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Mandibular Fracture in a Hemifacial Microsomia Patient following Implant Failure and Hardware Infection: A Case Report

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Abstract

Hemifacial microsomia (HFM) is a complex congenital condition with heterogeneous malformations of the facial skeleton that almost always involves mandibular hypoplasia. Here we introduce a unique case in which a patient with HFM had initially successful optimization of facial symmetry using a polyetheretherketone implant for mandibular augmentation. However, multiple factors associated with the intraoperative and postoperative course, including hardware failure and infection, led to diminished mechanical strength of the mandible, ultimately resulting in a mandibular fracture. In this unique case presentation of HFM, we discuss the various factors that contributed to mandibular weakness and increased susceptibility to fracture.

Keywords

- hemifacial microsomia
- polyetheretherketone implant
- mandibular fracture
- mechanical strength
- biomechanics

Introduction

Hemifacial microsomia (HFM) describes a spectrum of anomalies associated with abnormal embryological development of the first and second branchial arches.[1] Among all deformities of HFM, mandibular hypoplasia followed by facial muscle hypoplasia and microtia is the most common.[2] Both mandibular and soft tissue deficiencies in HFM must be addressed to achieve optimal occlusion, functional jaw movements, facial symmetry, and overall aesthetic improvement.[1] Depending on the severity of mandibular hypoplasia, various management options are available, such as mandibular osteotomy and autologous fat grafting, distraction osteogenesis, temporomandibular joint reconstruction, and alloplastic implants (i.e., porous polyethylene or polyetheretherketone [PEEK] constructs).[1] Regardless of operative technique, optimizing mandibular mechanical strength by ensuring appropriate hardware placement and mandibular stability as well as averting infection is imperative to minimizing bony weakness and subsequent susceptibility to postoperative fracture.

Here we present a case of a 16-year-old girl with HFM who underwent left mandibular augmentation with a PEEK implant, but subsequently developed hardware infection followed by a mandibular fracture in the bony area previously infected. To our knowledge, no known case has ever been reported in the literature regarding the susceptibility of a congenitally dysplastic mandible to fracture, particularly in an area that was previously manipulated by hardware, later resulting in hardware failure and infection. Our purpose is to discuss the factors contributing to mandibular fracture in this patient with underlying mandibular hypoplasia associated with HFM.

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Case

A 16-year-old female with congenital left HFM was referred to the craniofacial plastic surgery department for concerns regarding facial asymmetry. Clinical evaluation was notable for micrognathia with the left lower third of the face projected less anteriorly than the right. She had normal ocular and auricular features but had visible soft tissue deficits along the left nasal ala base. She had a class I occlusion with a 6-mm overjet and normal occlusal cant despite prior orthodontic treatment. Her temporomandibular joint movements were palpable bilaterally. Computed tomography (CT) imaging showed left mandibular hypoplasia (Pruzansky grade I) and a borderline dysmorphic right mandible with narrow anteroposterior dimensions (►**Fig. 1**).

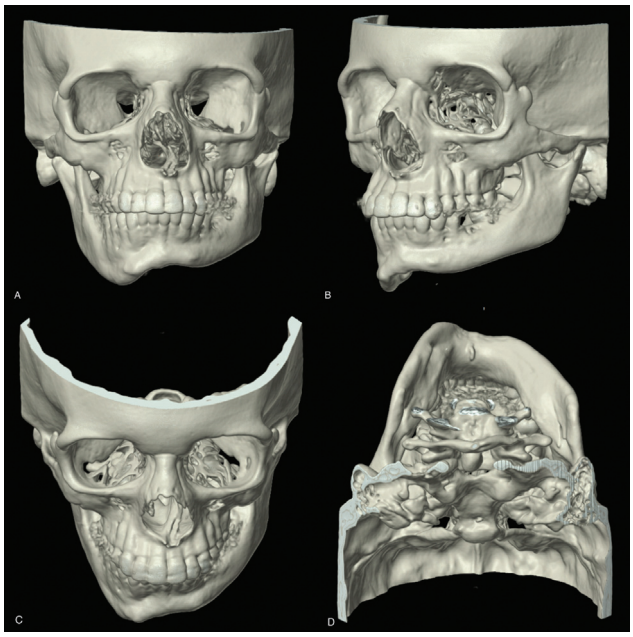

Fig. 1 Three-dimensional computed tomography analysis with anteroposterior view (**A**), oblique view (**B**), superior view (**C**), and inferior view (**D**) showing left mandibular hypoplasia and borderline dysmorphic right mandible contributing to facial asymmetry.

After discussion of treatment options, the patient and her family elected for left mandibular and chin contouring with a PEEK implant and autologous fat grafting to the left nasal ala base. The custom implant with predrilled holes was virtually constructed using computer-assisted manufacturing and consisted of two pieces that keyed into each other – one posterior piece for the mandible and one anterior piece for the chin (►**Fig. 2**). To properly position the implant, a left inferior buccal sulcus incision and an extraoral stab incision along the left mandibular border were made to expose the symphysis and body of the mandible superiorly to the inferior alveolar foramen. When positioning the implant, the most lateral right screw broke inside the anterior piece after it was fixated to the bone. The screw could not be removed without sacrificing implant integrity, so the decision was made to leave the screw given there was still good implant stabilization to the bone. The two pieces of the implant interlocked very well and had excellent stabilization after complete fixation to the bone. The superiorly positioned mental nerve was neurolyzed for the sake of placing the implant and then repaired with 8–0 nylon suture. Fifty grams of fat was harvested from the neck through the extraoral incision and another submental stab incision and were injected into the left midface just deep to the left nasal ala. Soft tissue and skin were sutured in layers to achieve watertight closure.

On postoperative days 7 and 21, she had improved chin projection, good occlusion, and well-healing incisions

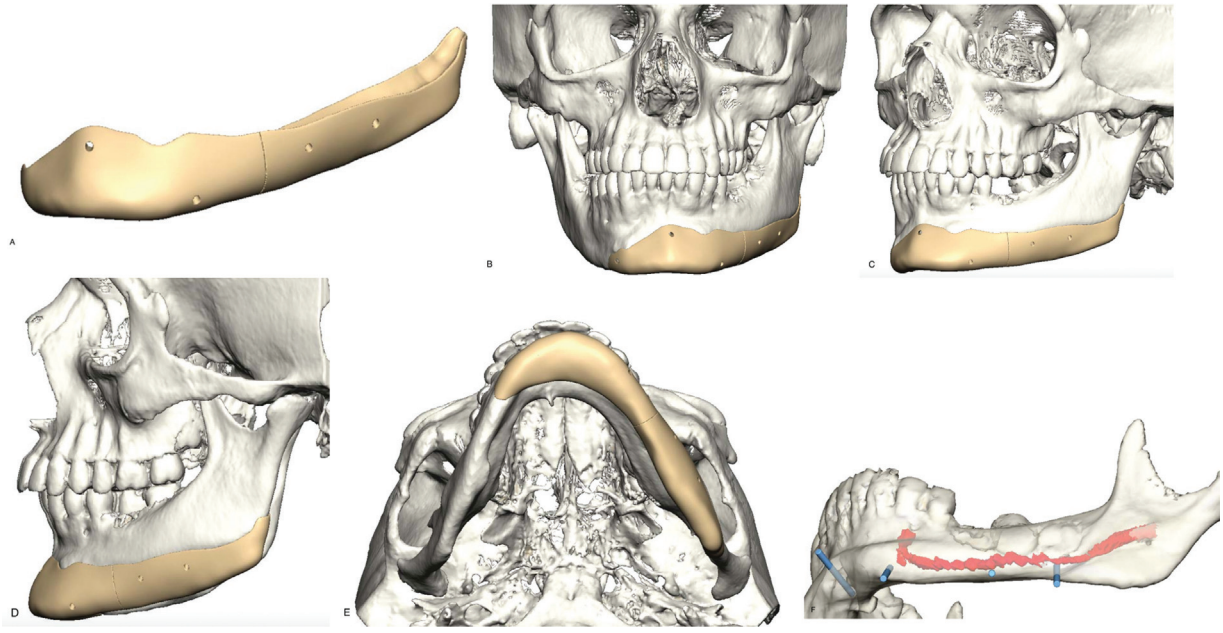

Fig. 2 Two-piece polyetheretherketone implant, designed with computer-assisted manufacturing, interlocked together for augmentation of the hemimandible (**A**). Anteroposterior view (**B**), oblique view (**C**), lateral view (**D**), and inferior view (**E**). Screws are angled to avoid the dental roots and inferior alveolar nerve (**F**).

Fig. 3 Facial profile prior to polyetheretherketone implant placement (**A, B**) compared with 3 weeks postoperatively (**C, D**).

(►**Fig. 3**). However, on week 5, she experienced left cheek swelling and purulent drainage from the submental incision site. CT images revealed significant lucency within the right parasymphyseal mandible around the most lateral right PEEK implant screw and a submandibular fluid collection that tracked all the way to the skin, indicating hardware infection (►**Fig. 4**). The infected PEEK implant was completely removed and the surrounding soft tissue of a total area of 8 cm^2 was debrided. The mandibular bone appeared healthy and was copiously irrigated with antibiotic solution. The wound was packed with gauze strips. Bacterial cultures grew *Prevotella* species and *Staphylococcus aureus* sensitive to the antibiotic course given.

Her condition improved and incisions appropriately healed. Two months after removal of the implant, she was in a golf cart rollover accident, suffering an open right mandibular fracture. Intraoral exam was notable for open bite and open right parasymphyseal fracture between the lateral incisor and canine with an exposed tooth root. Sensation was diminished bilaterally along the mental nerve distribution, which was stable since her prior jaw surgeries. No other facial injury or palpable bony defect was found. CT imaging showed a mildly displaced right parasymphyseal mandibular fracture at the same location where bony lucency was seen on prior imaging when her implant was infected (►**Fig. 5**).

Fig. 4 Significant lucency (**A, B**) within the right parasymphyseal mandible around the right most, partially broken screw of the polyetheretherketone implant. Submandibular fluid collection (**C**) suggests implant infection.

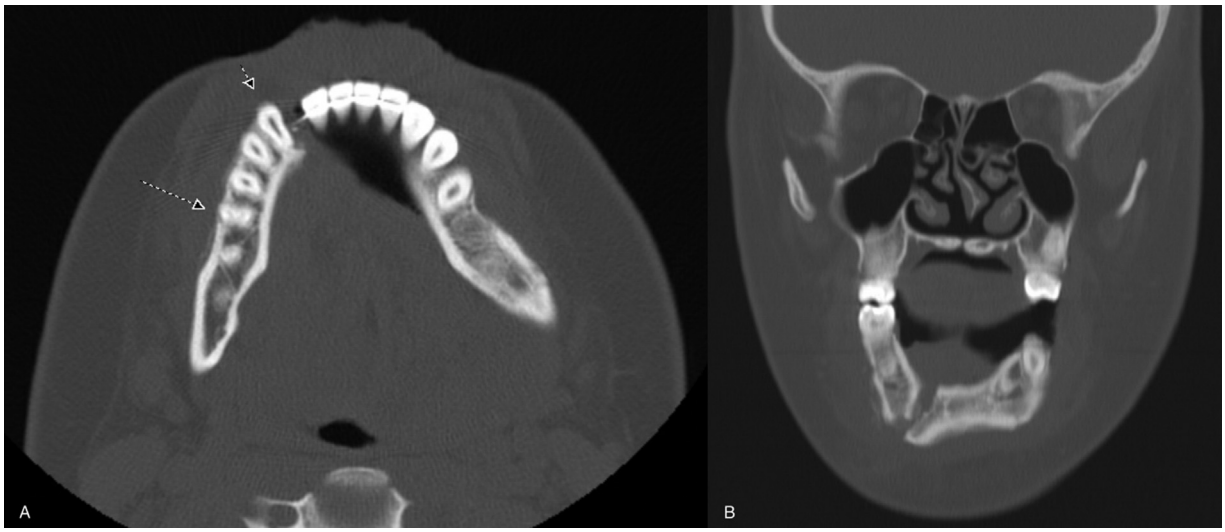

Fig. 5 Axial (**A**) and coronal (**B**) computed tomography images show right parasymphyseal mandibular fracture extending just lateral to right lateral incisor.

Fig. 6 Patient's profile is shown following removal of hybrid arch bars in anteroposterior (**A**), oblique (**B**), and lateral (**C**) views. Chin projection and occlusion remained stable since her initial augmentation surgery despite subsequent removal of the infected polyetheretherketone implant and operative fixation of mandibular fracture.

She underwent open reduction and internal fixation of the right mandible with intraoperative maxillomandibular fixation (MMF) to stabilize the occlusion. The right mental nerve was significantly stretched and encased in scar tissue, likely secondary to prior surgeries. Hence, the nerve was neurolyzed to safely place the plating system for mandibular repair. A 1.5-mm tension band was applied after reduction of the fracture. A 2.5-mm reconstructive plate was fixed into place along the lower mandibular border. MMF wires were then removed and occlusion was confirmed. Guiding elastics were placed on the arch bars. The wound was debrided and incisions were closed in interrupted suture layers.

Her postoperative course was unremarkable. At 2 weeks, the arch bars were removed. She had good occlusion consistent with her premorbid condition, so no orthodontic treatment was needed. Her asymmetric facial profile related to her congenital HFM was not significantly worsened after PEEK explantation or traumatic injury. Because of her residual facial asymmetry without functional deficits, a collaborative decision was made to hold off on aesthetic surgery to optimize symmetry until her swelling improved and postoperative scars had time to settle (►**Fig. 6**).

Discussion

HFM is a challenging condition to address since diagnosis requires identification and classification of all phenotypical elements involved, including non-craniofacial structures. In general, mild deformities like Pruzansky grade I mandibular hypoplasia can be treated with mandibular osteogenesis, osteotomy, or augmentation, while more severe grade III defects typically require graft reconstruction.[1] Our patient had mild mandibular hypoplasia classified as Pruzansky grade I with minimal soft tissue deficiencies along her left nasal ala base. Given her mild degree of HFM, mandibular augmentation with a PEEK implant and autologous fat grafting was performed with initially successful improvement in her facial symmetry. However, she developed an implant

infection requiring explantation and then subsequently suffered a mandibular fracture in the bony area that was previously infected. Because of these unique series of events, the question arose as to all the factors that may have increased susceptibility to mandibular fracture.

When undergoing implant placement, it is important to ensure appropriate tension is applied when placing screws intraoperatively. The torque generated from fixing the screw produces a force known as preload. The greater the preload, the greater the resistance to movement along the interface, which ultimately leads to reduced screw loosening.[3,4] Consequently, optimizing preload should diminish the risk of screw loosening and therefore maximize mandibular mechanical strength.

The broken screw that remained at the site of the PEEK implant likely weakened the mechanical strength of the mandible. Possible causes for implant screw fracture can frequently be attributed to mechanical problems or infection.[3] Given the acute, intraoperative nature of the screw fracture, the etiology is more likely attributed to issues related to technical placement of the PEEK implant–mandibular connection system. Lee et al concluded that screw loosening was associated with positioning and placement of the implant, type of implant, implant diameter, type of implant prosthesis, and type of implant–abutment connection. Among the 1,928 dental implants in their study, 7.2% demonstrated screw loosening.[5] In our case, the broken screw was likely inadequately placed and tightened along the mandible, rendering the implant less firmly fixed over time. Chronic mechanical manipulation, associated with mastication for example, may also possibly result in screw loosening.[3,6] Thus, in the instance where our broken screw was not appropriately tightened along the implant–mandibular interface, there is greater risk of progressive screw loosening and reduced mandibular mechanical strength, contributing to the resulting mandibular fracture.

Overall, PEEK implants are reliable, chemically inert, and biocompatible. They can be custom-made to treat complex, three-dimensional conditions such as HFM through computer-assisted manufacturing.[7] However, our patient presented with a hardware infection 5 weeks postoperatively following PEEK implant placement. Alonso-Rodriguez et al have shown that infection is a major complication affecting approximately 14% of patients who get custom PEEK implants for craniofacial deformities.[8] Additionally, one study demonstrated that when compared with several neurosurgically-implanted materials, PEEK implants presented with one of the highest infection rates (7.13%).[9] Olate et al reported a 22.2% postoperative infection rate for patients who underwent mandibular angle augmentation with custom PEEK implants.[10] Orthognathic surgery is also considered as a clean contaminated procedure; consequently, these procedures are more inherently prone to infection compared with those that are non-contaminated.[11] In our patient, the hardware infection itself and subsequent removal of the PEEK implant both likely further contributed to reduced mechanical strength of the mandible, leading to increased susceptibility of the mandibular fracture.

Surgical manipulation of the hypoplastic mandible with hardware placement should also be considered as a contributing factor to fracture development. Raghoebar et al explained that multiple implant fixation sites can diminish the mechanical strength of the mandibular bone, at least temporarily.[12] Consequently, caution must be undertaken when surgically handling a thin mandible. Although this patient's custom-designed PEEK implant had excellent stabilization when fixated, the lateral right-most screw had broken inside the bone. When the implant became infected, CT imaging showed bony lucency of the right parasymphyseal region where this screw had broken. This same region of diminished bone density of the mandible had later fractured under traumatic forces. Thus, decreased bone density of the right parasymphyseal mandible associated with prior implant screw fixation likely made the mandible more fragile and prone to injury. Like in elderly patients with atrophic edentulous mandibles, high stress loads on a mandible with reduced bony density can ultimately lead to fracture.[13–15] Our patient had a focal area of weakness in the mandible from surgical manipulation, which was predisposed to fracture from traumatic high tensile forces in that particular anatomical point. As aforementioned, hardware infection itself and then removal of the infected implant likely reduced the structural support and stability of the mandible, increasing the susceptibility to injury from a traumatic event.

It is important to consider the traumatic mechanism resulting in the fracture. A rollover golf cart accident presents as a significant mechanism of injury that can lead to facial trauma in any patient, regardless of predisposing factors that reduce mandibular mechanical strength. For our patient, the underlying biomechanics and suboptimal stability of the hypoplastic mandible following hardware failure and implant infection increased her susceptibility to fracture.

PEEK implants by themselves do not completely resolve facial asymmetry and often require concomitant autologous fat transfer for fine cosmetic refinements, as seen in our patient.[7,16] Nevertheless, PEEK implants are a viable alternative source to autogenous bone grafting for mandibular reconstruction and contouring in HFM. Our patient had initially successful results with PEEK implant mandibular augmentation, but unfortunately endured hardware infection and subsequent mandibular fracture. Any of these sequential factors – broken implant screw decreasing mandibular mechanical strength, infection and diminished bone density in that region, reduced structural support after removal of the implant, and surgical handling of thin mandibular bone – may have made our patient more prone to a mandibular injury beyond just the traumatic mechanism. Hence, significant care must be undertaken when surgically handling thin, hypoplastic mandibular bone, and postoperative complications must be managed immediately to prevent further compromise of mandibular bone integrity in patients with HFM.

Products/Devices/Drugs

None.

Authors' Contributions

K.A. was involved in conception and design, acquisition of data, analysis and interpretation of case, and writing manuscript. R.P.D. contributed with design, acquisition of data, analysis and interpretation of case, and writing manuscript. R.S.M. was involved in conception and design, acquisition of data, analysis and interpretation of case, writing manuscript, and in final edit of manuscript.

Ethical Approval

Informed consent was obtained from the patient for surgical management.

Patient Consent

Informed consent was obtained from the patient for surgical management.

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None.

Conflict of Interest

None declared.

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