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► **To cite this version:**

Thanh Thuy Nham, Pierre Corre, Justine Loin, Hélios Bertin. Maxillary protrusion in a black female, what is your diagnosis?. *Journal of Stomatology, Oral and Maxillofacial Surgery*, 2023, 124 (1), pp.101335. 10.1016/j.jormas.2022.11.011 . hal-04600620

**HAL Id: hal-04600620**

**<https://hal.science/hal-04600620v1>**

Submitted on 4 Jun 2024

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## TITLE

Maxillary protrusion in a black female, what is your diagnosis?

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## **Maxillary protrusion in a black female, what is your diagnosis?**

A 15-year-old patient with severe SS homozygous sickle cell disease (SCD) was referred by her orthodontist for correction of an unesthetic gum smile and a Class II dental occlusion (Figure 1). She was undergoing orthodontic treatment and four premolars had been removed under local anaesthesia. She reported having had five vaso-occlusive seizures in her life, and trans-cranial Doppler revealed severe neurovascular impairment. The clinical examination revealed a 7-mm gingival exposure when she smiled, a lack of occlusion of the lips at rest and retrogenia (Figure 2). The occlusal examination revealed the presence of an Angle Class II malocclusion. A lateral cephalogram and Delaire's cephalometric analysis revealed maxillary anterior protrusion and vertical excess from maxillary origin associated with mandibular retrusion (Figure 3). What is your diagnosis?

**The sickle cell gnathopathy** is an unusual maxillary protrusion considered as the result of a bone marrow hyperplasia and compensatory erythropoiesis induced by chronic hemolytic anemia [1]. The maxilla undergoes an exaggerated reactional development and can have a major functional and aesthetic consequence in patients. SCD is one of the most common recessive inherited haemoglobinopathies in the world, with 30% of the population carrying the sickle cell trait and nearly 2% of new-borns being affected [2]. Due to the increasing migratory population in recent decades, the sickle cell trait now has a global distribution according to the World Health Organization, especially in Europe and the French overseas departments [3]. The facial deformity could concern a third of the SCD population and should be known by maxillofacial surgeons [4]. The surgical management involves the classical techniques of Le Fort I osteotomy for maxillary impaction combined with a self-rotation of the mandible and genioplasty. However, orthognathic surgery should be given due consideration in patients at risk of severe and acute decompensation. Management can only be considered in a multidisciplinary team involving orthodontist, surgeon, anesthesiologist and hematologist [5]. Peri-operative measures include the monitoring of oxygen saturation and blood pressure. Pre-oxygenation and compensation for blood loss by isotonic solutes for normovolemia are usually performed. A one-step surgical procedure with concomitant jaw surgery and wisdom tooth extraction is indicated to

limit the repeated general anesthesia. The use of facial icing during and after surgery is contraindicated in order to maintain normothermia. A postoperative monitoring in the intensive care unit for the first 24 hours is often necessary, as well as a prolonged hospitalization for pain management.

## **FIGURE CAPTION**

Figure 1. Frontal photograph showing the 7mm-gingival smile in relation to the maxillary protrusion.

Figure 2. Frontal photograph revealing a vertical excess of the lower third of the face associated with retrogenia.

Figure 3 Delaire's cephalometric analysis on lateral cephalogram showing an anterior and vertical protrusion of the maxilla, partly compensated by the extraction of the premolars.

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