Unexplained permanent constriction of the jaws (Langenbeck's disease): about two cases

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Case Report

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ABSTRACT

Introduction Hypertrophy of the coronoid process is a rare pathology, described by Langenbeck in 1853. This anomaly can be uni- or bilateral. Two entities have been described with few cases in the literature, which may cause some diagnostic confusion. Through this work we have highlighted Langenbeck's disease by two cases treated in our department of maxillofacial surgery and stomatology of the Moulay Ismail military hospital in Meknes.

Observation we report two cases of patients consulting in our service for a chronic limitation of the mouth opening never followed. In both cases orthopantomogram and CT of the facial bone showed bilateral hyperplasia of the coronoid processes. Langenbeck's disease was retained as a diagnosis. The treatment for our two cases was based on bilateral resection via the endobuccal route under general anesthesia with nasotracheal intubation by fibroscopy. The histological study of the surgical specimens confirmed the diagnosis of Langenbeck's disease.

Discussion Among the bone causes of limitation of mouth opening, hypertrophy of the coronoid process or Langenbeck's disease is one of the most frequent. Panoramic dental imaging makes it possible to suspect hyperplasia but only 3D CT can confirm the diagnosis. Langenbeck's disease is an increase in the size of the coronoid process without bony abnormality. It is usually bilateral. The painless nature of this disease is the reason for the diagnostic delay as for our two cases presented. Coronoidectomy associated with postoperative maxillofacial physiotherapy is the treatment of choice.

Key Words: Langenbeck's disease, coronoidectomy, hypertrophy of the coronoid process, limitation of mouth opening,

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INTRODUCTION

Hypertrophy of the coronoid process is a rare pathology, described by Langenbeck in 1853 [1, 2]. This anomaly can be uni- or bilateral.

Two entities have been described with few cases in the literature, which may cause some diagnostic confusion.

Langenbeck's disease, characterized by a conflict between the coronoid process and the zygomatic arch; indeed the hypertrophied coronoid apophyses strike the zygomatic bone at the mouth opening.

The other entity is Jacob's disease, always unilateral affection. Described in 1899 to be characterized by the presence of an osteochondroma, sometimes post-traumatic [3, 4].

Through this work we have highlighted Langenbeck's disease by two cases treated in our department of maxillofacial surgery and stomatology of the Moulay Ismail military hospital in Meknes.

OBSERVATION

Our first case was that of a young patient aged 26 consulting in our service for a chronic limitation of the mouth opening never followed.

This limitation of mouth opening without any particular context had been present since childhood according to the patient, associated with temporomandibular joint cracking, with no notion of aggravation or improvement.

On clinical examination, a mouth opening limited to 20 mm is noted (Figure 1).



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Panoramic imaging and a CT scan of the facial bone were performed with sections in the different planes and threedimensional reconstructions having detected bilateral (Figure 2)



hypertrophy of the coronoid processes impeding the mouth opening, without any bone lesion of the jaws.

The diagnosis of Langenbeck's disease was then retained. The surgical procedure was scheduled under general anesthesia with nasotracheal intubation by fibroscopy. A bilateral intraoral coronoidectomy was performed

(Figure 3).



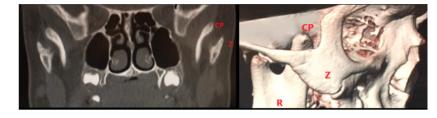
The anatomo-pathological examination of the surgical specimens confirmed the presence of bone hypertrophy without structural abnormality.

The immediate post-operative results have been promising (Figure 4).

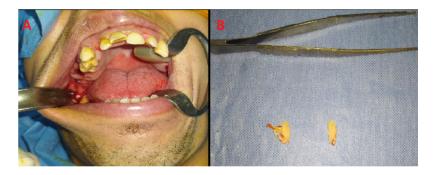


Several physiotherapy sessions were started early after the surgery and the reassessed mouth opening at three weeks and then at three months was 36 mm with improvement in temporomandibular joint noises.

The second case to present is that of a 35-year-old patient also consulting for a permanent limitation of mouth opening. The interrogation in search of a cause of permanent constriction of the jaws did not point to anything. The clinical examination objectified a mouth opening limited to 22 mm. Orthopantomogram and CT of the facial bone showed bilateral hyperplasia of the coronoid processes (Figure 5).



Langenbeck's disease was also retained for this case as a diagnosis. The treatment was based on bilateral resection via the endobuccal route under general anesthesia with nasotracheal intubation by fibroscopy. The histological study of the surgical specimens confirmed the diagnosis of Langenbeck's disease. The immediate postoperative (Figure 6)



results and at 3 months were positive with a satisfactory mouth opening at 37 mm.

DISCUSSION

Among the bone causes of limitation of mouth opening, hypertrophy of the coronoid process or Langenbeck's disease is one of the most frequent [3, 5].

There is no known precise etiology for this disease, but hypotheses are mentioned: hyperactivity of the temporal muscles, endocrine disorders, genetic abnormalities or chronic disc displacements [2].

Panoramic dental imaging makes it possible to suspect hyperplasia but only 3D CT can confirm the diagnosis. Langenbeck's disease is an increase in the size of the coronoid process without bony abnormality. It is usually bilateral. About 80 cases have been reported in the literature; the painless nature of this disease is the reason for the diagnostic delay as for our two cases presented [6, 7, 8].

The maxillofacial scanner with three-dimensional reconstruction and the histological study make it possible to distinguish the different forms of hyperplasia of the coronoid process. Coronoidectomy associated with postoperative maxillofacial physiotherapy is the treatment of choice. The surgical approach can be endobuccal, exobuccal or mixed.

Coronoidectomy via the endobuccal approach avoids the aesthetic and functional risks associated with lesion of the mental branch of the facial nerve [9]. However, this route is not always possible in cases of severe trismus.

The endobuccal route, the most practiced at present, presents little risk for the nervous structures and has the advantage of not causing unsightly scars and the postoperative course is simple and quick both in terms of healing and pain. It is important to underline the essential interest of postoperative maxillofacial physiotherapy for obtaining and maintaining a satisfactory mouth opening. This must begin immediately postoperatively and maintained for at least 6 months to maintain the result obtained in the long term.

CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

CONCLUSION

The ignorance of this type of pathology often leads to diagnostic delays as illustrated in this work. Its diagnosis is made easier by CT with three-dimensional reconstruction. It is interesting to know pathologies such as Langenbeck's disease and to know how to evoke it in front of cases of old mouth opening limitations in patients with no medical history.

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