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Pediatric cranioplasty: lessons from a 1940s neurosurgical procedure☆

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ABSTRACT

No clear consensus exists regarding the best material and technique for use in pediatric cranioplasty. The immature bone in pediatric patients poses several challenges, such as rapid growth, skull growth restriction, plate migration, and tissue erosion. We present a pediatric cranioplasty from the 1940s, and examine the characteristics of the plating and anchoring used.

The patient suffered a skull fracture after falling from a swing in 1946 at age six, requiring cranioplasty. Sixty-seven years later, she noted drainage from the old incision site, requiring reoperation. During surgery, the skull appeared normally shaped, and the area under the plate exhibited complete bone growth. The plate was noted to have several innovative design features that contributed to this outcome, notably that the plate and its anchors were “semi-rigid”; the hardware was softer than surrounding bone and easily pliable, deforming to accommodate skull growth and prevent restriction. The structure of the plate was also such that it allowed growth of underlying bone tissue to close the defect.

This case contains unique features that can foster discussion regarding plate design and surgical technique that might avoid traditional complications.

Introduction

How to best reconstruct or reattach the skull in pediatric neurosurgery remains a subject of debate among neurosurgeons. One aspect of the debate is the circumstances in which one should utilize standard plates, sutures, or resorbable plates. Another aspect involves which material to use in the event an autograft or customized implant is not available. Throughout history, pediatric cranioplasty has constantly evolved. Major issues are incomplete skull growth at the time of operation, rapid growth of the skull following the procedure, increased metabolic demand of bone tissue, and the requirement for the cranioplasty repair to have increased endurance (so as to last the length of the patient’s life) [1]. These factors lead to concerns about potential complications such as skull growth restriction, alteration of normal skull anatomy, tissue inflammation, foreign body reaction, and extrusion erosion and migration of implanted hardware [1,5]. To address these considerations, varying techniques have been developed, and multiple materials have been proposed, including metals such as steel and titanium, acrylics, autologous bone grafts, hydroxyapatite cement, organic polymers, or hybrid mixtures [4]. Here we present an interesting case of a patient who suffered wound breakdown sixty-seven years after receiving cranioplasty due to a traumatic skull fracture at age six. We then review the history of pediatric cranial implants.

Case report

History and examination

The patient is a 73 year old female with Parkinson's disease who in 1946, at the age of 6 years, fell off a swing and sustained a right frontoparietal skull fracture, requiring repair of the cranial defect. She presented to our care when she noted intermittent episodes of clear fluid trickling down her right face. She denied any fever, headache, or pain, and no progressive neurologic complaints. Physical exam revealed a 2 cm area of wound breakdown along her old incision, through which milky white discharge was expressed.

Operation and pathologic findings

Contrast CT of the head was grossly limited by artifact (Fig. 1A). Scout films demonstrated the underlying plate (Fig. 1B). Removal of the right frontal cranial plate and scalp wound reconstruction was planned. At the time of surgery, the plate was removed using a Penfield 1, after folding back two of the tabs anchoring the bone plate and using a drill to remove the posterior one, which had straightened over time with skull growth. (Fig. 2A and B). Bone growth was noted underneath the entire area, and no additional cranial repair was required. The scalp defect was closed primarily. Cultures were positive for *Helcococcus kunzii*, a gram-positive

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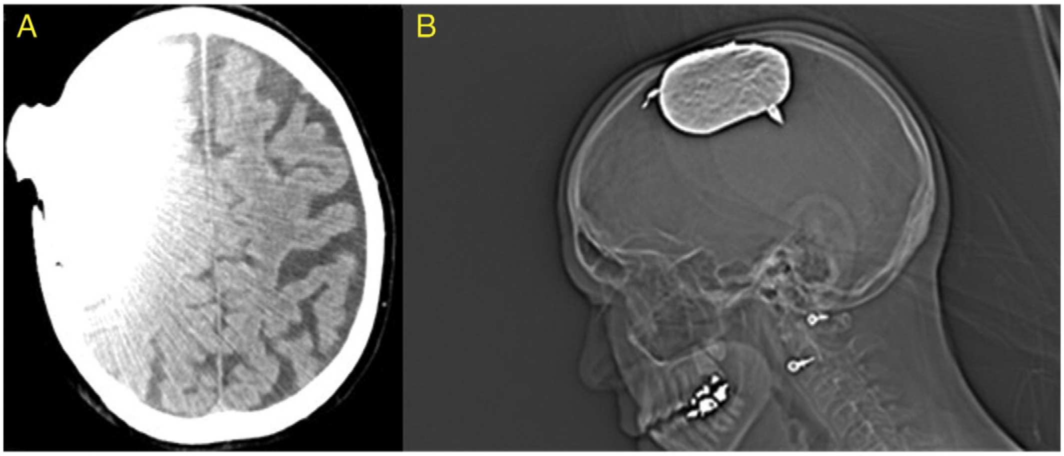

Fig. 1. A. A pre-op CT scan of the head revealed significant metallic artifact, limiting initial evaluation of the tissue and plate. B. Scout film showing the location of the metallic plate, along with some of its structure. Two of the tabs holding the plate in place are visible.

cocci, 2+ *Finegoldia magna*, and 1+ *Propionibacterium Acnes*. All bacteria were sensitive to Levaquin.

Discussion

We present a pediatric cranioplasty performed sixty-seven years ago. The surgical technique employed resulted in significant bone growth beneath the plate and normal skull development. The success of the cranial plate lies in its simple design. The anchors on the plate were designed such that they bent as the skull grew, minimizing skull growth restriction. Furthermore, the skull itself was stronger than the metal used for construction of the plate and anchors; this allowed the implanted hardware to deform, accommodating normal skull growth. Finally, the plate's thickness mimicked the thickness of the outer table of the skull, allowing underlying cancellous bone and diploe to continue to grow beneath the plate, and eventually fill in the cranial defect (Fig. 3).

There is a paucity of data regarding the best material and technique for use in pediatric cranioplasty. The technique used must account for the immature state of the pediatric skull, and the necessity that any implants last the patient's lifetime.

The earliest materials used for pediatric cranioplasty were stainless steel bone wire [4]. This wiring was inexpensive and resistant to infection. However, it did not allow for 3D contouring during reconstruction, often caused erosion, and failed to fix bone in the desired position. Rigid metal plate fixation prevented these issues, allowing surgeons to sculpt the plate to fit the shape of the skull, and providing better long term fixation and improved protection of underlying tissues [1]. Early plates were composed of lead, platinum, iron, nickel, or chromium alloys, which had risk of corrosion and toxicity, or in the case of platinum, was prohibitively expensive [9]. Due to these concerns, those metals were replaced with molybdenum, cobalt, vitallium, and tantalum [4,9]. Vitallium was very common in the 1940s, but eventually discontinued in favor of tantalum, as contouring was difficult [9]. Tantalum gained favor during World War II for use in repairing large cranial defects sustained during battle. It's advantages were that it was inert, had limited tissue inflammation, and was easily malleable and corrosion-resistant [9]. Tantalum and stainless steel, however, are radio-opaque and thermo-conductive, interfering with imaging and causing head pain in heat and cold [4,9]. Eventually, both were replaced by titanium, which has excellent tensile strength and pliability, and limited effect on imaging studies [4,11]. Interestingly, titanium became industrially available in the United States in 1946, the same year our patient underwent skull plate fixation, although titanium cranioplasty wasn't documented in literature until 1965 [2,9,10].

Screws used in plate fixation also had issues. They caused osteolysis at the site and migrated out of bone, resulting in plate migration and/or causing erosion [1,4,5,9]. Paradoxically, due to the apical growth pattern of pediatric bone (that is, new bone is laid down

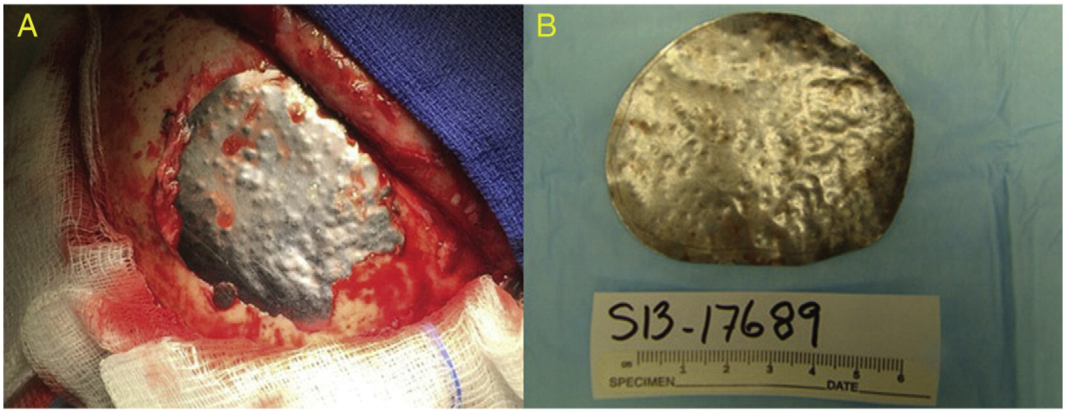

Fig. 2. A. Intra-op photograph of skull plate (two of the placement tabs are visible). The posterior tab was visibly deformed. B. Gross examination of the plate. The metal composition of the plate was not known, but it was soft and pliable, of similar hardness to the metal in a soda can.

Fig. 3. Simplified schematic of how the plate was set into the skull. Black lines represent the metal plate. Gray lines represent the patient's skull. Thick gray lines represent new bone growth beneath the plate at time of operation. Image A depicts the plate as it was initially implanted. The black lines represent the plate covering up the cranial defect in the skull (thin gray lines). The lower image B shows the plate just prior to removal. The black arrow points to where the posterior plate anchor was deformed by new bone growth (dark gray), getting forced upward into a vertical position.

near the galeal surface, and resorbed in deeper layers), plates/screws could also gradually be forced deeper into the calvarium, undergoing passive intraosseous translocation, and winding up adjacent to the dura and underlying brain tissue [4,5].

Studies show that infection of metallic plates is associated with placement near the nasal cavity or sinuses, and is also associated with recent head infections [12]. Our patient did not reports these complaints, though she had seborrhea of the skull which predisposed her to issue. We hypothesize that our patient displayed the phenomenon of osteolysis at anchor sites, leading to superficial tissue erosion. Once a small amount of erosion occurred, bacteria likely entered and colonized the plate. *Propionibacterium Acnes* is a well-recognized type of infection, usually affecting shunts [6]. *H. kunzii* has been previously implicated in superficial skin infections, primarily in the setting of diabetes mellitus [3,7]. *F. magna* is an anaerobe that has been implicated in a wide range of infections, such as skin infections, pneumonia, infectious endocarditis, prosthetic joint infections, and meningitis. It was recently described as an under-estimated pathogen, and genetic analysis has revealed that it possesses numerous virulence factors, granting it significant pathologic potential [8].

However, the interval in which this patient presented with this complication far exceeds most previously reported cases. Wiggins et al described the median interval from cranioplasty to removal of a graft due to infection as 120 days, with almost all cases presenting within 8 months of surgery [12]. Our patient's interval was 67 years. Goodrich, Sandler, and Tepper described a similar case to ours in which a patient had a metallic plate placed due to childhood trauma, and presented with skin erosion and infection 30 years later [4]. In that patient, the metallic plate had failed to undergo osseous integration and had mobilized, becoming what the authors referred to as a "free-floating unit" [4]. However, it should be noted that our patient's case is somewhat different from this previously described case in that our patient's skull plate had not mobilized, and had remained effectively fixed in its original location. Furthermore, the other described patient had a hybrid metallic plate composed of titanium mesh mixed with another component; early titanium implants were known to have higher incidence of osteolysis and superficial tissue erosion than the non-titanium metal fixation plates utilized in our patient [4].

Berryhill et al described the fears some physicians have of the potential for rigid plate fixation to cause growth restriction and other skull defects if utilized in pediatric patients. However, they also describe bone growth in the pediatric calvarium as being very fluid, and arises from multiple foci, including mesenchymal, embryonic, and periosteal sources [1]. In their opinion, this multifocal growth pattern makes it unlikely that rigid plate fixation could significantly interfere with the normal process of pediatric skull development [1]. They offer as support studies in Rhesus monkeys conducted by Yaremchuk et al, which indicated that there was no significant difference in skull deformities between monkeys receiving simple wire fixation, rigid plate fixation, and "extensive (excessive) fixation" [13]. Our patient supports the notion that at least semi-rigid plate fixation does not cause significant growth restriction in pediatric patients.

Conclusion

In conclusion, this case is interesting because it demonstrates a semi-rigid fixation strategy for pediatric cranioplasty. Because the growing bone was stronger than the hinges embedded in the surrounding area, as the skull grew, the hinges deformed along with it, allowing for normal development. The plate was as thin as a child's outer table, allowing for growth of underlying bone tissue, effectively filling in the defect underneath the plate. Whether these strategies would be as effective in younger patients remains to be seen. Furthermore, the material used here-stainless steel and/or alloy-obviously is suboptimal as it interference with neuroimaging. A new material used in employing this technique must still mimic the thickness, tensile strength, and inert properties of the original material.

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