

CASE REPORT

Stroke Due to Orthognathic Surgery: Case Report of a Rare Complication

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Rafael Maffei Loureiro, MD§ Andre Felix Gentil, MD, PhD* Roberto Franco Morgulis, MD¶ Eduardo Carvalhal Ribas, MD, PhD*|| Summary: Le Fort I osteotomy is a frequent surgical procedure used in orthognathic surgeries to treat severe malocclusions and is associated with relatively rare surgical complications. Here, the authors report a case of thrombotic ischemic stroke as a result of this procedure, a complication still not described in the literature. A 19-year-old man with class II malocclusion and retrognathia underwent orthognathic surgery for aesthetic purposes. The surgery included a Le Fort I maxillary osteotomy with vertical impaction, bilateral sagittal split ramus osteotomy for mandibular advancement, and genioplasty. Postoperatively, the patient developed left eye blindness, headache, somnolence, aphasia, and right hemiplegia. Medical imaging showed the Le Fort I line of fracture extending from the maxillary osteotomy to the left optic canal and to the left carotid canal, with osseous fragments impinging the petrous segment of the internal carotid artery, left carotid artery occlusion and associated to an ischemic stroke at the left middle cerebral artery territory. Treatment required decompressive craniectomy and later focused on clinical stabilization, infection management, orthognathic care, neurorehabilitation, and cranioplasty. The hemiplegia and aphasia partially recovered during 12 months, and final dental occlusion was appropriate. Our report demonstrates that an unfavorable Le Fort I fracture trajectory can lead to ischemic stroke and severe neurological deficits. (Plast Reconstr Surg Glob Open 2022;10:e4471; doi: 10.1097/ GOX.00000000004471; Published online 19 August 2022.)

A nteroposterior malocclusions are classified according to the mandible relative position to the maxilla: class I if the relation between teeth is acceptable but line of occlusion is impaired, class II if the mandible is posterior to the maxilla, and class III if anteriorly positioned.¹ Their overall global distribution is 74.7%, 19.56%, and 5.93% for class I, class II, and class III malocclusion, respectively.¹

Treatments for class II patients vary according to malocclusion severity, skeletal maturity level, and facial

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Copyright © 2022 The Authors. Published by Wolters Kluwer Health, Inc. on behalf of The American Society of Plastic Surgeons. This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal. DOI: 10.1097/GOX.00000000004471 appearance. Orthognathic surgery is usually performed for severe malocclusions. Le Fort I osteotomy, alone or in combination with sagittal split mandibular ramus osteotomy, is a frequent procedure for these cases, and is associated with relatively rare surgical complications.^{2,3}

Here, the authors report a case of a patient with class II malocclusion and retrognathia who developed left eye blindness and an ischemic stroke as a complication of Le Fort I maxillary osteotomy. To our knowledge, this is the first report of thrombotic ischemic stroke as a surgical complication of this procedure.

CASE REPORT

A 19-year-old man with class II malocclusion and retrognathia was referred for surgical treatment for aesthetic purposes. Orthognathic surgery was performed by an oral and maxillofacial surgeon in northern Brazil, and included a Le Fort I maxillary osteotomy with vertical impaction, bilateral sagittal split ramus osteotomy for mandibular advancement, and genioplasty. The patient complained of headache and somnolence on the first postoperative day

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Related Digital Media are available in the full-text version of the article on www.PRSGlobalOpen.com. (POD) and developed sudden right hemiplegia at POD 2. Computed tomography (CT) showed the line of fracture extending from the maxillary osteotomy to the left optic canal and to the left carotid canal, with osseous fragments impinging the petrous segment of the internal carotid artery. It also revealed an ischemic stroke related to the left middle cerebral artery territory. The patient underwent a left decompressive craniectomy on the same day.

At POD 12, he was transferred by airplane to our institution, located 3000 km away, to receive better medical treatment and start neurological rehabilitation. At admission, neurological examination revealed right hemiplegia, severe motor aphasia, and absent left direct pupillary light reflex with consensual preservation, suggesting left amaurosis due to optic nerve dysfunction. CT and AngioCT better detailed the fractures and neurological injuries (Fig. 1). [See figure, Supplemental Digital Content 1, which displays the coronal (A) and axial (B) CT images showing skull base fractures (arrowheads in B), one of them involving the left optic canal (arrows), http://links. lww.com/PRSGO/C133.]

Treatment was focused on clinical stabilization, infection management, orthognathic care, and neurorehabilitation, with specialized physiotherapy, occupational therapy, speech therapy, a nutritionist, and psychology. Several other complications during hospitalization were aspiration pneumonia, urinary tract and COVID-19 infections, hyponatremia, haloperidol impregnation, non-steroidal anti-inflammatory drug (NSAID) allergic reaction, and need for tracheostomy and gastrostomy. At POD 36, with hemiplegia, aphasia, and swallowing partially recovered, the patient was transferred to a specialized transition care clinic and later discharged home after a month.

CT scans performed during follow-up noted left frontal encephalomalacia development and compensatory ipsilateral ventricular enlargement, preventing the skin flap and brain to accommodate naturally inside the craniectomy skull defect. Cranioplasty, with an external ventricular

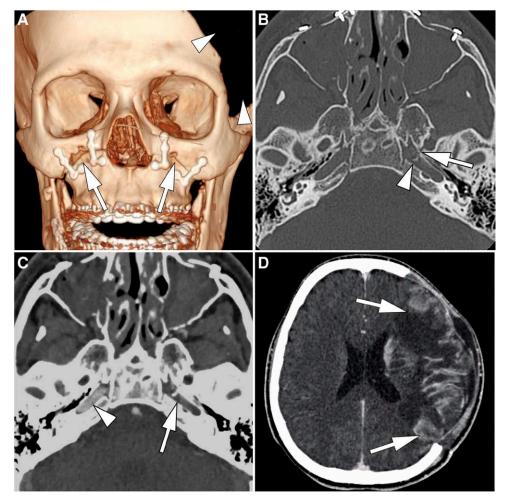


Fig. 1. Skull base fractures and stroke after Le Fort I surgery. Three-dimensional reconstruction CT image (A) shows a Le Fort I osteotomy (arrows) and a left craniectomy (arrowheads). Axial CT image (B) shows a skull base fracture involving the left foramen ovale and carotid canal (arrow) with bone fragments inside the left carotid canal (arrowhead). Axial angiographic CT image (C) shows occlusion of the left carotid artery (arrow). Note the normal right carotid artery for comparison (arrowhead). Axial contrast-enhanced CT image (D) shows early subacute stroke in the left middle cerebral artery territory (arrows) with brain herniation through the craniectomy defect.

drainage brief placement and polymethyl methacrylate prosthesis confection, was performed 7 months after the ischemic stroke and involved neurosurgeons and plastic surgeons. The hemiplegia and aphasia partially recovered during 12 months of follow-up, and final dental occlusion was appropriate. This report was approved by our institutional ethics committee, and patient consent was given.

DISCUSSION

Le Fort I osteotomies are considered important progress in maxillary surgery. In summary, the anterior surface of the maxilla is exposed by a gingival incision and a horizontal osteotomy is performed at the level of the nasal floor, from the nasal septum to the lateral pyriform rims, crossing below the zygomaticomaxillary junction and passing through the pterygomaxillary junction to interrupt the pterygoid plates. The nasal septum and vomer are also separated from the palate. Finally, the maxilla is downfractured with digital pressure or forceps. If osteotomies have been done completely, this movement can be performed smoothly. However, if excessive force for the downfracture is needed, previous osteotomies probably were inappropriately made, and the pterygomaxillary separation can be completed with multiple unfavorable fracture trajectories, which are associated with direct and indirect neurovascular damage.4

Complication rates range from 6% to 9%, most commonly including hemorrhages, infections, malocclusions, tooth loss, maxillary necrosis, and relapse.^{2–5} Pseudoaneurysms, arteriovenous fistulas, decreased visual acuity, and blindness are rare complications.^{6–9} A PubMed database search using the terms "stroke" and "orthognathic surgery" retrieved 11 articles, but most were related to facial sensitivity to brush stroke stimuli or chewing strokes performance, and none mentioned neurological strokes. Recent large case series^{3,5} and systematic reviews^{2,4} about orthognathic surgery complications mention cranial nerve impairment, but not strokes or hemorrhages.

Newhouse et al¹⁰ reported, in 1982, a hemorrhagic stroke after significant intraoperative bleeding following Le Fort I osteotomy and maxilla downfracture; angiography revealed an arteriovenous fistula between the internal carotid artery and internal jugular vein. No cases of thrombotic ischemic stroke as a Le Fort I osteotomy complication were found in the literature.

CONCLUSIONS

Orthognathic surgery is usually safe, but significant complications may arise. Our report demonstrates that Le Fort I osteotomies and maxilla downfracture have to be done with attention to avoid unfavorable fracture trajectories, which can lead to neurovascular damage and severe ischemic stroke.

ETHICAL APPROVAL STATEMENT

The present article was approved by our institutional ethics committee (Hospital Israelita Albert Einstein, São Paulo, Brazil).

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