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SPECIAL TOPIC

Pediatric/Craniofacial

30-year International Pediatric Craniofacial Surgery Partnership: Evolution from the "Third World" Forward

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Background: Craniofacial diseases constitute an important component of the surgical disease burden in low- and middle-income countries. The consideration to introduce craniofacial surgery into such settings poses different questions, risks, and challenges compared with cleft or other forms of plastic surgery. We report the evolution, innovations, and challenges of a 30-year international craniofacial surgery partnership.

Methods: We retrospectively report a partnership between surgeons at the Uniwersytecki Szpital Dzieciecy in Krakow, Poland, and a North American craniofacial surgeon. We studied patient conditions, treatment patterns, and associated complications, as well as program advancements and limitations as perceived by surgeons, patient families, and hospital administrators.

Results: Since partnership inception in 1986, the complexity of cases performed increased gradually, with the first intracranial case performed in 1995. In the most recent 10-year period (2006–2015), 85 patients have been evaluated, with most common diagnoses of Apert syndrome, Crouzon syndrome, and single-suture craniosynostosis. In the same period, 55 major surgical procedures have been undertaken, with LeFort III midface distraction, posterior vault distraction, and frontoorbital advancement performed most frequently. Key innovations have been the employment of craniofacial distraction osteogenesis, the use of Internet communication and digital photography, and increased understanding of how craniofacial morphology may improve in the absence of surgical intervention. Ongoing challenges include prohibitive training pathways for pediatric plastic surgeons, difficulty in coordinating care with surgeons in other institutions, and limited medical and material resources.

Conclusion: Safe craniofacial surgery can be introduced and sustained in a resource-limited setting through an international partnership. *(Plast Reconstr Surg Glob Open 2016;4:e671; doi: 10.1097/GOX.0000000000000650; Published online 6 April 2016.)*

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Plastic surgeons have long helped treat the global burden of disease, particularly through cleft, burn, and trauma care.[1–4] Care delivery is evolving from medical mission models to also include emphasis on continuity of care,[5] outcomes,[6] cost-effectiveness,[7,8] and integration into broader health services.[9–12] Craniofacial surgery poses different demands, risks, and challenges compared with cleft or other forms of plastic surgery. Requisite adjunct medical services are more complex—including

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intensive care, skilled anesthesia, neurosurgery, and blood transfusion capabilities—and continuity of patient follow-up is critical. Noordhoff[13] and Sharma[14] have described their experience with and imperatives for establishing high-volume craniofacial centers in highly populous low-income countries. Less clear is how to develop craniofacial surgery through partnerships or exchange programs that might be more suitable to smaller populations.

We report an international craniofacial surgery partnership based at the Uniwersytecki Szpital Dzieciecy (University Children's Hospital) in Krakow, Poland, which is now in its 30th year. This article describes the evolution of the program, patients treated, and innovations both in the surgical model and surgical technology that have enabled it and critically addresses ongoing challenges.

EVOLUTION OF A PARTNERSHIP

In 1986, a didactic medical conference in Krakow, Poland, convened by Project HOPE brought together American (S.P.B.) and Polish (J.S.) plastic surgeons. In operating together on several sundry plastic surgical cases the following week, disparities between standards of surgical care between their countries of origin were illuminated and discussed, and a mutual interest in future collaboration emerged. Over the following 10 years, the surgeons evaluated patients and operated together in Krakow once or twice annually, gradually increasing the complexity of cases. In 1995, after sufficient team training, familiarity, and equipment had been accumulated, the first intracranial correction of a congenital facial deformity was performed.

The Jagellonian University (established 1364 CE) in Krakow and the University of Pennsylvania in Philadelphia have together provided academic institutional support during the 30-year partnership. The American surgeon (S.P.B.) has made 68 trips to Krakow to conduct clinic or operate in conjuction with local Polish surgeon colleagues (J.S., and junior partner B.S.) His role has been primarily of an advisory and teaching capacity, through a standing appointment as a visiting Jagellonian University professor. The Polish surgeons have visited the University of Pennsylvania as observing surgeons 4 times, and ancillary staff members (nurses, hospital administrators) have made multiple visits as well. More recently, University of Pennsylvania craniofacial surgery fellows have joined in the collaboration, in an operative teaching capacity with Krakow faculty and residents. Ongoing relationships have developed with anesthesia and intensive care physicians in Krakow. From the outset, the exchange has emphasized mutual learning, cultural sensitivity and respect, and tailoring interventions appropriate to the patient needs and hospital context in Krakow.

PATIENTS TREATED

In the most recent 10-year period (2006–2015) for which data are available, 85 patients have been evaluated by the partnership, with the most common diagnoses of Apert syndrome, Crouzon syndrome, or single-suture craniosynostosis (Table 1). In the same period, 55 major surgical procedures have been undertaken in partnership, with LeFort III midface distraction, posterior vault distraction, and frontoorbital advancement performed most frequently (Table 2). A 17-year-old male with Crouzon syndrome presented with forehead as well as mid- and lower-face retrusion, having previously undergone only frontoorbital advancement (Fig. 1). His age of presentation and treatment course are representative of patients treated through the partnership (Figs. 2 and 3).

During the most recent 10-year period (2006–2015), there have been 3 (5.5%) major complications

Table 1. Patient Conditions Evaluated by the Krakow Craniofacial Surgery Partnership in the Last 10 Years (2006–2015)

Condition	Patients
Apert syndrome	25
Crouzon syndrome	11
Pfeiffer syndrome	3
Encephalocele	2
Craniofacial clefting	5
Craniofacial microsomia	8
Single-suture craniosynostosis	10
Treacher-Collins syndrome	2
Frontonasal dysplasia/hypertelorism	6
Facial paralysis*	4
Pierre Robin sequence	3
Other	6
Total	85

*Includes congenital facial paralysis (without craniofacial microsomia), and patients with prior neoplasm (post-resection).

Table 2. Surgical Procedures Performed by the Krakow Craniofacial Surgery Partnership in the Last 10 Years (2006–2015)

Condition	Patients
Frontoorbital advancement	8
LeFort III midface distraction advancement	11
Monobloc distraction advancement	3
LeFort I maxillary advancement	2
Posterior vault distraction osteogenesis	9
Four-wall box osteotomy	1
Cranial vault reconstruction	5
Mandibular distraction osteogenesis	5
Orbital reconstruction with cranial bone graft	1
Costochondral mandibular reconstruction	2
Cranioplasty	2
Other	6
Total	55

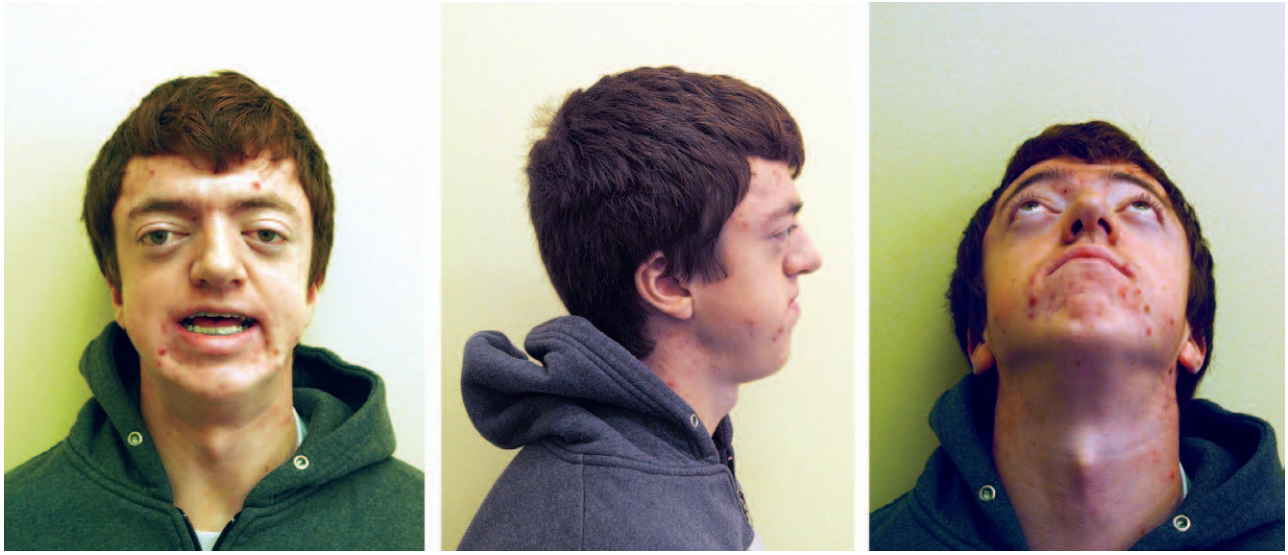

Fig. 1. Preoperative photographs of a 17-year-old boy with Crouzon syndrome presenting with forehead as well as mid- and lower-face retrusion, having previously undergone only frontoorbital advancement as a child.

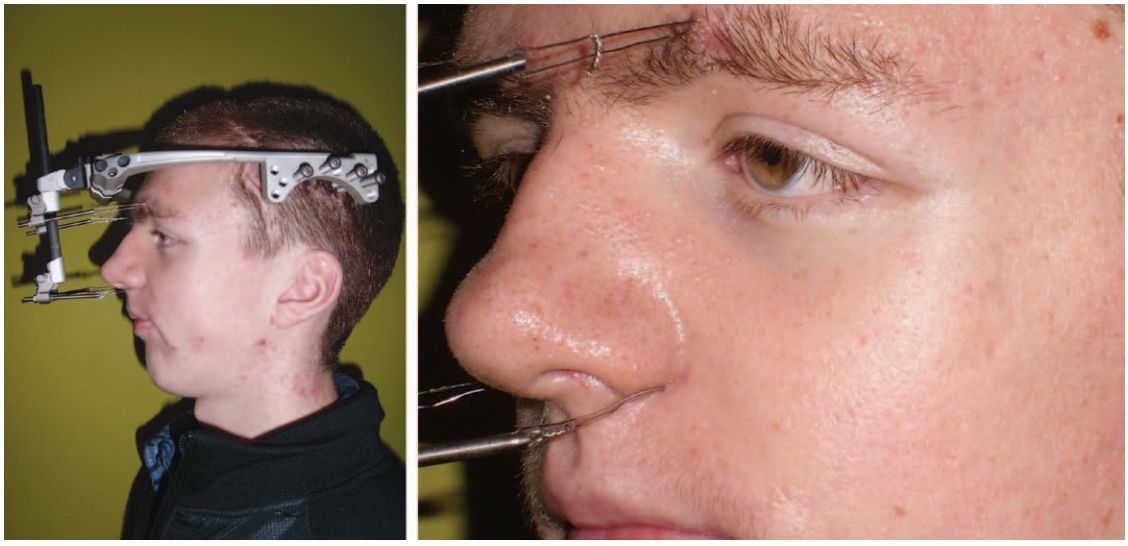

Fig. 2. The patient was treated with monobloc fronto-facial osteotomy and distraction osteogenesis with an external halo. Serial radiographs and photographs were taken during activation and posted electronically for all team members to review in real-time evaluation.

requiring surgical reintervention. Two of these cases involved LeFort III distraction of patients with syndromic craniosynostosis and midface retrustion. One patient developed premature consolidation of the midface, approximately 5 days into activation; this necessitated repeat midface advancement which was performed with shorter latency phase. The other patient experienced supratherapeutic distraction with an activation phase lasting approximately 20 days, which arose from miscommunication between the surgical teams in Krakow and North America and the patient's family. This patient will require orthognathic corrective surgery with maxillary setback (and/or mandibular advancement) to attain occlusion. A third patient developed a postoperative abscess which warranted return to operating room for incision. There were also 3 (5.5%) minor complications during this period of known superficial surgical site infection/cellulitis treated with antibiotics. There have been no patient deaths during the 30-year partnership.

FINANCIAL SUPPORT AND HEALTH SYSTEM INTEGRATION

Financial support for this partnership originated with Project HOPE, which from 1986 to 1991 funded international team travel; travel was subsequently

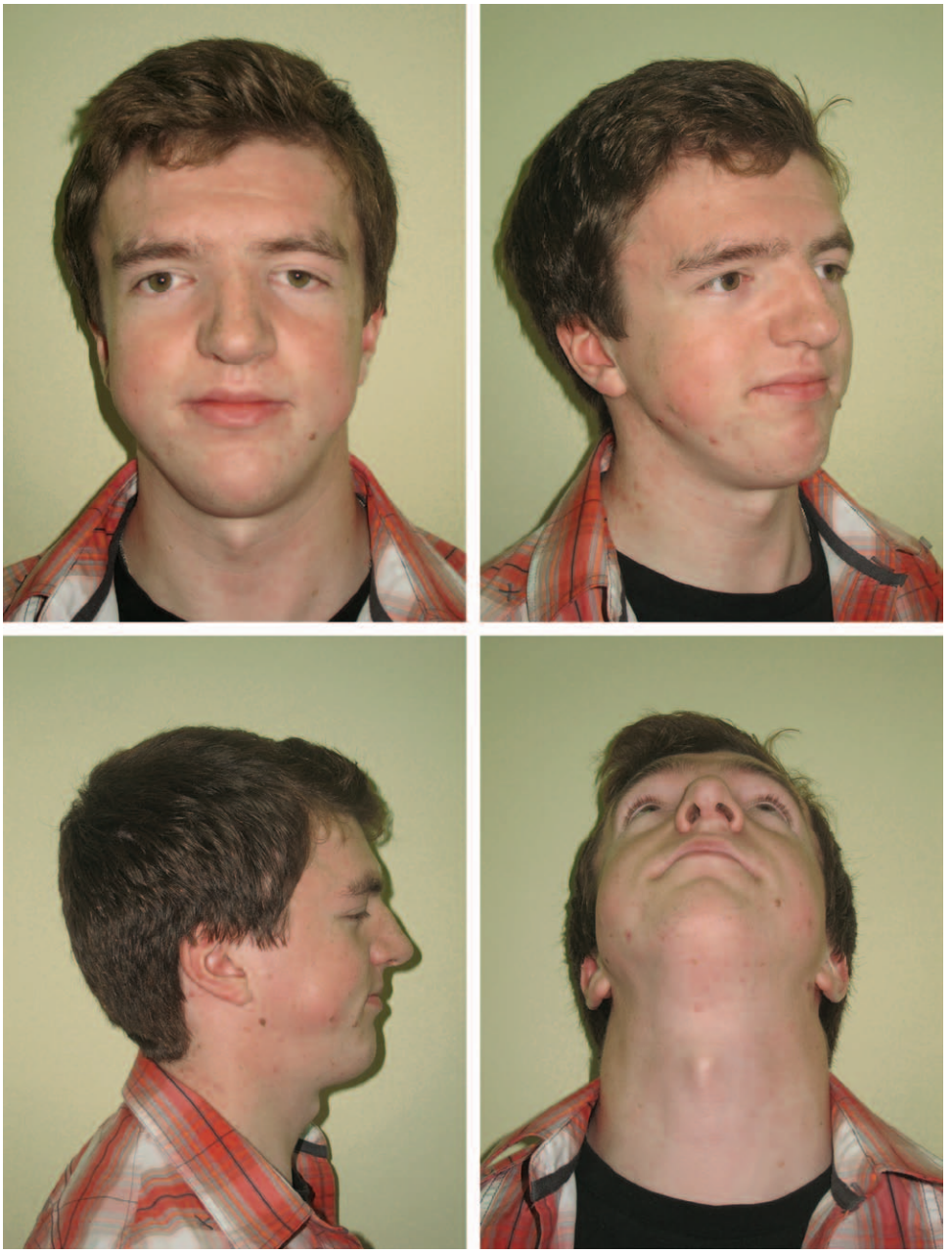

Fig. 3. Postoperative photographs 2 years later, after completion of monobloc advancement and bilateral sagittal split osteotomy and mandibular advancement.

funded by the Center for Human Appearance at the University of Pennsylvania and other agencies. Clinical care has been supported by the University Children's Hospital. Metal internal fixation plates were donated periodically by the manufacturers, in particular when 1 manufacturer transitioned from stainless steel to titanium miniplates in the North American market (Synthes Inc., West Chester, Pa.). And internal and external distractors were reused (after cleaning, checking condition, and sterilizing) after initial use in North America. The University Children's Hospital is now able to procure a limited number of titanium fixation plates for cases; distractors are not yet affordable.

The post-Soviet collapse in the 1990s illuminated the mounting barriers to all forms of medical and surgical care for children in Poland. Although the emphasis of this partnership has been craniofacial surgery, it has also been leveraged as a vehicle to support other forms of surgical care, in what might be considered "horizontal integration" into surgical disease management.[8] In 2005, the Children's Medical Foundation of Central and Eastern Europe was founded as a parallel 501(c)3 organization to provide

funding to well-managed yet underfunded hospitals of the region. Since inception, Children's Medical Foundation of Central and Eastern Europe has spent $600,000 to directly fund equipment ranging from laparoscopic equipment to oxygenators as well as thousands of hours of professional volunteer time.

INNOVATIONS

Four key innovations arose either to strengthen the partnership or to derive from it. First, craniofacial distraction in the early 2000s enabled a paradigm shift in the type and extent of surgeries performed. Shortly after McCarthy's[15] first description of mandibular distraction osteogenesis (and its early adoption in North America), we employed external mandibular distraction for micrognathic and retrognathic patients with craniofacial microsomia and Treacher-Collins syndrome. We subsequently evolved to internal distraction and also utilize posterior vault, LeFort II/III, and monobloc distraction in Krakow. Foremost, this has enabled larger and, in our view, safer advancements to be performed than with conventional fixation. The length of surgery seems to be shorter, concomitant blood loss reduced, low rate of infection experienced, and overall morbidity decreased (findings that parallel recent studies of distraction for posterior vault expansion).[16–18] Further, by utilizing consolidation rather than internal fixation to achieve osteosynthesis, we have a reduced need for (and cost of) internal fixation plates. Of course, this is offset by the need for distractors. The cost of new distractors (either internal or external halo systems) has not been historically feasible for the publicly funded hospital, but we have not yet encountered any equipment failures of reused devices. We use external halo devices for monobloc or midfacial advancement and find that it is easier for local surgeon partners to perform hardware removal and easier to cannibalize parts among the different distractors than with internal distractors. It is our hope that the cost of earlier-generation distraction may decrease to an affordable level allowing the use of new distractors in the future.

The second 2 innovations, Internet communication and digital photography, have gone hand in hand. At the outset of the partnership, postoperative patient management was done verbally (and hastily) by overseas telephone call and involved quite subjective descriptions and guidance. Beginning in about 2000, the availability of digital photography and email enabled photographs and pictures of radiographs to be shared postoperatively in a serial fashion and the management plan adopted accordingly. Further, learning through evaluation of serial radiographs facilitated teaching among the local team in Krakow. As the relationship has continued to evolve, the majority of postoperative management is directed by the Krakow team with guidance from North America only periodically modifying the local decision making. Of course there are limits to electronic communication and digital photography, and we have found that they do not replace a clear postoperative plan between the patient's family and both surgical teams.

The final innovation has been derived from the Krakow craniofacial exchange but has influenced surgical practice in North America. It is the observation that certain craniofacial morphology improves in the absence of surgical intervention. A specific example is that patients with Apert syndrome and associated frontal bossing, when treated initially with posterior vault distraction osteogenesis (PVDO), exhibit improvement in frontal bossing and frontal morphology. This has led us to both defer frontoorbital advancement and preferentially treat this patient cohort with PVDO even absent considerable posterior pathology, both of which appear to benefit patients in the form of less surgery in our experience to date. This realization would not have been as likely to arise in a North American practice, where the combination of a competitive marketplace of other providers, perceived "standards of care," and medicolegal defensiveness each reinforce the status quo. Furthermore, simply evaluating an older patient with less severe frontal morphology would have suggested a less severe syndromic variant. However, by following patients continually but with less opportunity to intervene surgically, we are thrust into a more actively observing role, and this can improve both our understanding of disease course and impact of treatment.

CHALLENGES

There continue to be several challenges to fully realizing a center in Krakow that delivers sustainable, high-quality craniofacial care and can be increasingly independent. First, recruiting and training successive pediatric plastic surgeons—and, furthermore, in craniofacial surgery—has proven difficult. The training pathway to pediatric plastic surgery in Poland originates from pediatric surgery not plastic surgery. Trainees complete general surgery then pediatric surgery residency and fellowship, the first and only plastic surgical training that comes as a 2- to 3-year apprenticeship subsequently. This regimen limits the depth of plastic surgical exposure, which likely compounds the challenge of complex craniofacial surgery. The long length and low salary of

this training regimen are discouraging and preclude those trained in plastic surgery from doing only a fellowship with pediatric or craniofacial focus. A plastic surgeon salary at the hospital, with an expected 38 hour-per-week commitment, is basic; and lack of formal plastic surgical training limits the opportunity to supplement this work with a private practice.

Second, as several surgeons elsewhere in Poland have expanded their surgical repertoire to incorporate some basic craniofacial procedures, how to best support them has been challenging. As a case in point, a neurosurgeon and an oral surgeon—each very competent and showing interest—have performed frontal-orbital advancement and orbital reconstruction with variable results and potentially incomplete corrections. Despite mutual interest, attempts to collaborate together on cases have been constrained by restrictive hospital credentialing policies. We are sensitive that our craniofacial outreach partnership fosters rather than undermines the abilities of local surgeons to provide craniofacial care. Although helping to train other surgeons is a logical long-term strategy, we struggle on behalf of our patients currently to advocate for other local surgeons when we perceive that at present the outcomes of our own partnership may be better defined. However, there are limitations to our partnership, specifically that we only operate on large cases together several times per year, and this limits early intervention. We also note that different subspecialties see deformities and treatment objectives differently. Paul Tessier charged that "craniofacial surgery should be performed only if it is the main interest of that surgeon"[19]; we struggle when the 2 options—a craniofacial outreach partnership or established surgeons doing occasional cases—are each imperfect.

Relating to each of these challenges, we must routinely critique the presence of North American surgeons to ensure it facilitates and does not impede the skill development of local surgeons. The Polish surgeon partners routinely perform smaller cases (eg, genioplasty, mandibular distractor removal) independently, and the acuity of these "smaller" cases continues to increase. At present, all team members seem to prefer to do the larger cases together. Also, recognizing the importance of high volume and a multidisciplinary team care, it is important that this partnership continue to grow in size and scope and potentially collaborate with any future similar efforts to achieve scale.

Third and finally are limited medical and material resources. As mentioned, neither distractors nor resorbable plates are available new, and the internal fixation sets are very limited. Reusing equipment such as distractors risks malfunction or device failure, inadequate sterility, and supply shortages. Further, lack of medical adjuncts such as gelatin hemostatic matrix likely contributes to what we perceive to be higher average blood loss compared with similar cases in North America. Although reports of mortality are rare in the literature, hemorrhage is the most common antecedent cause in intracranial surgery.[20] Given that there always exist uncontrollable risks to performing surgery in remote environments, being able to mitigate risks for which solutions exist would be optimal. Certain techniques we employ, such as perioperative hemostatic scalp sutures placed on either side of the bicoronal incision, have been adapted to the environment and may represent a form of "reverse innovation" from a lower-resource setting.[21] The environment of ingenuity fostered by these limitations is a silver lining, but clearly the availability of supplies used in wealthy countries is in the interest of superior patient care.

CONCLUSIONS

This partnership has enabled the development of a craniofacial surgery program in Krakow, Poland, over the last 30 years. It has facilitated the cultivation of surgical skills, capacity, and mutual learning in a lower-income setting. Until a future time when adequate experience and resources in Poland enable self-sufficiency, we plan to continue to strengthen craniofacial surgery incrementally through such a partnership, embracing safety, sustainability, and quality of care on behalf of our patients and their families.

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PATIENT CONSENT

The patient provided written consent for the use of his image.

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