

# Le Fort I osteotomy to treat recurrent pleomorphic adenoma and facial deformity

## Osteotomía Le Fort I para tratar un adenoma pleomórfico recurrente y la deformidad facial

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**ABSTRACT:** Pleomorphic adenoma is a benign lesion of the salivary glands. The most common intraoral site is the palate, with tendency to affect females between the fourth and seventh decades of life. The most reported treatment for this lesion is the surgical removal. Therefore, the aim of this report was to present the case of a 31-year-old female patient who presented a recurrence of pleomorphic adenoma and facial deformity. Recurrence was observed 22 year after first surgical treatment. Tumor involved the palatal area, nasal floor and maxillary sinus; the patient showed an augmented vertical growth of the maxilla and mandibular retrognathia with a previous compensatory orthodontic treatment. The treatment included Le Fort I osteotomy for enucleation of the tumor, upper reposition of the maxilla and a chin osteotomy for advancement. 5 year follow-up show a stable result with no recurrence. Diagnosis and treatment strategies are discussed. We concluded that this treatment is stable; Le Fort I osteotomy can help to treat the tumor and facial deformity at the same time.

**KEY WORDS:** Salivary Gland, tumor, orthognathic surgery, pleomorphic adenoma.

### INTRODUCCIÓN

Tumors of the salivary gland are uncommon with a incidence of 2.5 to 3 cases per 100,000 per year in the Western world; 80% of these lesions are with no malignance (Nourwali & Dar-Odeh, 2019). Pleomorphic adenoma (PA), originally called a benign mixed tumor in 1866, was described microscopically in 1874. Histological morphology of the PA shows epidermoid and myoepithelial cells and this condition (Clauser *et al.*, 2004).

PA usually occurs in the fourth to sixth decades of life mainly in females, with a female-to-male ratio of 2:1 (Patigaroo *et al.*, 2014; Kumar *et al.*, 2015; Wu *et al.*, 2016; Singh *et al.*, 2019; Sarma *et al.*, 2020) and it is present in the

40-70% of the salivary gland tumors (Patigaroo *et al.*, 2014). 70% of the tumors of the minor salivary glands are pleomorphic adenomas (Clauser *et al.*, 2004) and the most common intraoral site is the palate (42.8-68.8%), followed by the upper lip (10.1%), buccal mucosa (5.5%) and floor of the mouth (Patigaroo *et al.*, 2014).

Unusual sites are the tongue, tonsil, pharynx, sinuses, larynx, epiglottis, trachea and even external auditory canal (Sahoo *et al.*, 2013). Palatal PA tumors are slow, painless growths, observed as a well-delineated submucosal mass and covered with a normal mucous membrane (Clauser *et al.*, 2004). Sometimes, mucosal ulcerations are observed due

to trauma during chewing. Major gland tumors are usually encapsulated, as opposed to minor gland tumors (Moon, 2019).

Abnormalities in the chromosome loci 8q12 and 12q15 (Sarma *et al.*, 2020) and the exposure to radiation or viruses some has been related to the etiology; however, it is controversial. In term of treatment, the standard treatment of palatine PA, includes wide local excision and removal of the involved bone, requiring proper visualization and complete removal of the lesion without rupture to avoid recurrence (Moon, 2019).

An alternative approach to treatment palatine PA includes Le Fort I osteotomy, which provides direct visualization to the pathological area (Buchanan & Hyman, 2013). Although this technique is related to treat facial deformities, Le Fort I osteotomy was first described by Langenbeck in 1861 for the treatment of benign tumors of the pterygopalatine fossa in two patients and in 1864 by Cheever (1870) for the resection of a nasopharyngeal tumor. Therefore, the purpose of this report is to show the use of Le Fort I osteotomy to treat recurrent PA at the same time with a facial deformity with a long-term follow up.

## CASE REPORT

A 31-year-old female patient was referred to our department with a four-month evolution of swelling in her face, with no other symptom. She reported previous surgical removal of a pleomorphic adenoma in the palate 22 years ago. The soft tissue over the lesion was no change in term of color and morphology. Computed tomography (CT) demonstrated a well-defined hypodense lesion (18.7 mm x 25.0mm x 23.3mm) involving the hard palate, right maxillary sinus and right nasal cavity (Fig. 1). Biopsy was taken in the center of the palate swelling.

The main concern in this case was the PA; however, a second concern was included by the patient in term of a high smile and a short chin (Fig. 2). Facial analysis showed a higher vertical growth, gummy smile and mandibular retrognathism. Furthermore, she was treated by orthodontics exclusively for dental compensation with an Angle Class II malocclusion without surgery indication at those moment. Due to the position of the tumor and the diagnosis of facial deformity, Le Fort I osteotomy and chin surgery were planned. Orthognathic planning was performed as regular technique

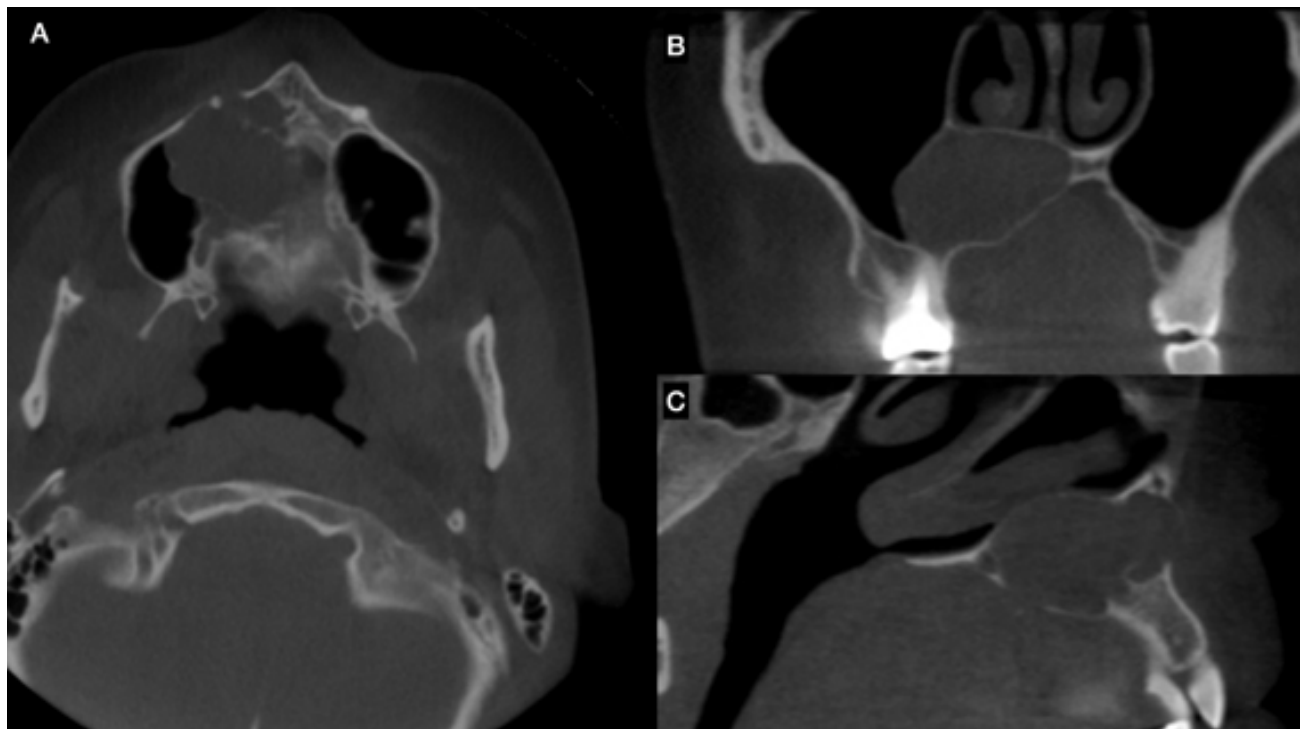


Fig. 1. CT showing a well-defined area in the maxillary bone. A) axial section involved the maxillary sinus and nasal floor. B) Coronal section showing the displacement of the right turbinate. C) Sagittal section showing the compromise of the teeth with the tumor.

using 3D image, photography and splint print and 3D print of the skull and the tumor was realized for surgical analysis previously to surgery.

After the down fracture of the maxilla the tumor was observed; the approach included a peripheral osteotomy (Fig. 3); a careful enucleation of the tumor was realized with observation of the integrity of the palatine tissue and retropharyngeal flap. After that, the maxillary impaction for correction of the vertical maxillary position was performed and then the chin osteotomy for advancement was realized. The maxilla was stabilized with L-shaped 2.0 plate with four screws in the nasomaxillary and zygomaticomaxillary buttress and a X-shaped plate with four screws used in the chin. Turbinectomy was not realized.

No teeth were removed, but endodontic treatment was needed for some teeth due to the involvement of the tumor. After 7 year follow-up, no signs or symptoms of local recurrence is observed and the stability of the procedure was confirmed.

## DISCUSSION

Pleomorphic adenomas are described as benign epithelial tumors with capsule. Although these lesions show as benign, tumor infiltration into the capsular tissue may represent a risk factor for recurrence. Therefore, this lesion can recur even several years after surgery, so long-term follow-up is necessary mainly in pediatric patients (Taiwo *et al.*, 2018; Moon 2019).

In this case, the patient reported having undergone surgical removal of a PA in the same region at 9 years old (22 years previous to our diagnosis); in the case of close follow-up, an early diagnosis could be performed with a more conservative surgical treatment. PA show a slow painless growth. However, large tumors often produce swelling, which could be associated with pain and ulceration. The most common region is the palate (hard palate) (Clauser *et al.*, 2004; Kumar *et al.*, 2015; Singh *et al.*, 2019).



Fig. 2. Clinical and histological sample. A) Le Fort I osteotomy with the upper tumor view. B) The Le Fort I approach to confirm the excision of the tumor with an adequate hemostasia and prepared to perform the impact surgery and fixation. C) Stratified pavement epithelium, with eroded areas, rich in histiocytes in cord structures immersed in a fibromyxoid stroma associated to central cystifications conferring an adenoid aspect, without cell atypia, necrosis, or ciliated epithelium; E) Tubulo-cystic neoplasm organized in the myxo-hyaline stroma composed of two types of cells, anastomosing in adenoid spaces, with central eosinophilic protein secretion.

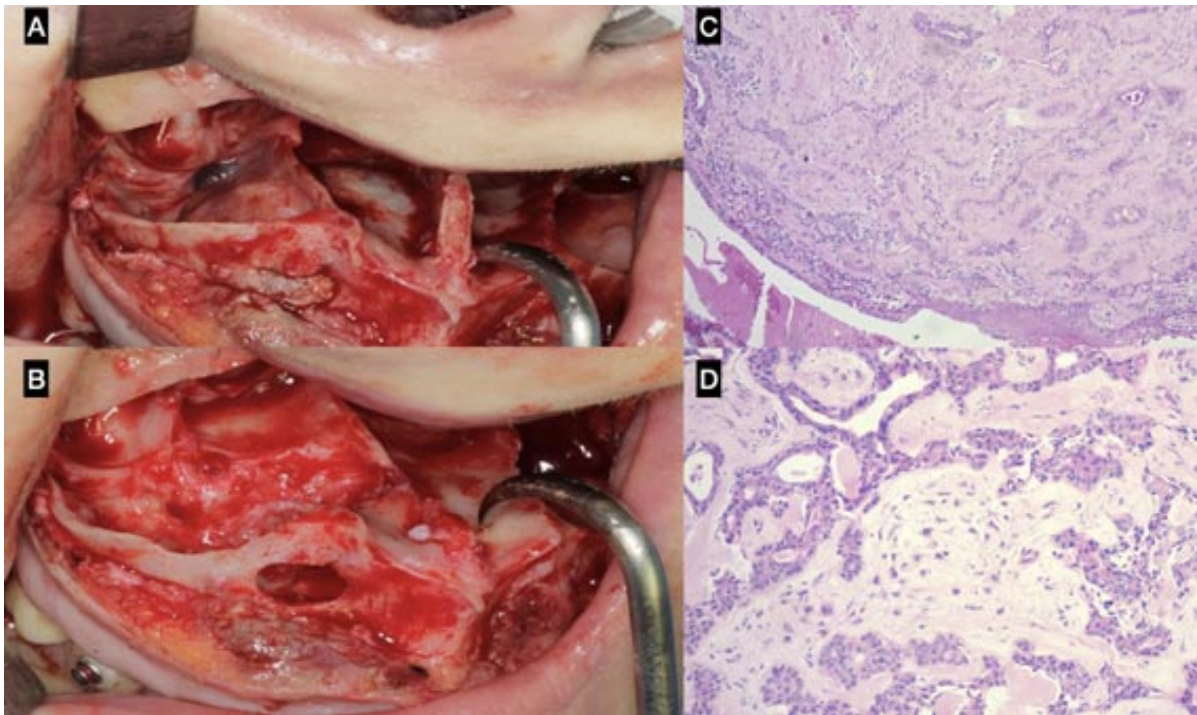


Fig. 3. Clinical view of the patient before and 5 year after of surgery. A) Maxillary protrusion with a high vertical growth. B) Chin and mandibular retrognathia with previous dental compensation. C) Frontal view with a better facial projection and stability after surgery. D) New position of chin after advancement surgery showing a better facial balance.

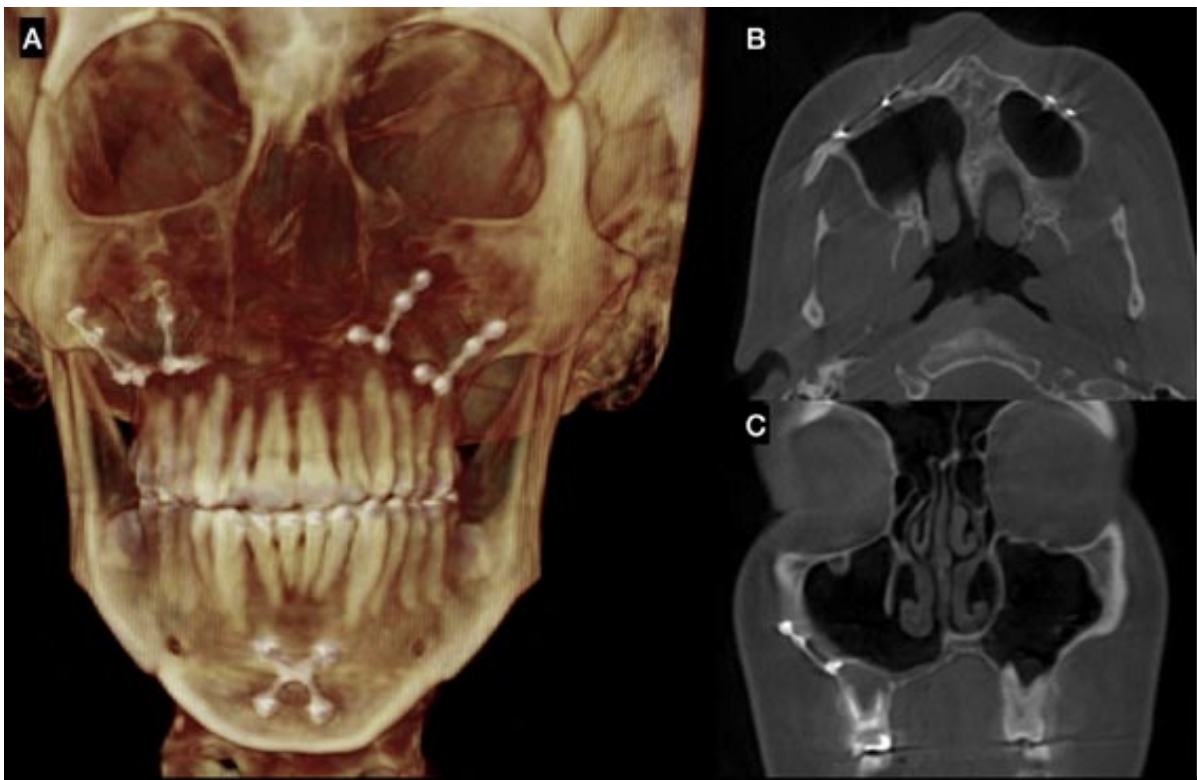


Fig. 4. CT 5 year after surgery. A) 3D reconstruction showing stability of new bone position and fixation. B) Axial section with no evidence of recurrence and good function of the maxillary sinus. C) Nasal and maxillary condition 5 year after surgery with a lower and lateral defect with no functional complications.

CT was necessary to confirm the involvement of the hard/soft tissue of the palate, the nasal cavity and the maxillary sinus. Due to the possibility of seeding cells in the biopsy time, it is recommended that this procedure be performed at the center of the tumor rather than at the margins, preferably using a punch, because this approach is accompanied by minimal morbidity and minimal damage to the specimen (Moon, 2019).

Histopathological description of this entity is clear (Wu *et al.*, 2016; Daniels *et al.*, 2007) and this case was consistent with the description. Differential diagnosis includes palatal abscess, mucoepidermoid carcinoma, adenoid cystic carcinoma, fibroma, lipoma, lymphoma, neurilemmoma, neurofibroma and rhabdomyosarcoma, cysts and soft tissue tumors (Jorge *et al.*, 2002; Dhanuthai *et al.*, 2009; Patigaroo *et al.*, 2014).

The goal of the surgical management of the PA in the hard palate is the excision with the capsule and the remove of the periosteum or bone (if they are involved) to avoid recurrence (Patigaroo, 2014; Sotong *et al.*, 2015). Where the overlying mucosa is ulcerated, it is removed to resolve with a spontaneous soft tissue repair or the reposition using a local flap. Any rupture of the capsule or incomplete excision will leave residual tumor cells behind, which will result in recurrence (Moon, 2019).

Some techniques could be used for excision, depending on the tumor position and extension. In palatal area, the inability to delineate adequate posterior and upper margins is a major drawback of the usual direct mucosal approach to excise the tumor (Sotong, 2015). Le Fort I osteotomy allows for proper observation of the tumor from the upper view; this technique assist to obtain a wide excision margin, thereby minimizing the possibility of recurrence (Daniels *et al.*, 2007). The use of 3D models is an important step in the surgical planning of this case and in term of tumor analysis and orthognathic planning (Mayrink *et al.*, 2013).

Long-term follow-up of at least five years is necessary because the local recurrence should be considered (Alves *et al.*, 2018). In our case we observed a recurrence after 22 years and other cases reported PA recurrence in the palate after 16 years in a 39-year-old male patient (Berardi *et al.*, 2009).

The prognosis for PA of minor salivary glands is usually considered to be better than in the parotid gland, because the morphology of the parotid gland and the nerve and vessels in the area. PA recurrence rates of 2% to 44% have been reported in the literature (Kumar *et al.*, 2015); in

the case of recurrent tumors, it can be multiple nodules or a single mass; recurrences on the palate can be serious, because they can go through the palatine foramen and take place close to the skull base (Yasumoto *et al.*, 1999; Daniels *et al.*, 2007). In the present case, the recurrence of the PA was included in the treatment of a facial deformity to obtain a better position of the maxilla and to build better conditions for the bone repair in the area of the tumor; the long time follow up showed a good stability and repair of the area.

We can conclude that the Le Fort I osteotomy is a good technique to perform the excision of the PA in the palatal area; this can be included in a global face treatment to perform another osteotomies and orthognathic surgery as well.

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**RESUMEN:** El adenoma pleomorfo es una lesión benigna de glándulas salivales. El sitio más común en la boca es el paladar, con tendencia a afectar mujeres entre los 40 y 70 años. El tratamiento utilizado de forma más habitual es de tipo quirúrgico. El objetivo de este reporte fue mostrar un caso de un paciente de 31 años de edad, de sexo femenino que presentó recurrencia de un adenoma pleomorfo en conjunto con una deformidad facial. La recurrencia se observó 22 años después del primer tratamiento quirúrgico. El tumor incluía el área del paladar, piso nasal y seno maxilar; la paciente mostró un aumento vertical del crecimiento maxilar con retrognatia mandibular junto a un tratamiento ortodóncico compensatorio previo. El tratamiento realizado incluyó una osteotomía de Le Fort I para enucleación del tumor, reposición superior de maxila y osteotomía del mentón para avance de 5 mm. 5 años después se presentó con resultados estables, sin recurrencia de la enfermedad. El diagnóstico y tratamiento es analizado y discutido. Podemos concluir que el tratamiento es estable; la técnica de Le Fort I puede ayudar a tratar el tumor maxilar y la deformidad en el mismo tiempo quirúrgico.

**PALABRAS CLAVE:** Glándula salival, tumor, cirugía ortognática, adenoma pleomorfo.

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