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CASE-BASED UPDATE

Infantile cranial fasciitis: case-based review and operative technique

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Abstract

Background Cranial fasciitis (CF) is an uncommon benign primary lesion of the skull that typically affects the pediatric age group. Due to the rarity of CF, no prospective studies exist. Earliest description of this condition dates to 1980. The limited scientific and clinical literature regarding CF is dominated by case reports. For these reasons, questions pertaining to the true incidence, genetic risk factors, prognosis, and long-term outcome remain unanswered.

Discussion Clinically, CF presents as a firm, painless, growing scalp mass that is typically not considered in the differential diagnosis. Preoperative pathognomonic signs and symptoms are absent, and imaging features are often nonspecific. Treatment is typically through complete surgical resection, at which time histopathological examination confirms the diagnosis of CF. Reconstruction of the skull defect in the child is critical. Autograft techniques help maintain a rigid construct that integrates with the native skull while preserving its continued ability to grow. Generally, a good outcome is observed with complete resection.

Exemplary case We report a case of CF in an infant with emphasis on operative nuances and early follow-up results.

Conclusion CF is a rare fibroproliferative disease that has a poorly defined incidence and long-term follow-up. Due to its locally invasive nature and nonspecific presentation, CF is often difficult to differentiate from malignancies and infections. Complete surgical resection is the best approach for diagnosis and cure. Its occult clinical presentation often allows it to achieve considerable growth, leaving a sizeable skull defect following resection. Since CF presents in the pediatric population, allograft reconstruction is preferred over titanium mesh or other synthetic materials to allow osseous integration and continued uninterrupted skull growth.

Keywords Cranial fasciitis · Fibroproliferative · Primary skull lesion · Rib resection

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Introduction and historical background

Cranial fasciitis (CF) is a rare, benign fibroproliferative lesion of the skull that typically presents in young children as a firm, painless, growing scalp mass. Earliest description of CF dates to 1980 in a report by Lauer et al. [32]. Radiographically, it often presents as a soft tissue lytic mass eroding through the skull. It may appear isointense on T1 magnetic resonance imaging (MRI) with peripheral contrast enhancement and demonstrates a hyperintense core and an isointense periphery on T2 MRI [27]. This typical radiographic description is not always encountered [57], making the initial diagnosis challenging. CF can present with ambiguous radiographic findings that can mimic neoplastic and infectious conditions [59]. CF commonly occurs spontaneously, but may also arise following trauma [27] or whole-brain radiation [8, 36, 60]. While intralesional steroid injection has been described in a patient

had no bone involvement [34], complete surgical resection provides a definitive diagnosis and cure.

To further investigate this entity, we conducted a PubMed literature search for all CF cases and were able to identify only 54 articles [1–9, 12–18, 20–34, 36–42, 44–46, 48–55, 57–61]. The majority of those articles were case reports (Table 1). A total number of 76 patients from 1980 to 2016 were reported in the literature that we reviewed. Average age of all the patients was 6 years, with a range of 1 month to 61 years. Male to female ratio was approximately 2 to 1 (Fig. 1). The most common site of CF lesion was the temporal region (26%), followed by the parietal (20%), occipital (14%), and frontal (13%) (Fig. 1). There are no large studies describing CF's natural history, incidence, recurrence rates, or long-term follow-up after treatment. In this paper, we conduct a case-based review of CF and discuss a clinical case that was treated with complete resection with a focus on the technical intraoperative nuances and follow-up.

Clinical presentation

CF is a commonly misdiagnosed primary skull mass that can mimic conditions such as malignancies and infection (e.g., dermoid, epidermoid, Langerhans cell histiocytosis, tuberculosis). Due to its rare occurrence, it is not usually considered in the differential diagnosis of primary skull lesions. No clear natural history was present in the literature; however, several reports have shown that the disease can progress to a significant size and cause brain compression [2, 17, 42, 49, 55].

For multiple reasons, delayed diagnosis of CF is common. Because CF is painless, children often ignore it until it becomes relatively large and noticeable to the parents. Furthermore, because the growth rate is slow, the lesion may grow to a substantial size, over a significant amount of time, prior to causing any neurological symptoms.

Diagnosis

The imaging characteristics of CF have been described as isointense on T1 MRI with peripheral contrast enhancement. On T2 MRI, CF demonstrates a hyperintense core and an isointense periphery on T2 MRI [27]. The imaging characteristics noted in our case did not match the proposed description in the literature. The lesion in our case was diffusely isointense on T1 and T2 MRI. It also demonstrated diffuse postcontrast enhancement on T1-weighted sequences. This mismatch between the reported classic imaging characteristics [27] was also noticed in other reports [57], corroborating the impression that there are no reliable radiologic imaging findings that are distinctly diagnostic for CF.

Management

Surgical resection remains the definitive way to diagnose and treat this condition. The planning phase of surgery begins with patient positioning. The lateral decubitus position allows simultaneous access to a rib bone donor site and the lesion. If the location of the lesion is in the occipital area, then the prone position may be a preferred position to access the donor rib. The periosteum should be carefully preserved and gently elevated from the bone while keeping in mind the location of the neurovascular bundle below the inferior border of the rib. In our experience, once the periosteum is preserved and sutured back, the rib will regrow within 4–12 months (Fig. 2e, f). Depending on the surgeon's preference, the rib may be morselized or eburnated and plated to the healthy skull edges. The advantage of plating large pieces of eburnated rib is that it provides a strong scaffold when compared to morselized bone.

During the lesionectomy procedure, it is important to keep in mind that the dura mater has osteogenic capability and therefore should be preserved whenever possible [19, 43]. Involved segments of the dura should be carefully inspected, excised, and repaired appropriately using synthetic dura or fascial grafts. In some instances, the outer dural leaflet can be split under the operative microscope. If the inner dural leaflet is not involved, it should be preserved. In such case, no further dural repair is necessary.

Large skull defects in infants can be reconstructed with harvested bone from rib or split calvarial bone grafts. Unlike adults, where continued skull growth is not a consideration, cranial defects in very young children are preferably not corrected with synthetic implants such as titanium mesh, polyether ether ketone (PEEK), or polymethyl methacrylate (PMMA) when possible. Studies have shown that shortly after birth, the human skull grows rapidly, reaching 90 % of its final size by the age of 7 years, and slows down after puberty [10, 35]. Therefore, in this age group, autograft is usually a good biocompatible option to reconstruct the skull and preserve its ability to grow unconstrained [11, 47, 56]. During lesionectomy, valuable bone dust should be carefully collected and used at the end of the reconstruction procedure.

Similar to the skull, rib grafts provide a tripartite structure consisting of a rigid inner and outer cortical bone separated by a softer inner cancellous bone. The rib's natural curvature makes it easy to incorporate into large skull defects while maintaining a low-profile construct level with the skull surface (Fig. 2c, d). The ability of ribs to regenerate makes them excellent autologous donor bone candidates. Disadvantages include performing an extra incision, postoperative pain, injury to the neurovascular bundle, and potential for pulmonary complications such as pneumothorax. Split calvarial bone

Table 1 Summary of the "reviewed literature depicting authors, year that the manuscript was published, type of the study, number of CF patients with in the study, patient's average age and age range, gender ratio, and location of the lesion as mentioned in the study

| Authors	Year	Study type	Number of patients	Mean age [range] (years)	Gender ratio (F/M)	Location of the lesion(s)
Alder	1986	Case report	1	1.7	0/1	Frontal
Agozzino	2006	Case report	1	8.0	1/0	Temporal
Barohn	1980	Case series	1	0.1	0/1	Frontoparietal
Boddie	1997	Case report	1	2.5	1/0	Parietal
Clapp	1997	Case report	1	0.1	0/1	Temporal
Coates	1990	Case report	1	3.4	0/1	Frontal
Curtin	2014	Case report	1	2.0	1/0	Temporal
Delfini	2004	Case report	1	11.0	0/1	Parietal
Du Toit	2009	Case report	1	0.0	0/1	Frontonasal
Fissenden	2014	Case report	1	1.17	0/1	Temporal
Foureur	2002	Case report	1	0.3	0/1	Parietal
Gonzalez-Garcia	2013	Case report	1	4.0	0/1	Temporal
Govender	2001	Case report	1	2.0	0/1	Frontoparietal
Halder	2012	Case report	1	10.0	1/0	Frontotemporal
Hattab	2014	Case report	1	2.7	1/0	Parietal
Hoeffel	1993	Case report	1	6	1/0	Temporal
Hoya	1996	Case report	1	1.2	0/1	Frontotemporal
Hunter	1993	Case report	1	0.8	0/1	Occipital
Hussein	2008	Case report	1	2.0	0/1	Occipital
Imafuku	2011	Case report	1	5.0	0/1	Frontal
Inamura	1991	Case report	1	2.0	0/1	Occipital
Iqbal	1995	Case report	1	5.0	1/0	Occipital
Johnson	2008	Case series	4	3 [0.3–11]	1/3	Frontal (1)–parietal (3)
Keyserling	2003	Case report	1	0.6	0/1	Parietotemporal
Kumon	1992	Case report	1	5.0	0/1	Frontal
Lang	1996	Case report	1	0.3	0/1	Frontoorbital
Larralde	2003	Case report	1	0.2	0/1	Temporal
Lauer	1980	Case series	9	1.9 [0.1–6]	3/6	Temporal (3), occipital (1), parietal (1), frontal (1), temporoparietal (1),temporoparietal (frontoparietal (1), parietooccipital (1)
Lecavalier	2014	Case report	1	4.0	0/1	Temporal
Lee	2004	Case report	1	3.0	1/0	Occipital
Longatti	2004	Case report	1	11.0	0/1	Parietal
Marciano	1999	Case report	1	32.0	0/1	Temporal
Marshall	2009	Case report	1	0.8	0/1	Temporal
Martinez-Lage	1997	Case report	1	6.0	0/1	Frontoparietal
Mollejo	1990	Case report	1	11.0	1/0	Occipital
Noguchi	1999	Case report	1	0.1	0/1	Frontoparietal
Oh	2007	Case report	1	0.3	0/1	Temporal
Pagenstecher	1995	Case report	1	7.0	1/0	Parietal
Pasquier	1984	Case report	1	0.0	0/1	Parietal
Patterson	1989	Case report	2	5 [3–7]	1/1	Unknown (1), temporoparietal (1)
Rapana	2002	Case report	1	45.0	0/1	Frontal
Ringsted	1986	Case report	1	6.0	0/1	Parietal
Sajben	1999	Case report	1	1.5	0/1	Frontal (1)
Sarangarajan	1999	Retrospective study	4	4.6 [1–14]	1/3	Frontal (1)–parietal (1)–temporal (1)–temporoparietal (1)
Sato	1993	Case report	1	7.0	0/1	Temporal
Sayama	1995	Case report	1	0.8	0/1	Frontotemporal
Summers	2007	Case report	2	38.5 [16–61]	1/1	Occipital
Takeda	2008	Case report	1	3.0	1/0	Temporooccipital
Wagner	2016	Case report	1	0.1	0/1	Parietal
Wu	2013	Case report	1	13.0	1/0	Occipital
Garza	2012	Case report	1	2.0	1/0	Temporal
Rakheja D	2008	Genetic study	7	2.8 [0.3–6.3]	3/4	Unknown
Santacruz L	2007	Case report	1	27.0	1/0	Temporal
Yebenes M	2007	Case report	1	8.0	0/1	Occipital
		Total:	76	6	24/52	

F female, *M* male

Fig. 1 Blue pie chart showing the gender ratio (*upper right corner*) and a color-coded pie chart showing the common locations of the reported CF lesions in the reviewed literature (*center*)
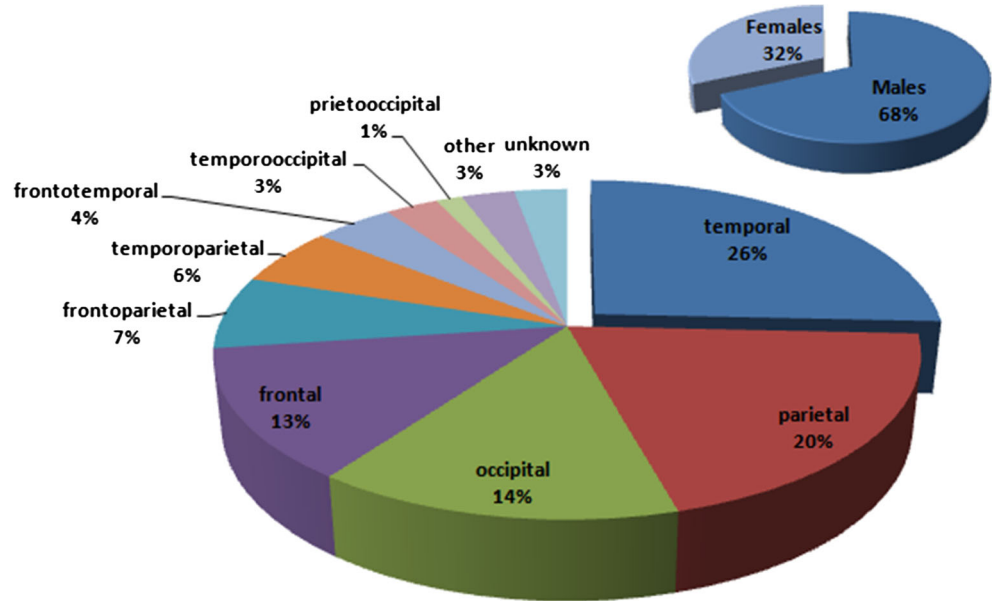
grafts are also excellent candidates to reconstruct large skull defects. Their round and malleable profile provide the ideal

contour for the skull. Split calvarial grafts help avoid the morbidity associated with extracranial bone harvesting at the
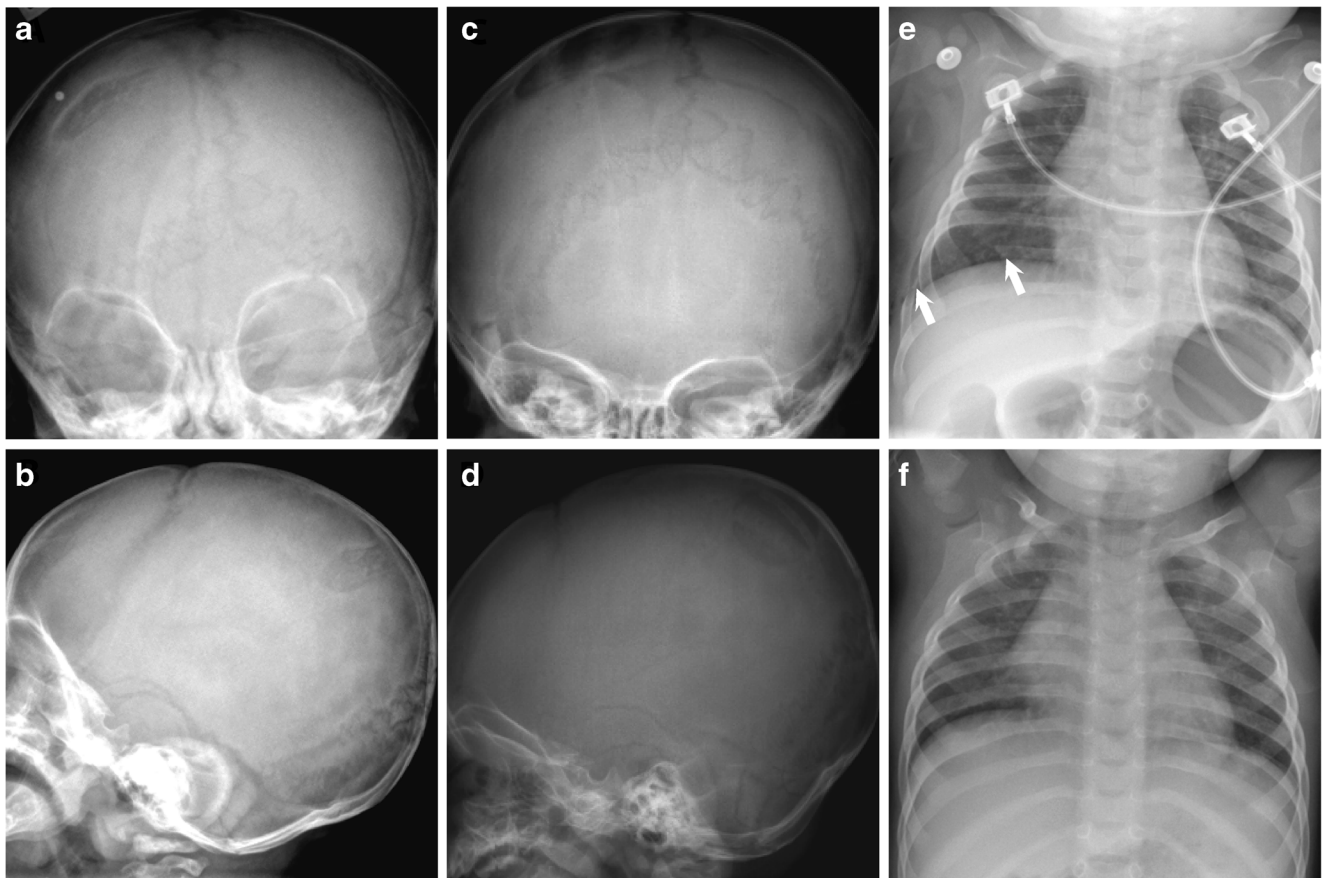
Fig. 2 **a**, **b** Preoperative anteroposterior and lateral skull radiographs demonstrating a right parietal lytic lesion with sclerotic border. **c**, **d** Postoperative anteroposterior and lateral skull radiographs demonstrating resection of the mass and rib allograft cranioplasty. **e** Immediate postoperative chest radiograph showing absent eighth rib on the right side (*arrows* depict the location of the harvested rib). **f** Four-month follow-up X-rays showing complete regeneration of the rib

Fig. 3 Preoperative computed tomography (CT) imaging. **a** Compiled axial slices 1.5 mm in thickness apart showing the lytic lesion (cranial fasciitis) involving the inner and outer tables of the parietal bone with associated overlying soft tissue swelling. **b** Three-dimensional volume rendering of the CT scan using Slicer 4.6.2 software
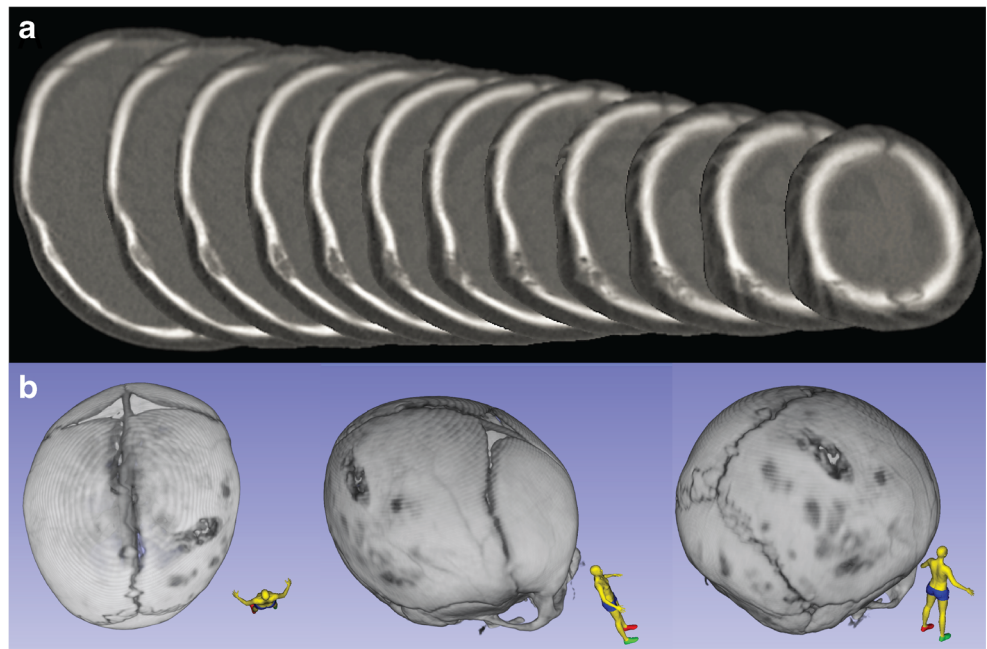

expense of larger cranial incisions to incorporate a large surface area of intact skull. This makes them ideal for large craniosynostosis operations to a greater extent than smaller lesionectomy procedures in infants. In summary, we believe that rib and split thickness calvarial grafts are equivalent and choosing one over the other ultimately depends on the surgeon's preference.

Fig. 4 **a** Axial T2, **b** saggital T1, **c** axial, and **d** saggital postgadolinium T1 MRI showing a solid mass involving the right parietal skull
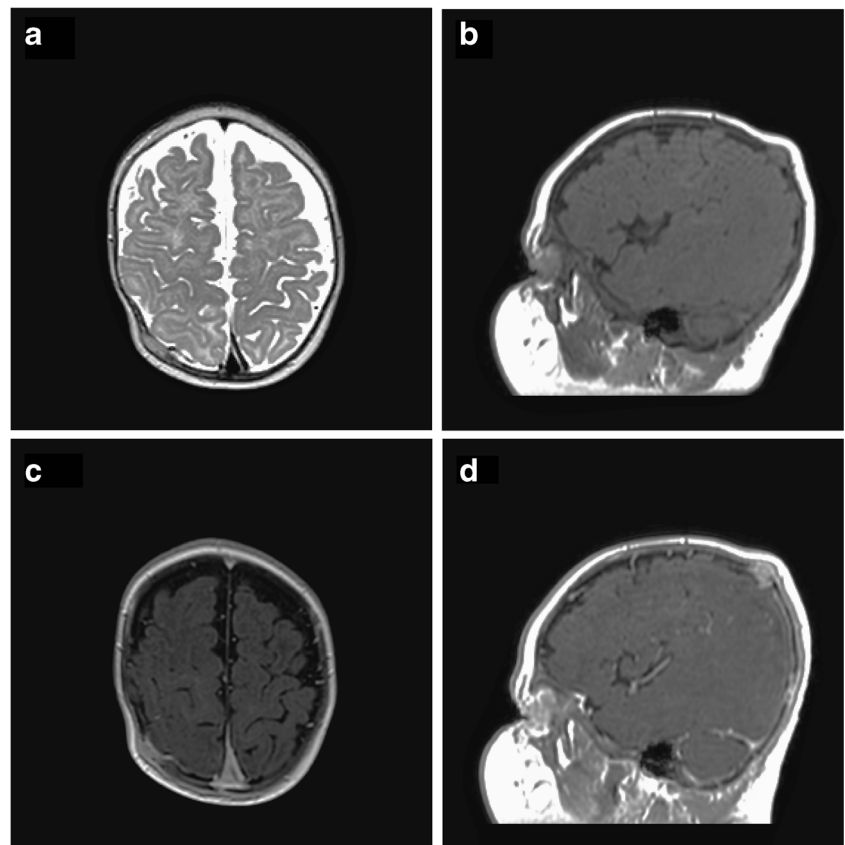
Fig. 5 Intraoperative pictures showing patient positioning (**a**) and preresection and (**b**) postresection (**c**) of the lesion
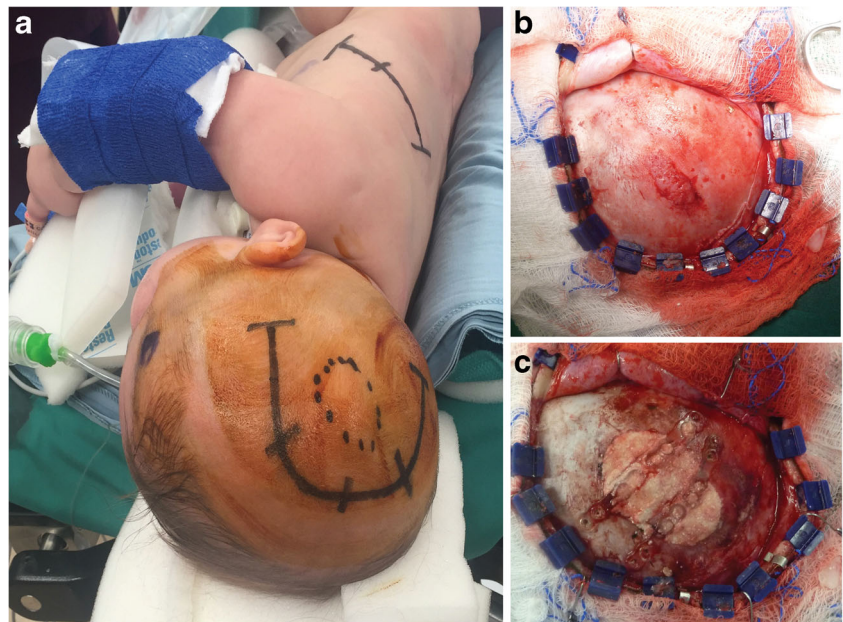
Prognosis and outcome

There are many questions pertaining to CF that remain unanswered. Further larger studies can aid in elucidating the true incidence, genetic risk factors, prognosis, and long-term outcome of this rare condition.

Exemplary case presentation

Clinical history and examination

A 6-month-old boy presented to a local physician with a slowly growing painless parietal scalp mass. The child was born full term via a nontraumatic normal vaginal delivery after an uncomplicated intrauterine course. The child had no history of head trauma. Neurologically, the child was normal. Head examination revealed a right parietal firm immobile lesion measuring approximately 3 cm in largest diameter. Overlying skin and scalp hair appeared normal.

Imaging

Skull radiographs demonstrated a parietal lytic lesion a few centimeters lateral to the midline with sharp sclerotic borders (Fig. 2a, b). Computed tomography (CT) scan of the head showed a right parietal soft tissue abnormality measuring 3 cm and eroding through the inner and outer tables of the skull (Fig. 3). MRI redemonstrated the soft tissue lesion previously described on CT. The lesion was isointense on T1 and T2 MRI and enhanced with administration of intravenous gadolinium (Fig. 4).

Operative technique

Positioning The patient was positioned in the left lateral decubitus position to allow simultaneous lesionectomy and rib harvesting. The head was rested on a cerebellar headrest. An inverted U-shaped incision was outlined to incorporate the palpable lesion. A diagonal straight incision overlying the ninth rib was outlined (Fig. 5).

Lesion resection Scalp incision was made as outlined (Fig. 5a, b). The periosteum was then incised and reflected along with the scalp flap inferiorly. A portion of the periosteum that was partially adherent to the lesion was excised and sent for frozen section, which showed a spindle cell proliferation. A grayish tumor eroding through the outer cortical bone was identified (Fig. 5b). A high-speed electrical drill was used to fashion a small craniotomy around the tumor maintaining a grossly clean margin of 5 mm. All of the bone dust was saved for later use. The outer layer of the dura was removed with the skull mass and sent en bloc for histopathological examination. The inner layer of the dura appeared to be grossly free of abnormal tissue, and bipolar cautery was utilized to achieve hemostasis.

Rib harvesting The chest incision was made with a no. 15 blade scalpel parallel to the underlying eighth rib. Blunt dissection was performed to spread the muscle layers, and bipolar cautery was used to dissect down through the muscle attachments to the ribs. The periosteum of the eighth rib was then incised sharply, and straight curettes and no. 1 Penfield instrument were used to dissect the periosteum over and around the rib. A semi-sharp was

Fig. 6 Schematic of rib resection technique. After exposing the desired rib (**a**), the periosteum is incised sharply and the underlying bone is exposed (**b**). The periosteum is reflected off the bone using a Penfield no. 1 instrument and straight curettes (**c**). A bone cutter is introduced after circumferential dissection of the periosteum away from the cortical surface of the rib (**d**)
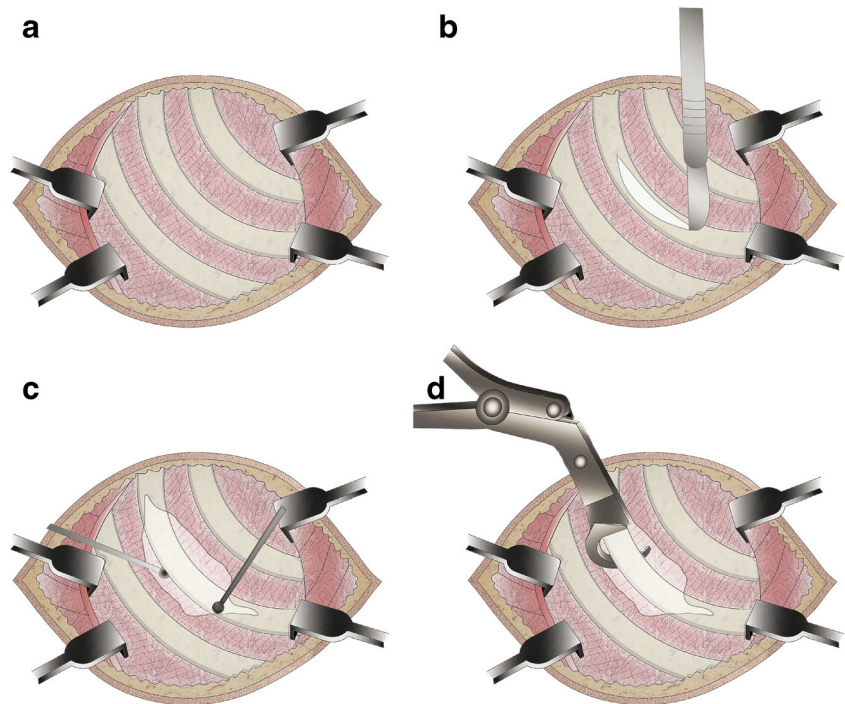
then used to complete circumferential dissection of the periosteum off the surface of the exposed rib. A rib cutter was then used to incise 6 cm of rib (Fig. 6). A Valsalva maneuver was performed and no air leak was noted. A Marcaine nerve block was infiltrated proximally at the T7, T8, and T9 ribs for postoperative pain control. The periosteum was reapproximated with absorbable sutures. The muscle fascia and the dermis were closed in layers. The harvested rib was set aside for later use.

Cranial reconstruction and autologous rib cranioplasty
The harvested rib was eburnated using an electric drill. It was then cut in half and plated over the defect using a

tissue-compatible absorbable osteosynthesis plating system consisting of amorphous pure poly(D,L-lactide) acid (PDLLA) polymer (Fig. 5c). The construct was solid. Approximately 3.5 cm^3 of bone putty containing demineralized bone matrix and bone chips combined with the harvested bone dust was placed in the empty spaces (Fig. 5b).

Pathology

Histopathologic examination of the lesion demonstrated a fairly uniform lesion composed of loosely arranged, broad fascicles of spindle cells. The background was variably edematous/
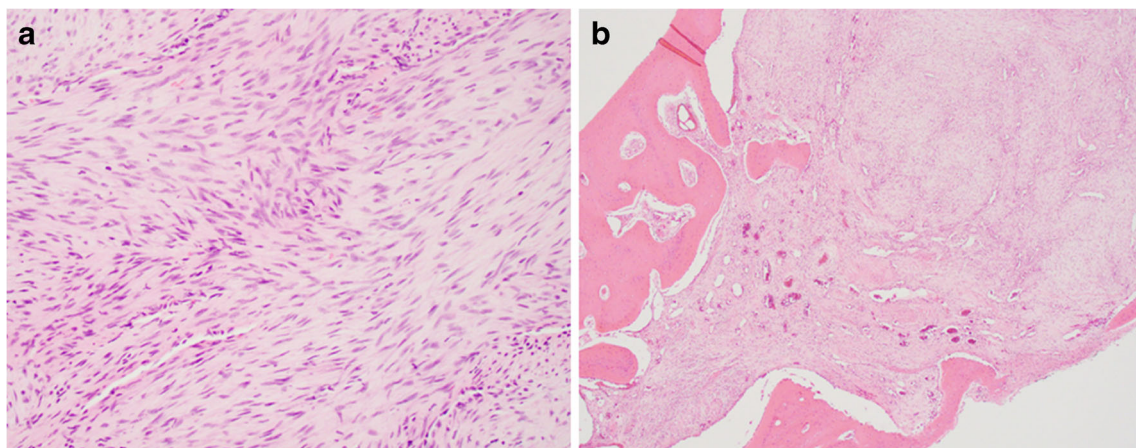
Fig. 7 Histopathologic examination of the specimen showed a spindle cell lesion with occasional mitotic figures. **a** H&E stain, ×200 with prominent infiltration through the bone. **b** H&E stain, ×40

Fig. 8 Four-month follow-up CT scan showing early bone and cross bridge formation in the axial (**a**), sagittal (**b**), and coronal planes (**c**). Three-dimensional reconstruction of the CT scan showing new bone formation (*P* posterior, *A* anterior, *L* left). Volume rendering was made using Slicer 4.6.2 software). *Arrows* denote the areas where new bone was noted around the construct (**d**)
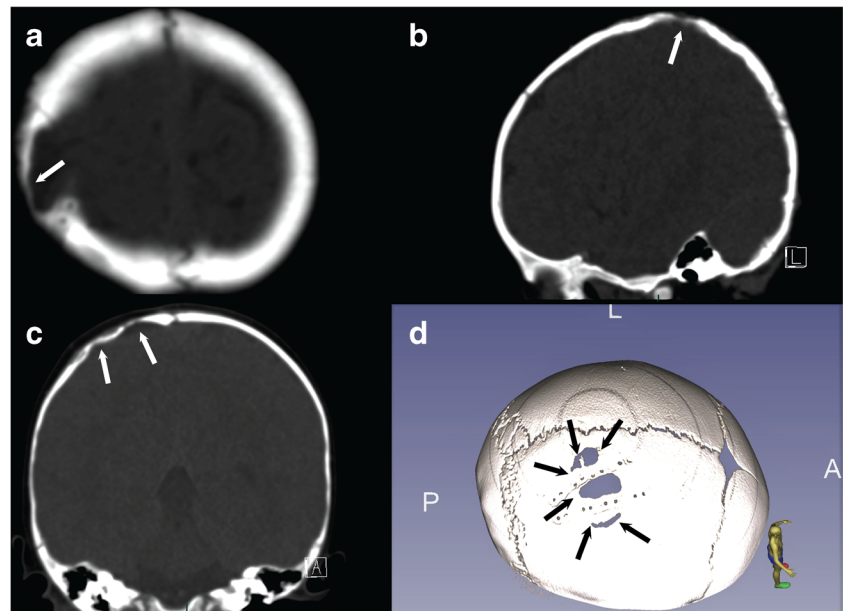
myxoid with focal areas of increased collagen. The spindle cells showed minimal nuclear atypia; however, mitotic activity was present. The spindle cell proliferation extensively involves the skull bone (Fig. 7a, b). Immunohistochemical staining showed the cells to express vimentin and smooth muscle-specific actin, while the cells were negative for desmin, muscle-specific actin, and CD34. The morphologic features and immunophenotype in this clinical setting are characteristics for cranial fasciitis.

Clinical outcome

The patient tolerated the surgery well with no complications. Operative time was 2 h and 15 min. Estimated blood loss was 5 ml. A plain chest X-ray (CXR) performed postoperatively did not demonstrate any pneumothorax and showed the area of rib resection (Fig. 2e). Postoperative radiographs at the time showed stable skull construct (Fig. 2c, d). Follow-up CXR at 4 months showed complete regeneration of the rib in the chest region (Fig. 2f). A CT scan of the head showed that the rib grafts had integrated into the skull and new bony cross-bridge formations were noted 4 months after the procedure (Fig. 8).

Conclusion

Due to its locally invasive nature, cranial fasciitis is often difficult to differentiate from malignancies and infections. Its nonspecific clinical and radiographic presentation makes complete surgical resection a necessary avenue for diagnosis and cure. Some large lesions can leave a sizeable skull defect behind warranting cranial reconstruction. Allograft reconstruction is superior to titanium mesh or other synthetic materials as cranial fasciitis is a disease of childhood and often presents while the skull continues to grow.

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