritation and pressure over vital structures in the tonsillar fossa (Yavuz et al., 2008). A history of trauma or tonsillectomy is uncommon in these patients (Eagle, 1958; Messer and Abramson, 1975; Gossman and Tarsitano, 1977).

ANATOMY AND EMBRYOLOGY

The styloid process is usually a slender, elongated, cylindrical bony projection of the temporal bone, located between the internal and external carotid arteries and the internal jugular vein and anteromedial to the stylomastoid foramen (Perez Carro and Nunez, 1995). Although typically straight, it is occasionally curved (Das et al., 2008). Three muscles (the styloglossus, stylopharyngeus and stylohyoid) are attached to the styloid process extending to the tongue, pharynx, and hyoid bone, respectively (see Fig. 5) (Camarda et al., 1989). The styloglossus and the stylohyoid muscles are innervated by the hypoglossal and facial nerves, respectively. The stylopharyngeus muscle is innervated by the glossopharyngeal nerve (Palesy et al., 2000).

Two ligaments attach to the styloid process: the stylohyoid and stylomandibular ligaments. The stylohyoid ligament is attached to the tip of the styloid process and extends to the lesser cornu of the hyoid bone (see Fig. 6) (Hollinshead, 1969; Du Brul, 1980; Balbuena et al., 1997). Stylohyoid and stylomandibular ligaments help regulate the movements of the mandible, the hyoid bone, the tongue and the pharynx (Shimada and Gasser, 1988).

Many important anatomic structures are closely located to the styloid process and the stylohyoid ligament. Lateral to the styloid process one finds the facial and hypoglossal nerves, the occipital artery and the posterior belly of the digastric muscle. The facial nerve emerges from the stylomastoid foramen posteriorly and passes laterally through the parotid gland (Fig. 7). Medial to the styloid process (from posterior to anterior) the internal jugular vein (with the accessory, hypoglossal, vagus and glossopharyngeal nerves) and the internal carotid artery are seen (Fig. 8). The superior constrictor muscle and the pharyngobasilar fascia are situated *medially to the tip of the styloid process* and adjacent to the tonsillar fossa (Yavuz et al., 2008).

A precise understanding of the mechanism of styloid process elongation requires knowledge of its embryogenesis (Ghosh and Dubey, 1999). Embryologically, the styloid process, stylohyoid ligament, lesser cornu of the hyoid bone and the superior portion of the hyoid body are derived from Reichert's cartilage, which arises from the second pharyngeal arch.

Reichert's cartilage is divided into four distinct segments (Lorman and Biggs, 1983). The proximal tympano-hyal part appears before birth and fuses with the petrous temporal bone to form the tympanic portion and stapes. The lower part of the styloid process and the upper part of the stylohyoid ligament stem from the stylo-hyal part, which appears after birth. These do not unite with the tympanic portion until after puberty and in some skulls never unite. The ossification of the stylo-hyal part and its fusion with the tympano-hyal part results in a long styloid process, while failed ossification results in a shortened styloid process. The cerato-hyal part usually deteriorates with its fibrous sheath forming the stylohyoid ligament and the hypo-hyal part forming the lesser cornu

Fig. 4. Enlarged styloid process on the right side of a male skull. [Color figure can be viewed in the online issue, which is available at www.interscience.wiley.com.]

of the hyoid bone (Dwight, 1907; De Chazal, 1946; Stafne, 1969; Hamilton et al., 1972; Gray, 1977; Kiely et al., 1995; Ghosh and Dubey, 1999; Satyapal and Kalideen, 2000; Chuang et al., 2007).

The styloid process begins to ossify at the end of pregnancy (Lavine et al., 1968) and continues to undergo calcification over the first 8 years of life. The ossification time and degree of ossification vary

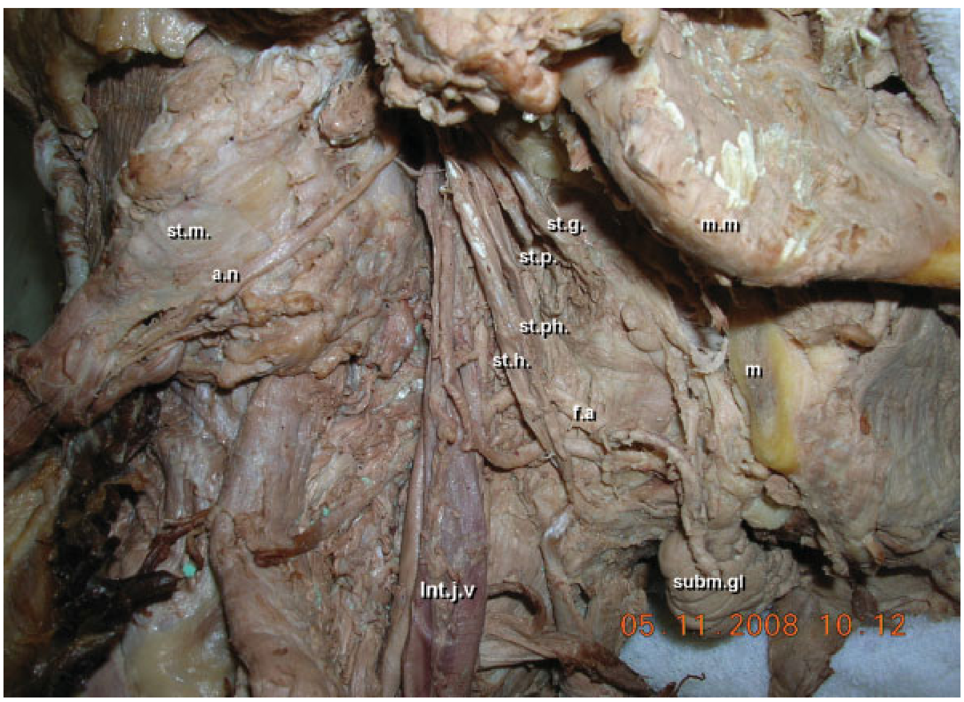

Fig. 5. Deep dissection of the right side of a female cadaver. st.m., sternocleidomastoid muscle; a.n, accessory nerve; st.g., styloglossus muscle; st.p., styloid process; st.ph., stylopharyngeus muscle; f.a., facial artery; int.j.v., internal jugular vein; subm.gl, submandibular gland; m, mandible; m.m, masseter muscle. [Color figure can be viewed in the online issue, which is available at www.interscience.wiley.com.]

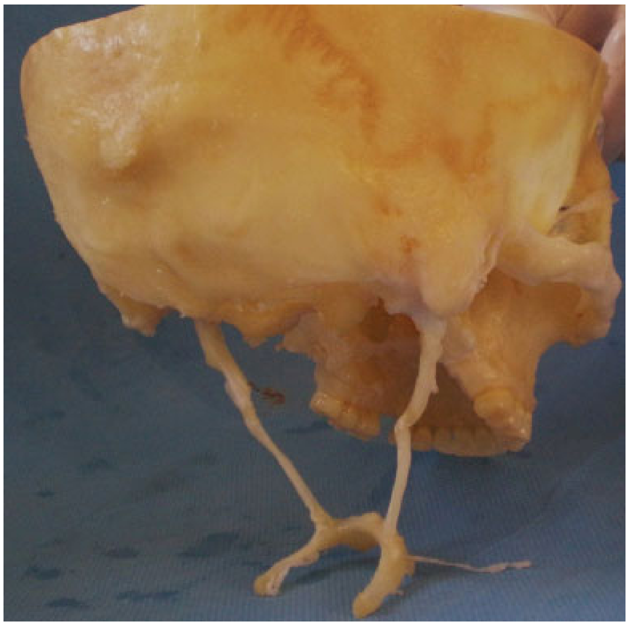

Fig. 6. Special preparation of the human hyoid apparatus. The mandible was removed. [Color figure can be viewed in the online issue, which is available at www.interscience.wiley.com.]

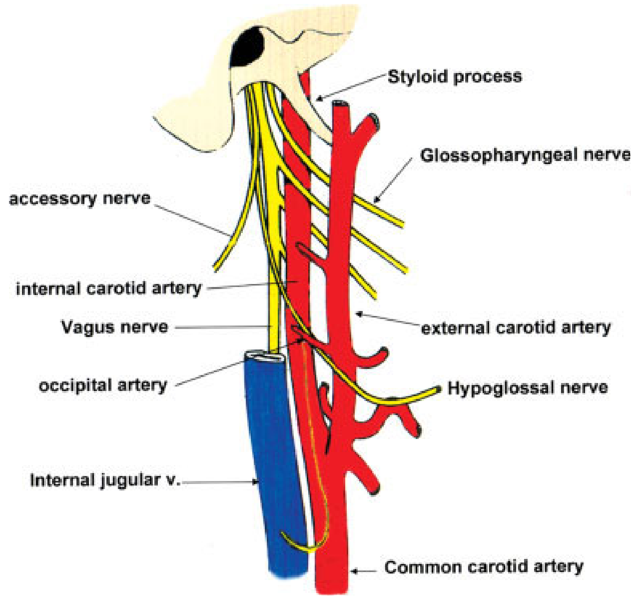

Fig. 8. Styloid process anatomy. From posterior to anterior are located the internal jugular vein (with the accessory, hypoglossal, vagus, and glossopharyngeal nerves) and the internal carotid artery. [Color figure can be viewed in the online issue, which is available at www.interscience.wiley.com.]

greatly (Stafne and Hollinshead, 1962; Stafne, 1969).

ETIOLOGY

A number of prevalent etiological and causal theories correlates anatomy, embryology, and physiology in order to derive a clearer understanding of Eagle's Syndrome (Baugh and Stocks, 1993; Sela et al., 2003) but the exact cause is still considered a mystery (Massey and Massey, 1979; Quereshy et al., 2001; Dinkar and Amonkar, 2003; Sela et al., 2003).

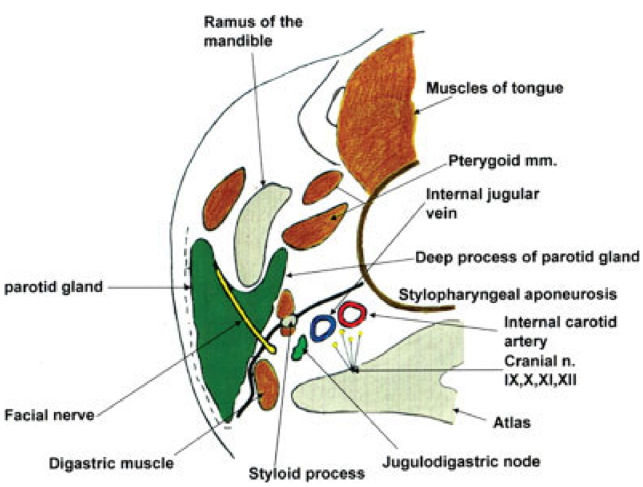

Fig. 7. Deep parotid space anatomy. The facial nerve passes laterally through the parotid gland. [Color figure can be viewed in the online issue, which is available at www.interscience.wiley.com.]

Steinmann (1968, 1970) proposed three different theories to explain such abnormal ossification. The ''Theory of Reactive Hyperplasia'' suggests that if the styloid process is appropriately stimulated by pharyngeal trauma, ossification may continue from its tip against the stylohyoid ligament. This may occur during the post-traumatic healing period and cause symptoms related mainly to impingement of the carotid artery (Camarda et al., 1989).

The ''Theory of Reactive Metaplasia'' (Eteromorphosis) also includes a traumatic stimulus causing multiple metaplastic changes to the stylohyoid ligament and resulting in its partial ossification. The occurrence of metaplasia might be attributable to the presence of osseous centers within these fibrous formations. In this case, symptoms would originate from the region of the stylohyoid ligament and affect the soft tissues of the neck causing increased symptoms such as dysphagia. These two theories explain the remarkable ossification of the stylohyoid apparatus following a traumatic event like tonsillectomy in any age group.

The ''Theory of Anatomic Variance,'' suggests that the styloid process and the stylohyoid ligament are normally ossified and that the elongation of the process is simply an anatomical variation. This theory explains the early radiological findings of such ossification in children and young adults who have not undergone any prior cervicopharyngeal trauma. In order to explain similar symptoms in adults without any prior cervicopharyngeal trauma and radiological

TABLE 2. Types of Elongation, Patterns of Calcification and Angulation of the Styloid Process

Type of elongation (radiographic appearance)	Pattern of calcification	Angulation of the styloid process
Type I = elongated (most frequent)	A = calcified outline (most frequent)	Narrow (<65°)
Type II = pseudoarticulated	B = partially calcified	Normal (65–75°)
Type III = segmented	C = nodular	Wide (>75°)
	D = completely calcified	

findings, Camarda et al. (1989) proposed the Theory of Aging Developmental Anomaly. According to this theory, the soft cervical tissues lose their elastic properties with age. As a result, the increased resistance to the movement of the joint between the stylohyoid ligament and the lesser cornu of the hyoid bone can cause secondary tendonitis in elderly people. In this case, the ossification procedure is complete at an early age; however the loss of the elasticity in the surrounding tissues causes symptoms much later. As this is not true elongation of the styloid process, the term ''pseudostylohyoid syndrome'' is proposed to diagnose older patients with no history of trauma in whom, because of aging, tendonitis develops at the junction of the stylohyoid ligament and the lesser cornu of the hyoid (Van der Westhuijzen et al., 1999; Bafaqeeh, 2000).

Other theories have also been proposed. According to the genetic hypothesis, this anatomic variation might be genetically transmitted as a recessive autosomal trait (Lentini, 1975). The dysendocrine theory suggests that abnormal ossification is the result of an endocrinological dysfunction in postmenopausal women (Epifanio, 1962). Another theory suggested a degenerative process, followed by ossification, may occur in patients with rheumatoid disease (Nelson, 1940).

Depending on the morphology and the calcification of the styloid process, it can be described by different types (Lavine et al., 1968; Langlais et al., 1986; Monsour and Young, 1986; Kiely et al., 1995; Guo et al., 1997; Satyapal and Kalideen, 2000). The most common type of elongation is a Type I process usually partially or completely calcified (Ilgüy et al., 2005) (Table 2).

PATHOGENESIS AND SYMPTOMS

Symptoms depend on a variety of factors, including the length and width of the styloid process, the angle and direction of its deviation and the degree of ossification (Ilgüy et al., 2005). The pathogenesis of the syndrome was described by Eagle, who discussed types (Eagle, 1937, 1949, 1958).

The first type, ''classic Eagle's syndrome,'' typically occurs in patients after tonsillectomy, although it can also occur after any other type of pharyngeal surgery. A palatable mass may be observed in the tonsillar fossa, its palpation sometimes exacerbating the patient's symptoms. Symptoms include ear pain, neck pain extending to the oral cavity and the maxilla (Le Toux, 1991; Siqueira et al., 2004; Weidenbecher et al., 2006), dysphonia (Blatchford and Coulthard, 1989; Sundmaker,1989), dysphagia (Kehrl and Hartwein, 1990; Manganaro and Nylander, 1998; Weidenbecher et al., 2006; Unlu et al., 2008), odynophagia (Tiago et al., 2002; Andrade et al., 2008), persistent sore throat (Takada et al., 2003), the sensation of a foreign body in the pharynx (Lindeman, 1985), painful trismus <25 mm, vertigo and tinnitus (Riley, 1996; Diamond et al., 2001; Ferreira de Albuquerque et al., 2003; Beder et al., 2005; Yavuz et al., 2008). Pain is also observed when turning the head or extending the tongue (Woolery, 1990). Apart from turning the head, yawning can also trigger symptoms, particularly those resembling migraine (Jacome, 2001, 2004). Other symptoms may include tongue pain in general, a sensation of increased salivation (Van der Westhuijzen et al., 1999), alterations in taste (Baddour et al., 1978; Kay et al., 2001), vocal changes (Schmidt, 1951; Harma, 1967), pain in the upper limbs (Prasad et al., 2002), chest (Shenoi, 1972), and temporomandibular joint (Ettinger and Hanson, 1975), facial paresthesia (Gonzalez Salceda, 1983), pharyngeal spasm (Stark, 1965), pain triggered by the movement of the mandible (Ozawa et al., 1995), cough, dizziness, or sinusitis. Eagle's syndrome has also been reported as the most important cause of secondary glossopharyngeal neuralgia (Soh, 1999; Slavin, 2002) or atypical craniocervical pain (Hernandez and Velasco, 2008). All of these symptoms are attributed to the irritation of cranial nerves V, VII, IX or X, all of which are situated very close to the styloid process. The observation of symptoms after tonsillectomy generates the hypothesis that these nerves are entrapped in the locally formed granular tissue. Trauma to the soft tissues during tonsillectomy may cause bone formation, leading to an elongated styloid process or ossified stylohyoid ligament. Ossification typically appears from 2 to 12 months after the trauma (Salamone et al., 2004). However, Fritz, (1940) reported that among 43 cases, only 11 had been subjected to tonsillectomy.

Isolated rheumatologic symptoms are rarely observed (Kurmann and Linthoudt, 2007).

In the stylo-carotid artery syndrome, an elongated styloid process deviating slightly from its normal direction can impinge the internal or external carotid artery, stimulating the sympathetic nerve plexus accompanying the artery and causing pain during artery's palpation. Stimulation of the internal carotid artery causes pain along the artery that is sometimes accompanied by pain in the eye and parietal cephalalgia (Jackson, 1974). These symptoms can result in wrong diagnoses, such as cluster headache or migraine. Symptoms may also include aphasia, sight disturbances, weakness (Chuang et al., 2007) or even syncope episodes (Correl and Wescott, 1982). Stimulation of the external carotid artery causes facial pain, mainly in the area under the eyes. Histological examination of the vessel wall in such cases may reveal arteriosclerosis (Koebke, 1976). Stylo-carotid artery syndrome might also results in arterial variation.

Another hypothesis has suggested that the styloid process can directly stimulate the formation of the mucosa and soft tissues of the pharyngeal region (Dwight, 1907; Eagle, 1958; Donohue, 1959).

Balasubramanian (1964) suggested that if the stylohyoid ligament is ossified and then fractured by a sudden jerk of the head, the constant movement of the hyoid bone prevents the unification of the broken parts. The subsequent formation of granular tissue compresses the soft tissues in the area and causes symptoms.

Babbit (1933) gave additional documentation of the pain associated with elongated and fractured styloid processes. Spontaneous avulsion fractures were caused by sudden laughter, coughing and epileptic seizures (Steinmann, 1970). Symptoms of the fracture may mimic tumours, foreign bodies, infections or neuralgia (Blomgren et al., 1999).

Steinmann (1968) has reported patients exhibiting symptoms of Eagle's syndrome but without any radiological finding, thus attributing it to degenerative and inflammatory processes in the area (insertion tendonitis).

Okabe et al. (2006) examined 659 panoramic radiographs of 80-year-old patients, in an effort to find other factors associated with the length of the styloid process. They found statistically significant positive relations between the length of the styloid process, serum calcium concentration and heel bone density. No relation with other factors, like arterial pressure, ECG findings, smoking and number of teeth, were identified. Bilateral ossification of the stylohyoid ligament may present as bilateral tinnitus, globus hystericus and hoarseness (Lugmayr et al., 1997).

There are three syndromes closely connected with the styloid process syndrome: Costen's, Trotter's, and Myofacial painful syndromes (Sandev and Sokler, 2000). Clinicians should consider coexisting locomotor system disorders such as Myofacial pain syndrome (MPS), Fibromyalgia syndrome (FMS), and Temporomandibular dysfunction in patients with Eagle's Syndrome (Zinnuroglu et al., 2008). The existence of an elongated styloid process in patients with classic temporomandibular disorder pain may be associated with psychological distress (Zaki et al., 1996). Many patients with stylalgia suffered primarily from a psychiatric disorder (Hampf et al., 1986). Also changes in masticating muscle activity are associated with Eagle's syndrome (Siessere et al., 2006).

Finally, Eagle's syndrome may manifest itself as pain in the molar region of the mandible, thus resembling dental pain and prompting the patient to visit their dentist (Zohar et al., 1986; Aral et al., 1997).

It is evident that Eagle's syndrome may be accompanied by a wide range of clinical manifestations, depending on the underlying pathogenetic process. Even though Eagle's syndrome is a rare condition, it should be kept in mind in patients suffering from chronic cervicofacial pain that is refractory to treatment.

DIAGNOSIS

The diagnosis of Eagle's syndrome is based on four different parameters: (1) clinical manifestations, (2) digital palpation of the process in the tonsillar fossa, (3) radiological findings and (4) lidocaine infiltration test. A patient exhibiting the symptoms associated with Eagle's syndrome, may consult their family physician or an otolaryngologist, a neurologist, a surgeon (neurosurgeon, maxillofacial or oral surgeon), a dentist or even a psychiatrist in order to be diagnosed. Persistent pain and other symptoms could aggravate the psychological state of a patient. By the time the syndrome is actually diagnosed, many patients have already visited some of these doctors, who have unsuccessfully tried to treat their symptoms (Har-El et al., 1987). This is quite understandable considering that the clinical manifestations of Eagle's syndrome resemble those of many other diseases. Consequently, it is quite difficult to make a correct diagnosis based solely on clinical manifestations. However, it is very important for physicians and dentists to include Eagle's syndrome in their differential diagnosis when treating patients experiencing pain in the cervicofacial and cervicopharyngeal regions (Bartoloni and Charlton, 2001).

The differential diagnosis includes: (Montalbetti et al., 1995)

- *Head pain:* migraine, cluster, chronic tension and cervicogenic headaches, caroticodynia, atypical facial pain, paroxysmal hemicrania (Casale et al., 2008)
- *Facial pain:* temporomandibular joint dysfunction, myofascial pain dysfunction syndrome, glossopharyngeal, trigeminal, superior laryngeal, occipital, pterygopalatine ganglion, intermediate nerve and geniculate neuralgias, clicking jaw, non-erupted or distorted third molar, faulty dental prostheses, salivary gland disease (Nishihara et al., 1986; Chi and Harkness, 1999)
- *Neck pain:* degenerative disc disease, chronic laryngopharyngeal reflux (Casale et al., 2008; Gelabert-Gonzalez and Garcia-Allut, 2008)
- *Ear nose and throat diseases:* chronic tonsillitis, tonsillar calculi, spasm of the pharyngeal constrictor muscle, otitis, mastoiditis, fracture of the hyoid bone, pterygoid hamulus bursitis
- *Other diseases:* psychosomatic diseases, foreign bodies, inflammatory and neoplastic processes in the oropharyngeal area, pharyngeal and base of tongue tumours, cervical arthritis, temporal arteritis, nuchal cellulitis and fibrositis, and neck-tongue syndrome (Eversole, 1978; Dolan et al., 1984; Haas et al., 1991; Barrett et al., 1993; Alcalde et al., 1994; Godden et al., 1999; Van der Westhuijzen et al., 1999; Gaul et al., 2006). A granular cell tumor is rarely associated with Eagle's syndrome (Philipp et al., 2001).

Variable types of styloid process-stylohyoid ligament abnormalities have been found to have significant correlation with ligamentous ossification and osteophytes of the cervical spine (Fig. 9) (Guo et al., 1997). Eagle was the first to describe the reproduc-

Fig. 9. Elongated styloid process bilaterally and ossification of the atlas. [Color figure can be viewed in the online issue, which is available at www.interscience.wiley.com.]

tion of a patient's symptoms during palpation on the tonsillar fossa as a diagnostic indicator (Eagle, 1937). However, no controlled study has proven the sensitivity or specificity of this indicator. The styloid process can only be palpated in the tonsillar fossa if it is longer than 7.5 cm (Hampf et al., 1986). A much shorter styloid process, however, can also produce symptoms (Ghosh and Dubey, 1999).

An elongated styloid process is typically confirmed radiologically using a lateral head and neck radiograph, a modified Towne's radiograph and an orthopantomograph. A disadvantage associated with the lateral radiograph is that the styloid processes of the two sides may overlap. The anterolateral modified Towne's radiograph calculates both the medial and lateral deviation of the process. In both cases, the angle of the head must be very precise throughout the entire procedure. In the orthopantomograph, the styloid process is typically considered to be elongated if its length is more than 1/3 of the length of the ramus of the mandible (Fig. 10) (Prasad et al., 2002). An advantage of the orthopantomograph is that the entire length of the styloid process is visible and its deviation can be measured quite accurately (Ghosh and Dubey, 1999). CT is an effective method for evaluating styloid process length, angulation and other morphological characteristics (Fig. 11) (Wada et al., 1991; Murtagh et al., 2001; Renzi et al., 2005; Savranlar et al., 2005). In cases of vascular compression, a sagittal CT angiography of the neck can also be effective in assessing blood flow disturbance (Chuang et al., 2007; Petrovic et al., 2008). During the lidocaine infiltration test, 1 ml of 2% lidocaine is administered to the area where the styloid process is palpable in the tonsillar fossa. If the patient's symptoms and local sensitivity subside the test result is considered positive and Eagle's syndrome is diagnosed (Prasad et al., 2002). 3D-CT is considered the gold standard in the radiological diagnosis (Nayak et al., 2007). It provides an accurate measurement of the length and angulation of the styloid process and is considered to be the best supplement to the plain radiograph (Onbas et al., 2005). It is also used to determine surgical approaches in some cases (Nickel et al., 2003; Lee and Hillel, 2004; Wang et al., 2006; Karam and Koussa, 2007).

TREATMENT

Eagle's syndrome can be treated either conservatively or surgically by excising the elongated styloid process. The appropriate treatment is chosen based on the severity of the symptoms (Ettinger and Hanson, 1975) and the pathogenesis of the syndrome. It is widely believed, however, that therapy should

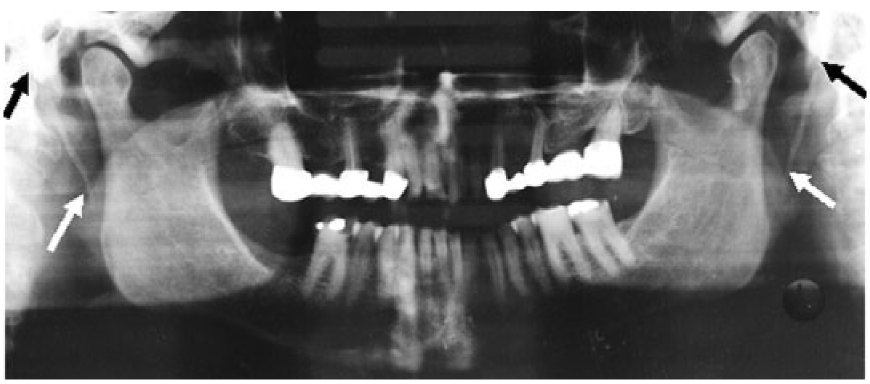

Fig. 10. Panoramic radiography showing bilateral elongation of the styloid process (arrows: the measured points of styloid process).

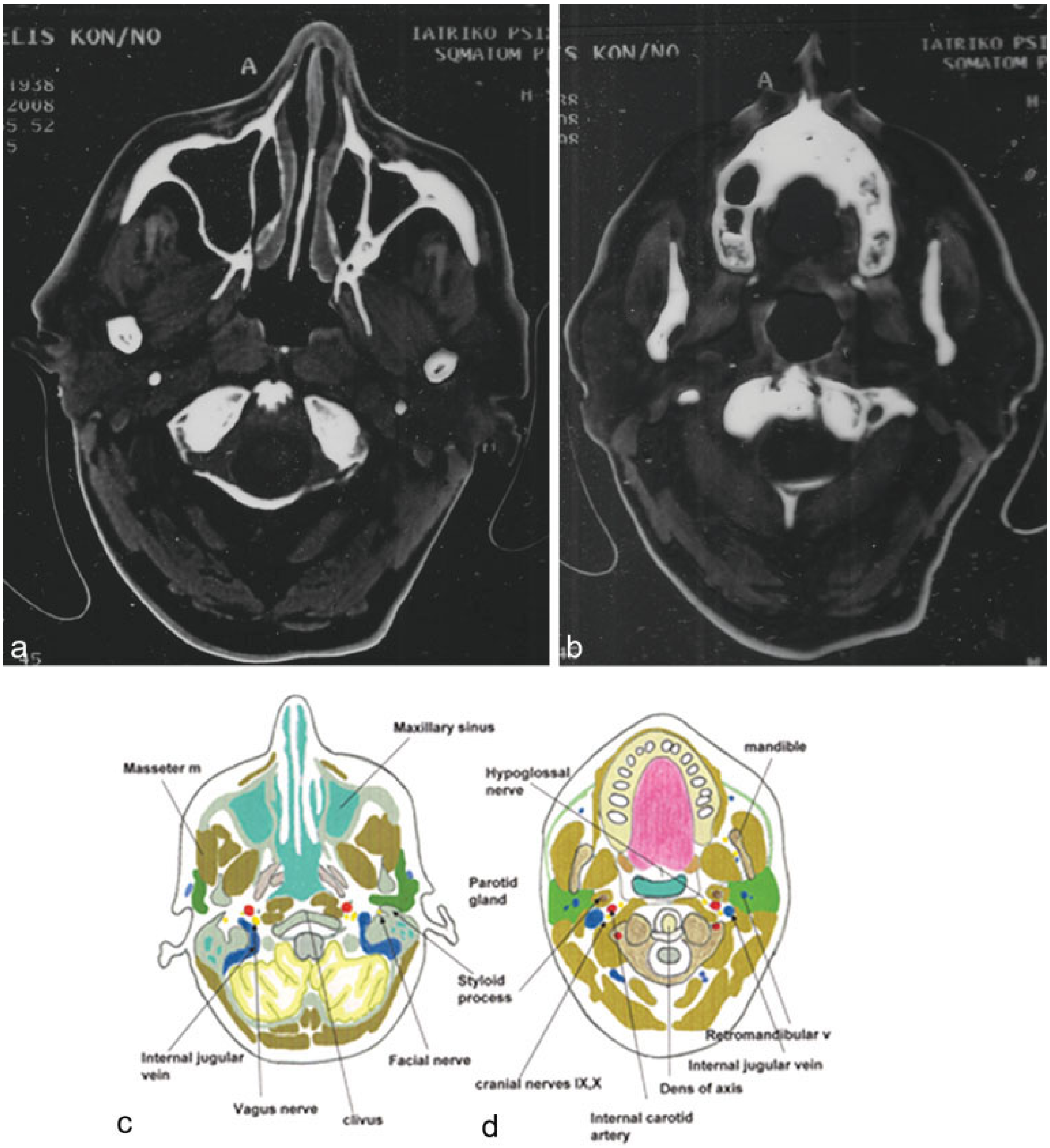

Fig. 11. a–d: CT reconstruction with schematic representation of axial cross-sectional anatomy of the neck (nasopharynx and oropharynx). [Color figure can be viewed in the online issue, which is available at www.interscience.wiley.com.]

begin with conservative treatment followed, if necessary, by surgical excision (Messer and Abramson, 1975; Camarda et al., 1989).

Conservative treatment involves injecting steroids or long-lasting anesthetics into the lesser cornu of the hyoid or the inferior aspect of the tonsillar fossa in order to relieve symptoms (Camarda et al., 1989; Jan, 1989; Bozkir et al., 1999). Treatment with nonsteroidal anti-inflammatory drugs is considered adjunctive therapy after surgical treatment for Eagle's syndrome (Salamone et al., 2004).

Transpharyngeal manipulation, which involves fracturing the elongated process manually under local anaesthesia, has also been proposed (Chase et al., 1986), although long-term results have not been satisfactory (Murthy et al., 1990).

For the surgical excision of the elongated styloid process, two different approaches have been proposed: the extraoral or transcervical and the intraoral or transpharyngeal approach. The extraoral approach was described by Loeser and Caldwell in 1942 (Fini et al., 2000). This approach involves positioning the patient in the supine position with their ipsilateral shoulder slightly elevated off the surgical table (Glogoff et al., 1981). With the head and neck extended and rotated contralaterally, landmarks are made on the patient's skin in order to guide the procedure. After making the incision and identifying and dividing the posterior extension of the platysma muscle, the anatomical elements along the posterior border of the mandible are carefully dissected. A portion of the external carotid artery system can be identified and retracted anteriorly. Directly below the investing fascia of the external carotid or the internal maxillary artery, the styloid process can be identified and easily palpated. After dissecting the fascia and revealing the process, an incision is made into the periosteum, thereby facilitating the reflection of the periosteum and muscle attachments. The styloid process can then be cut close to its base, severing it from the stylohyoid ligament at a point distal to the calcified portion (Loeser and Cardwell, 1942; Chase et al., 1986). Histopathologic examination of the stylohyoid ligament has revealed trabecular bone with normocellular marrow (Salamone et al., 2004). The extraoral approach offers better visibility of the area and lower risk of deep cervical infections, but is more complex, time consuming and leaves a scar on the neck (Ghosh and Dubey, 1999). The risk for transient marginal mandibular nerve weakness is notable (Martin et al., 2008). To avoid scarring, a modified incision described by Appiani and Delfino is used to approach the stylohyoid complex (Buono et al., 2005). Edema of the tonsillar fossa produces compromised speech and swallowing (Chase et al., 1986). Patients with a clinically and radiologically established elongated styloid process (longer than 25 mm) may be managed successfully by surgical resection using an external approach (Ceylan et al., 2008).

The first step in the intraoral approach is to put the patient under general anesthesia and perform a tonsillectomy in order to obtain better visibility of the area. Supporting evidence, however, suggests that this might not be necessary (Perellò, 1995). The tonsillar fossa is then palpated and the tip of the styloid process is identified. The underlying muscles are carefully dissected, divided and reflected downwards. An incision is made into the periosteum of the styloid process, which is removed from the tip to the base of the process. Finally, the process is excised close to its base and the soft tissues of the tonsillar bed are sutured (Prasad et al., 2002). The intraoral approach leaves no scars and is a simpler and less time consuming procedure (de Souza Carvalho et al., 2009). This approach also prevents injury to anatomical structures located in the maxillo-vertebro-pharyngeal region (Fini et al., 2000). The styloid process is less visible and there is a greater risk of deep cervical infection (Ghosh and Dubey, 1999). It is generally accepted that the intraoral approach should only be used if the surgeon is familiar with the technique (Diamond et al., 2001). Apart from the traditional removal of the styloid process, amputating the lesser cornu of the hyoid is also considered as an alternative in cases of pseudostylohyoid syndrome (Van der Westhuijzen et al., 1999).

Complications related to the surgical excision of the styloid process include facial nerve injury, osteomyelitis and infection at the operative site and in deeper tissues of the head and neck (Baddour et al., 1978). Other complications include surgical emphysema of the neck, edema of the submandibular and retromandibular areas (Baddour et al. 1978; Prasad et al., 2002) and thrombosis of the internal carotid artery (Riediger and Ehrenfeld, 1989; Montalbetti et al., 1995). A case of ''first-bite syndrome'' has also been reported, likely caused by damage to the sympathetic innervation of the parotid gland during the operation (Cernea et al., 2007). Also, reossification in the region of the styloid process and stylohyoid ligament may occur and up to 20% of the patients may have a recurrence of symptoms (Zaki et al., 1996).

The overall success rate for treatment, whether it is surgical or not, is in the range of 80% (Baugh and Stocks, 1993).

CONCLUSIONS

It is important to note that an elongated styloid process does not necessarily signify Eagle's syndrome, as the majority of individuals exhibiting this anatomical anomaly experience no symptoms. Additionally, although an elongated process is found bilaterally in most cases, patients typically display unilateral symptoms. Also it is noteworthy that the occurrence of the syndrome correlates with the length of the styloid process, its width and its angulation. In fact a number of mechanisms can result in the onset of the syndrome and are responsible for the variety of symptoms. Consequently, patients may experience any number of symptoms, which often mislead physicians and necessitate the use of other data such as radiological findings to confirm the diagnosis. Both physicians (head and neck, oral and maxillofacial surgeons) and dentists must have a high index of suspicion for this clinical entity. Eagle's syndrome should be included in the differential diagnosis of cervicofacial and pharyngeal pain. The fact that it is often excluded in such cases results in underdiagnosis and, consequently, an underestimation of the incidence of this syndrome.

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