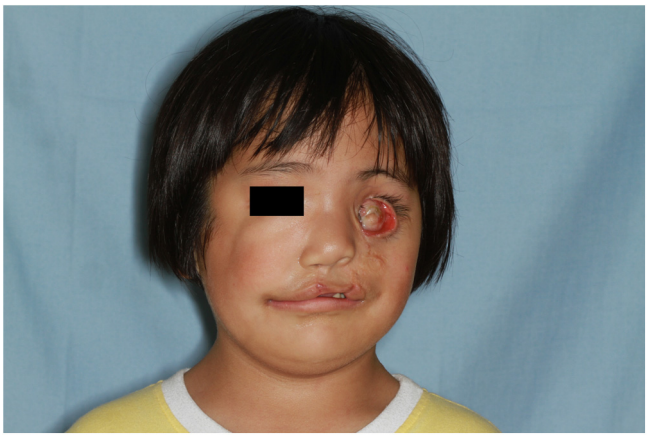

Fig. 1. Frontal view of the patient.

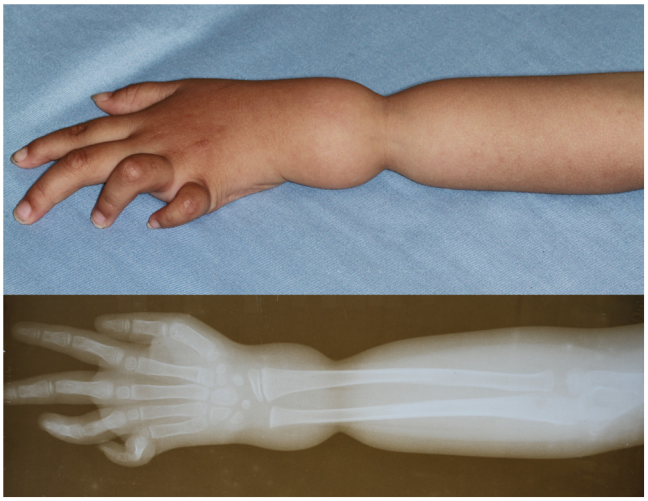

Fig. 2. Posterior view and radiograph of the left forearm.

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Available online 2 July 2019
https://doi.org/10.1016/j.bjoms.2019.06.019

Amniotic band syndrome with Tessier number 4 and 7 clefts: a case report

Sir,

Our patient was a 7-year-old girl who had been abandoned at birth, so there was no prenatal history available, and information about her parents could not be accessed. Physical examination showed a constriction ring on her left forearm, and multiple types of craniofacial clefts.

The left-sided Tessier number 4 cleft passed through the high point of the Cupid's bow and extended laterally to the nasal alae up into the inferiomedial canthal region, and had been repaired when she was two. The bony alveolar cleft arose between the central and lateral incisors on both sides, and coursed laterally to the pyriform aperture, medial to the infraorbital foramen. The hard and soft clefts in the palate were complete on both sides. The left lower eyelid was cleft with involvement of the nasolacrymal duct, the left orbit was exposed, and there was slight macrostomia on the right side. A thorough physical examination showed no malformations other than the facial clefts and constriction ring.

At admission, her left eye was blind with severe injection and chemosis of the conjunctiva and subepithelial haze. Her left forearm had a constriction ring, but the bones were not involved. Her middle, ring, and little fingers had adopted a curled-up position when relaxed, but the hand functioned normally (Figs. 1 and 2).

A Tessier number 4 facial cleft is a rare, complex, and challenging craniofacial malformation, and when found in conjunction with amniotic band syndrome, is even more rare. Nivaldo Alonso et al[1] reported 21 cases of the cleft, in which four patients had amniotic bands in the limbs. The coexistence of clefts number 4 and 7 is extremely rare, however. Sigler et al[2] presented a case of a unilateral partial Tessier number 7 cleft accompanied by incomplete number 2 and 3 clefts. As far as we know, a combination of Tessier number 4 and 7 clefts has not yet been reported.

Two theories have been proposed to explain the pathogenesis of amniotic band syndrome. The intrinsic theory is based on a developmental anomaly of the embryonic germinal disc, which makes the amniotic band a by-product rather than the cause of the fetal anomalies. The extrinsic theory holds that an early rupture of the amniotic sac causes the formation of amniochorionic mesodermal bands that lead to the development of a constriction band. Early rupture may decrease amniotic fluid, which could cause the compressive consequences of early constraint such as scoliosis and clubfoot.[3]

Keller et al[4] observed similar anomalies in cousins affected by the syndrome, which suggested a genetic cause. Conversely, Torpin[5] reasoned that its malformations might be the result of external influences. He proposed several criteria such as: asymmetrical findings; syndactyly that starts distally with free proximal segments; no bony defects of the remaining parts; and lesions associated with fibrous bands that had originated from a denuded chorion.

Based on these criteria, we think that the extrinsic theory can better explain all abnormalities in this patient. According to previous publications and our own experiences, both intrinsic and extrinsic theories together explain a subset of cases of amniotic band syndrome more clearly, which suggests to us that it is aetiologically heterogeneous.

Conflict of interest

We have no conflicts of interest.

Ethics statement/confirmation of patient's permission

We have ethics approval and the permission of the patient's guardian was given to publish the information in this paper.

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Available online 2 July 2019
https://doi.org/10.1016/j.bjoms.2019.07.003