Decompression of Odontogenic Cystic Lesions: Past, Present, and Future

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Tumors and cystic lesions of the jawbones have been described since the late 1600s and it took another 200 years for classification systems to appear in the medical, surgical, and dental literatures. In the late 1800s, Carl Partsch introduced cystostomy, a method by which the cyst is converted into a pouch by suturing its lining to the mucosa of the oral cavity. The purpose of this article is to analyze the history, present, and future of cystic conditions of the jaws and decompression, a modality of treatment that during the past few years has regained the attention of oral and maxillofacial surgeons and pathologists owing to its relative simplicity and effectiveness compared with other conservative options.

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The history of cystic conditions of the jawbones and decompression (also known as *marsupialization* or *exteriorization*)[1] as a treatment modality is intimately related to the birth of oral and maxillofacial pathology. Odontogenic cysts and tumors were noted long before oral pathology was recognized as a specialty of dentistry by the American Dental Association in 1950, just 2 years after the founding of the American Academy of Oral Pathology.

Certainly, oral and maxillofacial pathology as currently understood began during the 1930s and 1940s, an epoch when the world was shaken by the Second World War and whose end led to a flourishing in virtually all fields of knowledge. Textbooks on oral pathology, such as those written by Russell Welford Bunting[2] and Kurt Hermann Thoma,[3] and the first issues of the *Archives of Clinical Oral Pathology* in 1937 and *Oral Surgery, Oral Medicine, Oral Pathology* in 1948 greatly contributed to the consolidation of the specialty.

The purpose of this article is to analyze the history, present, and future of cystic conditions of the jaws and decompression, a modality of treatment that during the past few years has regained the attention of oral and maxillofacial surgeons and pathologists owing to its relative simplicity and effectiveness compared with other conservative options.

Early Descriptions of Odontogenic Entities

The texts by Bunting[2] and Thoma,[3] published in 1929 and 1941, respectively, were the epitome of work that had started 3 centuries previously, mostly with French and British doctors. For example, in 1671 Iean Scultet[4] described cysts of the jaws as "liquid tumors." The idea that a cyst was capable of exerting enough pressure to cause bony expansion was formed at that time.[5] In 1746 Pierre Fauchard[6] provided the first accurate description of an odontoma. In 1774 John Hunter[7] described what seems to be the case of a patient with an odontogenic cyst. Four years later, Anselm Louis Bernard Bréchillet Jourdain[8], on page 19 of his *Traité des maladies et des opérations réellement chirurgicales de la bouche, et des parties qui y correspondent*, described 3 cases that appear to be dentigerous cysts.

During the following decades, dentists, pathologists, and surgeons started to describe with greater regularity cystic lesions and other entities of the maxillary bones. It seems that the first efforts to identify the

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many entities of the oral and maxillofacial region started in approximately 1840, thanks to the release of the *American Journal of Dental Science* (*AJDS*) in 1839, in which such pathologies were available for the dental community to be studied, debated, and characterized. One must not forget that all this happened during the time that is known in history as the *Golden Age of Dentistry* (1835 to 1860).

The first volume of the *AJDS* contained a report of an odontogenic tumor (OT) that currently would be diagnosed as "cementoblastoma."[9] The "periapical cyst" (sac) was described in 1839 by Brown[10] and the dentigerous cyst (distended capsule, osseous cyst, serous cyst) in 1842, although it seems it had been described in France in 1778. The "keratocystic odontogenic tumor" (KCOT; encysted tumor, cyst, cystic carcinoma) was reported in 1844.[5] The following years witnessed an explosion of articles related to oral pathology in the *AJDS* that helped dentists, surgeons, and physicians understand the complex pathologic processes that take place in the mouth and jaws.

It was the famous British pathologist James Paget[11] who in 1853 coined the term *dentigerous cyst* to refer to any cystic condition of dental origin. Other pathologists used many other terms to refer to unilocular cysts. For example, what Emile Magitot called the "radicular cyst" was the same entity Amédée Forget knew as the "periosteal cyst" and Louis-Charles Malassez knew as the "radiculo-dental cyst."

Classifying Odontogenic Tumors

Owing to the increased activity in research and the wide variety of reports on the subject in Europe and the Americas, in 1869 Paul Pierre Broca[12] published *Traité des tumeurs*, where he suggested the first classification of OTs:

I. Ordinary odontomas
  1. Embryoplastic period
    Embryoplastic odontomas
      Fibroplastic
      Fibrous
  2. Odonto-plastic period
    Odonto-plastic odontomas
      Cemental
      Bulbar
  3. Crown formation period
    Coronal odontomas
      Cemental
      Pulpal or dentinal
  4. Root formation period
    Radicular odontomas
      Cemental
      Dentinal

II. Composed odontomas
III. Heterotopic odontomas

Broca used the term *odontoma* for any tumor arising from the formative dental tissues and classified them according to the stage of tooth development when abnormal growth started. In 1885 Malassez[13] made slight modifications that did not gain much attention. Three years later, in 1888, John Bland-Sutton[14] cleverly classified OTs based on the nature of the cells from which the malignancy arose. In this system, he included odontogenic cysts and fibrous odontogenic entities, but the term *odontoma* remained as the usual designation for OTs.[15]

In 1885 Poulet and Bousquet proposed the term *unilocular*[13] to designate single-cavity lesions. The first mention of a "simple cyst" was by the American pathologist Charles Freeman Geschickter. In an article published in 1935, Geschickter[16] mentioned the "simple follicular cyst," which meant that the entity originated from simple (primitive) odontogenic tissues.

Classification of Odontogenic Cysts

At the beginning of the 20th century, a myriad of tooth-related cystic conditions had been described and a classification system was urgently needed.[17] In his 1937 oral diagnosis and treatment planning textbook, Thoma[18] subdivided the classification of the follicular cyst in the following manner:

1. Simple (without tooth formation)
2. Dentigerous
3. With odontoma

In 1949 he added a fourth category, the multiple cyst.[19]

Also in 1937, Hamilton Robinson,[20] one of the most influential early oral pathologists in addition to Thoma and William Shafer, subdivided dentigerous cysts into 4 categories:

1. Simple
2. Compound
3. Eruption
4. Heterotropic

Eight years later (in 1945), Robinson[21] proposed another classification for the cysts of the jawbones.

1. Developmental cysts from odontogenetic tissue
  1.1. Periodontal
  1.2. Dentigerous
  1.3. Primordial
2. Developmental cysts of non-dental origin
  2.1. Median
  2.2. Globulomaxillary
  2.3. Incisive canal cysts
3. Ameloblastomas

In this classification of 1945, the term *primordial* was mentioned for the first time and it seems to be the same "simple cyst" described by Geschickter in 1935. The new term was favored owing to the researcher's beliefs that the cyst arose from remnants of the dental lamina or enamel organ (primordial origin) before enamel had formed, and it replaced a tooth.[22] During the first half of the 20th century, many classification methods for odontogenic cysts and tumors (odontomas) were proposed,[15] and 4 terms were generally used to describe odontogenic cysts: dentigerous, simple or primordial, and follicular. The term *follicular* was applied to dentigerous and primordial cysts, until Mervyn Shear[23] established in 1976 that *dentigerous* and *follicular* were synonymous.

Rise and Fall of Decompression: the Past

Understanding the behavior of benign and malignant conditions of the maxillofacial region has taken almost 4 centuries, if the work of Scultet[4] is used as a starting point. Four hundred years of research have helped to not only undress these pathologies to the point of knowing them as they are, but also to understand how they are best treated. Long before World War II, investigators knew that odontogenic cysts and tumors had the potential to recur, regardless of the treatment modality used. Factors such as type, size, and nature of the lesion, localization, etiology, and age of the patient play a role in determining the therapy.

During the past 20 years, researchers have been able to give reliable statistics on the chances an entity has to recur after a given treatment plan. It is not the purpose of this article to animate the controversy of whether marsupialization should be used. Rather, this article addresses the historical facts related to this surgical option, which is currently used to treat unicystic ameloblastomas,[1,24] KCOTs,[25] and other odontogenic cysts and tumors.[26]

A cyst is a pathologic cavity in the soft tissue or bone with an outer wall composed of connective tissue and an inner wall composed of epithelium. The cavity has a watery, semisolid, or colloidal content. Cysts gradually enlarge owing to a combination of osmotic pressure[27] and release of growth factors and prostaglandins.[28,29] This persistent pressure exerted on the bony walls coupled with biomolecules cause bone resorption while the entity expands. Therefore, mitigating the pressure by making a small window into the cyst and keeping it open guarantees permanent drainage, thus preventing its enlargement. Eventually, bone growth will reduce the space.

This applied physical principle was the rationale that Polish professor Carl Franz Maria Partsch (Fig 1) used to introduce the concept of cystostomy in 1892[30] and cystectomy in 1910.[31] In an article published in German at the end of the 19th century, Partsch reported the conversion of a cyst into a pouch by suturing its lining to the mucosa of the oral cavity (Fig 2). Today, cystostomy is known as Partsch I or marsupialization, a term from the modern Latin *marsupialis*, from the Latin for the Greek *marsupion*, meaning "pouch." Cystectomy (Partsch II) is enucleation and primary closure.

The antibiotic era was not in sight when Partsch introduced cystostomy, which, fortunately, was not associated with the development of infection, therefore becoming the treatment of choice during the first decades of the 20th century. In 1932 E.B. Dowsett[32] noted that enucleation often led to wound infection and treatment failure. Expanding on Partsch's concept, Thomas[33] described decompression in 1947. Thomas pointed out its benefits, which were maintenance of pulp vitality, preservation of the inferior alveolar nerve or maxillary sinus, prevention of fracture of the jaw, and low risk of recurrence.

FIGURE 1. Carl Franz Maria Partsch (1855 to 1932).

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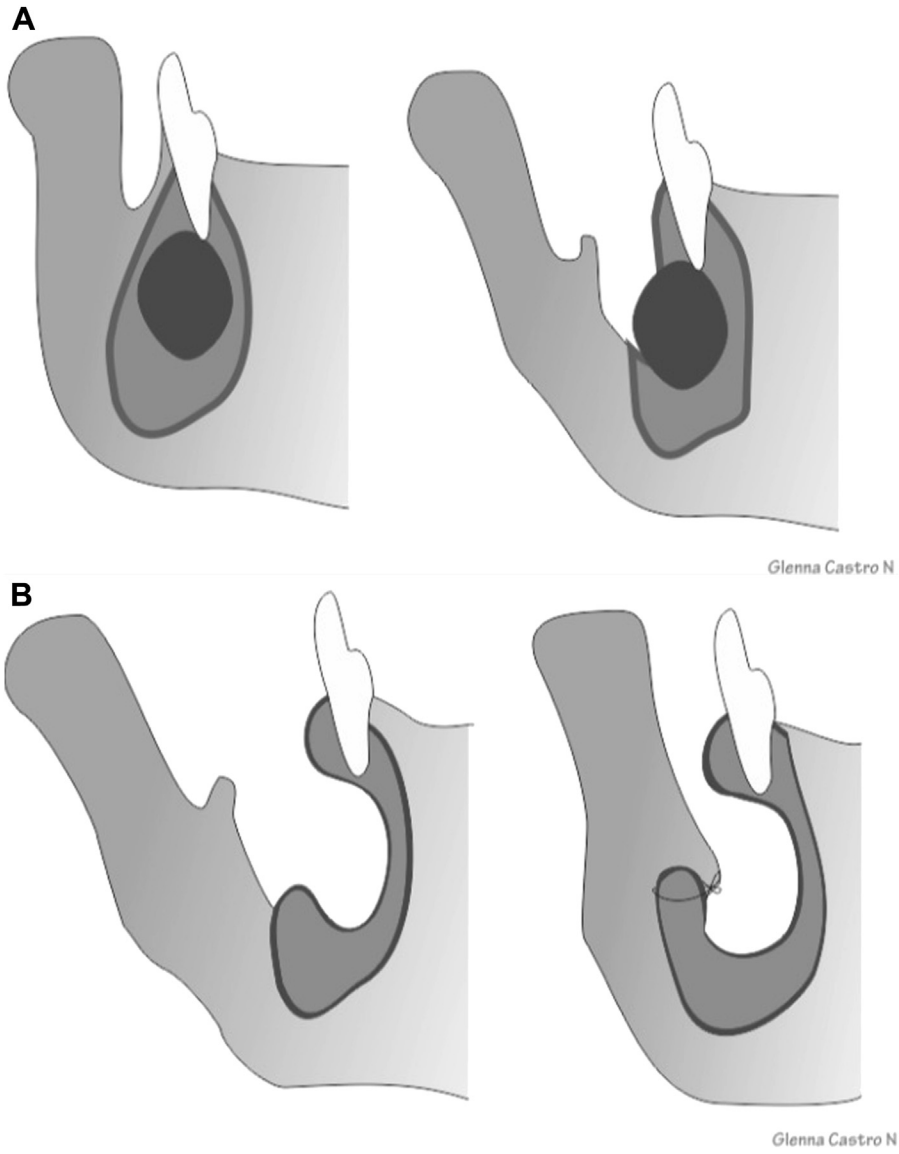

FIGURE 2. *A*, Odontogenic cystic condition. *B*, Partsch I (cystostomy).

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The term *decompression* encompasses marsupialization and is defined as any method used to relieve intracystic pressure by keeping a patent opening into the exterior, which could be the mouth (Fig 3), nose (Fig 4), or maxillary sinus (Fig 5). Two decades after Dowsett's article, the panorama for marsupialization and decompression was about to change. The introduction of antibiotics was almost a death knell for marsupialization and decompression and at the same time acted as a lifeboat for enucleation and primary closure, which, aided by antibiotics, became the more accepted method of treatment owing to faster results. The real understanding of the many pathologic conditions of the mouth started during the 1950s and the aggressive behavior of some of them was fully documented during this time.

Another factor that reinforced the idea that marsupialization and decompression should be avoided was the better understanding of what Hans Peter Phillipsen[34] called the *odontogenic keratocyst* in 1956 (ie, KCOT). During the next 30 years, articles would warn against malignant transformation occurring within odontogenic cysts.[35-37] Although marsupialization was deemed unpredictable at that time, a minority still had faith in it.[38] What is more, the great surgeon Harry Seldin[39] reported the usefulness of marsupialization as a presurgical treatment for unicystic ameloblastoma in children. To combat recurrence, aggressive methods prevailed until the end of the 20th century.[40-44]

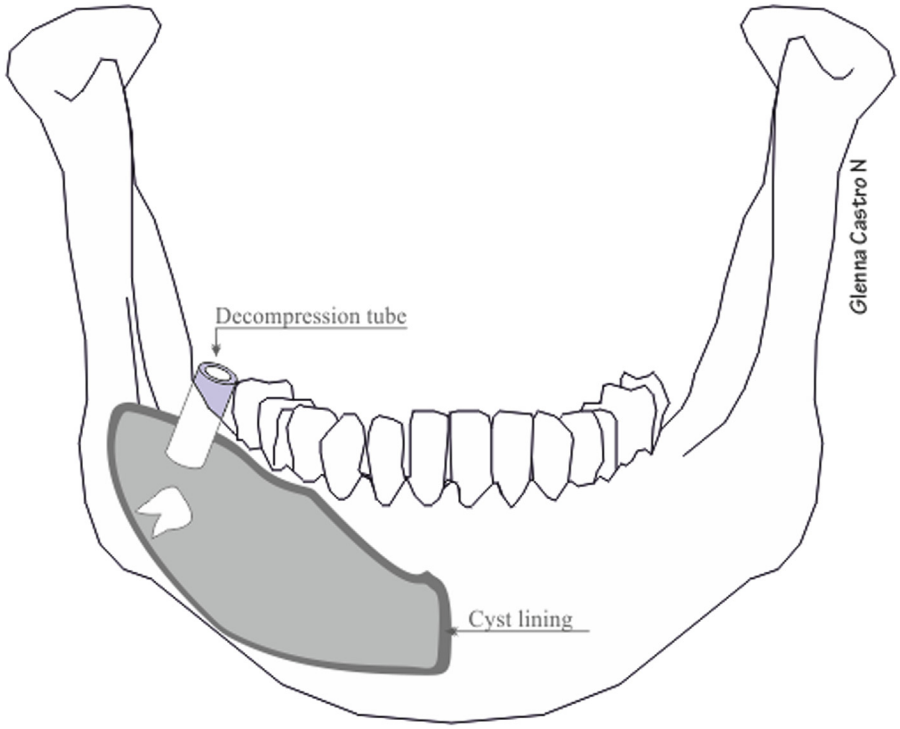

FIGURE 3. Decompression to the mouth.

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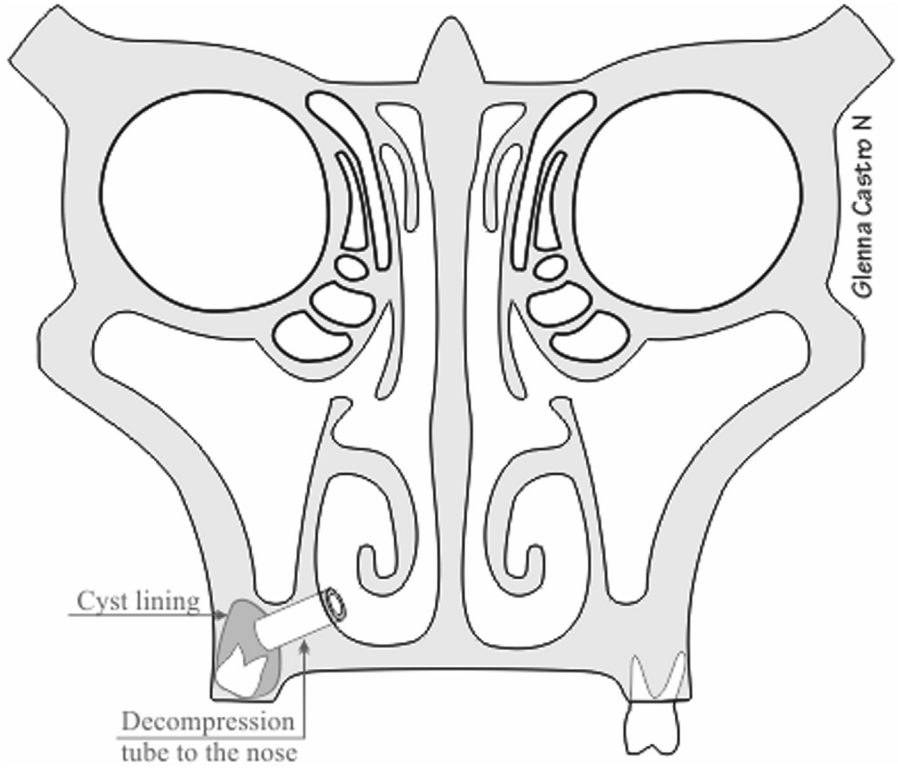

FIGURE 4. Decompression to the nose.

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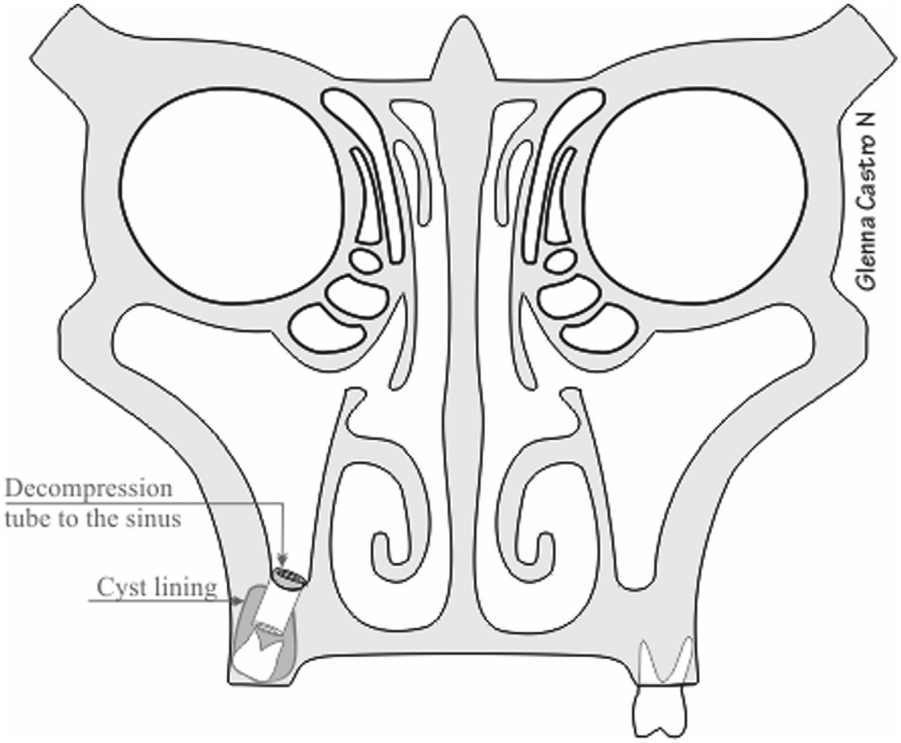

FIGURE 5. Decompression to the nasal sinus.

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Reborn as the Phoenix: the Present

In 1991 an article by Brøndum and Jensen[45] paved the way for the reintroduction of marsupialization and decompression, which recently had resurrected like the Phoenix. In 1996 Marker et al[46] successfully decompressed 23 odontogenic keratocysts using small polyethylene tubes (Fig 6). They concluded that decompression resulted in new bone formation, thickening of the cyst wall, and conservation of bone and anatomic structures. Another interesting conclusion was that the keratocyst epithelium was modulated histologically to non-keratocyst after decompression. Despite such optimistic results, from the literature review it seems that it was not until the early 2000s that marsupialization and decompression regained the attention of surgeons and pathologists, much at the urging of Pogrel.[47,48]

The past 10 years have observed how researchers design conduit systems to decompress odontogenic cysts and tumors. Of note, in a systematic review published by Lau and Samman[1] regarding recurrence related to treatment modalities of ameloblastoma, they called attention to the fact that 3 reported cases treated by marsupialization had complete resolution, which is paradoxical because tumors do not regress by exteriorization.

In 2007 Huang et al[24] described a decompression plug for ameloblastomas. This plug, which is basically the same as that described by Marker et al,[46] was effective in decreasing tumor volume and minimizing the extent of surgery. The following year, Tolstunov[49] used a catheter for the same purpose. One of the drawbacks of decompression is the size of the tube, which could be dislodged over time. This issue was cleverly tackled by Kolokythas et al[26] using a 16-gauge needle to create a passage for a 28-gauge

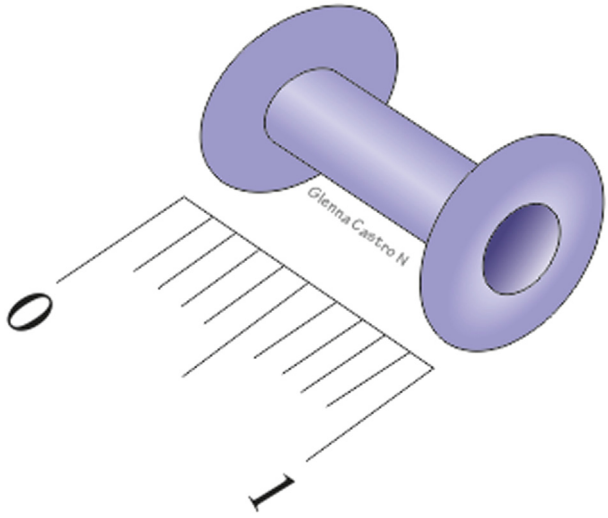

FIGURE 6. Decompression plug as described by Marker et al.[46]

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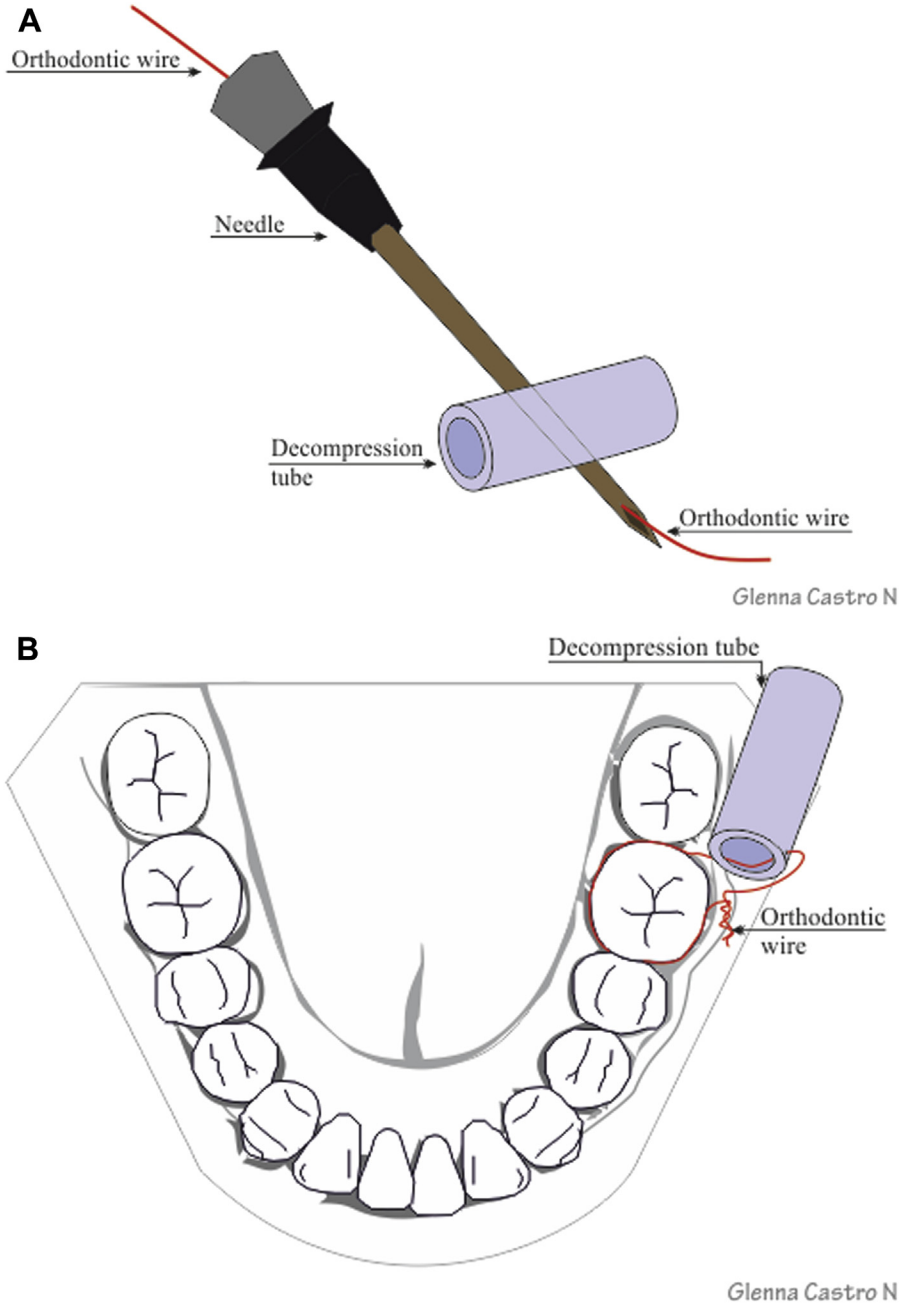

FIGURE 7. Fixation method using a wire as described by Kolokythas et al.[26] *A*, Passing the wire through the needle. *B*, Fixing the tube to the teeth.

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wire that would be secured to the teeth (Fig 7). Another stent fixation method, using 1.2-mm screws (Fig 8), was proposed in 2012 by Swantek et al.[50] In 2014 Gao et al[51] introduced a thermoplastic resin stent, with and without a clasp. In 2015 Delgado-Rueda et al[22] maximized the benefits of decompression by using 2 decompression anesthesia tubes in the same patient (Fig 9).

The Future of Decompression

Decompression, as any other treatment technique, is not a panacea. The benefits of this old method have been described and proved over the years, and as patients and surgeons become less aggressive, this technique will become more popular when indicated. Cases must be carefully selected and the surgeon has

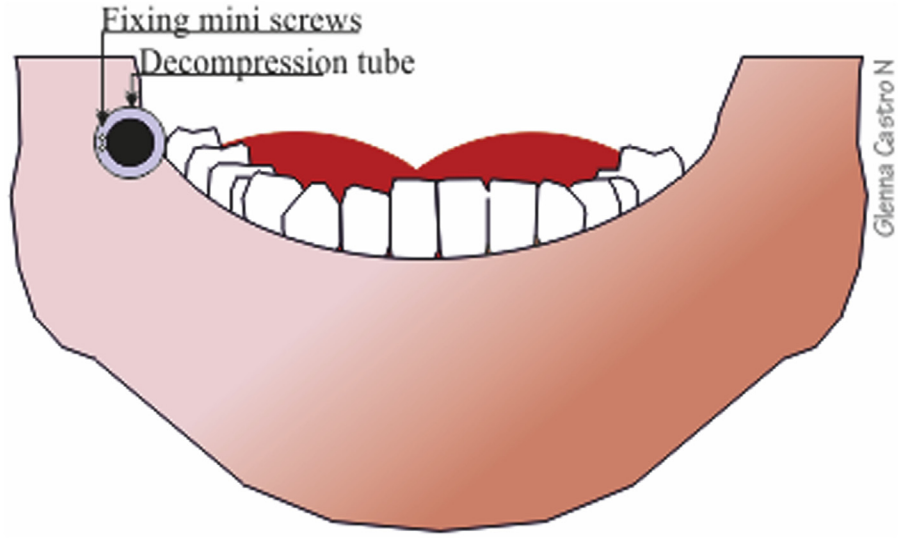

FIGURE 8. Fixation method using small screws.

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to weigh many factors, such as patient age, type of lesion and time of evolution, and the patient's cooperation.

A Closing Remark: Introduction and Acceptance of a Pleonasm

To the unprepared eye, the word *pleonasm* might be confused with *neoplasm*, but they are different! A *pleonasm* is the use of more words than necessary to convey meaning, as a fault of style or for emphasis. This seems to be the case of the expression *odontogenic origin*, which was introduced to the dental literature in the late 1970s. In this sense, Shafer's *Textbook of Oral Pathology*[52], a book widely used in North American and Latin American dental schools, helped disseminate the pleonasm. In the Spanish version of the book, the expression was translated as *origen odontogénico*. The word *odontogenic* comes from Greek *odous, odont-*, meaning "tooth," and *guénesis*, which means "origin." Therefore, the expression *odontogenic origin* (and its Spanish translation, *origen odontogénico*) must be avoided because the term *odontogenic* already means that the condition being described is related to dental tissue.

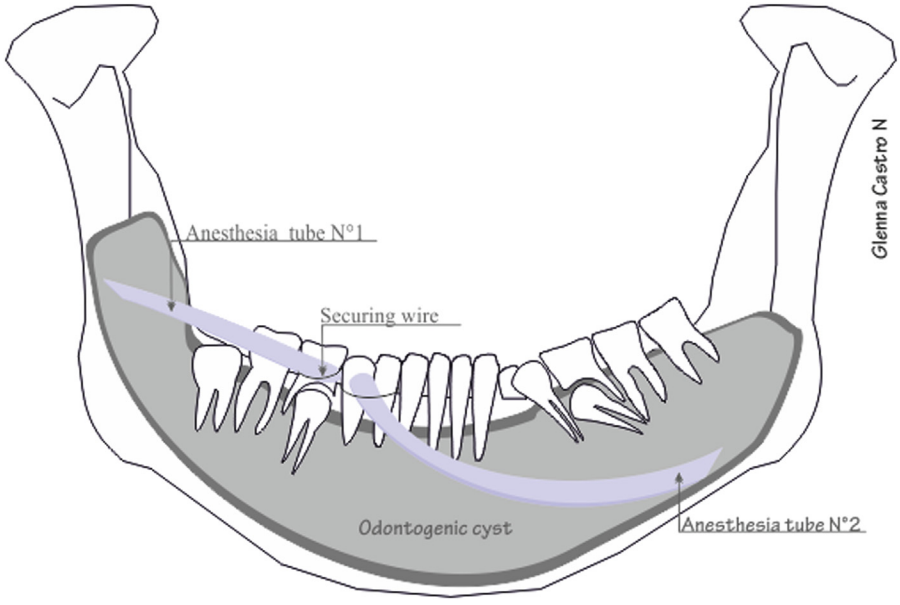

FIGURE 9. Two decompression tubes (double decompression) in the same patient.

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