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Case Report / *Olgu Sunumu*

Asymmetric conjoined twins: Gnathopagus parasiticus

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Abstract: In this study, a rare case of asymmetric conjoined twins that were connected to each other by their mandibles is described. There was an amorphous mass in between mandibles of the newborn calf. Radiological examination revealed that the amorphous mass consisted of a structure similar to the mandible and skull. According to the findings, the present anomaly was an asymmetric conjoined twins connected to each other by mandibles and named as gnathopagus parasiticus. Gnathopagus parasiticus case has not been reported in the literature previously.
Keywords: Asymmetric conjoined twins, autosite, gnathopagus parasiticus.

Asimetrik yapışık ikiz: Gnathopagus parazitikus

Özet: Bu çalışmada, çeneleri ile birbirlerine bağlanmış nadir bir asimetrik yapışık ikiz olgusu tarif edildi. Yenidoğan buzağının çene kemikleri arasında amorf bir kitle vardı. Radyolojik inceleme, amorf kitlenin mandibula ve kafatasına benzer bir yapıdan oluştuğunu ortaya koydu. Elde edilen bulgulara göre, mevcut anomalinin mandibulalar vasıtasıyla birbirine bağlanmış asimetrik yapışık ikiz olduğu anlaşıldı ve gnathopagus parazitikus olarak adlandırıldı. Gnathopagus parazitikus vakası daha önce literatürde bildirilmemiştir.
Anahtar sözcükler: Asimetrik yapışık ikiz, gnathopagus parazitikus, otosit.

Twins are the result of a complete or incomplete division of the morula stage of a zygote. Conjoined twins develop as a result of an incomplete division of a morula or blastocyst. The conjoined twins can develop symmetrically or asymmetrically (3). Symmetrical conjoined twins are complete and connected at various body regions (2, 9); whereas in the condition of asymmetric conjoined twins one of the twin is incomplete (heteropagus or parasite) and smaller, and it attaches on the body of the fully-developed twin (autosite) (3). Live births of asymmetric conjoined twins are rare due to fetal dystocia caused by twins and thus a Caesarean section is required in most of the cases (3).

Twins may arise from the influence of genetic or environmental factors such as infectious agents, drugs, toxic plants, and physical agents (8). However, making assumptions on the etiology is often not possible due to the role of multiple factors on developmental anomalies in animals (4).

Conjoined twins are named according to the location of the junction of the body and whether it is symmetrical or asymmetrical (4, 9). In this case, the calf had an abnormal mandible structure and an amorphous mass attached to this mandible. Detailed examination revealed that the abnormal mass was a parasitic twin of the calf which has not completed its development in time. Both parasitic and autosite twins were found to be stuck together from their mandibles. Therefore, the case was named as gnathopagus parasiticus in accordance with the literature. To the best of our knowledge, case of gnathopagus parasiticus has not been reported in the literature.

A Simmental calf (two-days-old, male) was admitted to the clinic due to incapability of food intake caused by an abnormal mandibular structure. After radiological

examination, an amorphous mass between the mandibles was surgically removed and the remaining mandibles were appropriately connected to enable self-feeding of the calf. Tissues obtained from the amorphous mass were fixed in 10% neutral buffered formalin, embedded in paraffin wax, and sectioned at 5 µm thickness. The bone samples of amorphous mass were decalcified with formic acid and sodium citrate method. Histologic sections were stained with hematoxylin-eosin (HE). There was no maternal or pedigree history available and karyotype analysis could not be performed.

In the clinical and radiological examination of the calf's head, we observed two short and disconnected mandibles of parasitic twin attached to the normal-sized disconnected mandibles of the autosite (Figure 1 A, B). Additionally, there was a structure, similar to a skull, attached to these mandibles of the parasitic twin with a joint. Hence, it is determined that the case represents asymmetric conjoined twins which were attached to each other via their mandibles. The parasitic twin was surgically removed from the autosite and examined in detail.

The parasitic twin was covered with normal skin. Cheeks and lips were present and attached to the mandible of the autosite. They exhibited normal oral mucosa with conical papilla (Figure 1 A). Hard palate and soft palate were protruding into the oral cavity of the parasitic twin (Figure 1 D). The hard palate had palatal rugae and palatal raphe, and they extended laterally. A soft palate in the aboral of hard palate was protruding into the oral cavity. Premolar and molar deciduous teeth were present in the maxilla of the parasitic twin (Figure 1 C). There were no symphysis mandibulae in the mandibles of both the autosite and the parasitic twin. The incisive teeth belonging to each of the mandibles were facing each other in parasitic twin. The nasolabial plate of the parasitic twin had a shape similar to the mentum (Figure 1 C, D).

The nasal cavity, nasal conchae, and paranasal sinuses were absent in the parasitic twin. However, the temporomandibular joints were fully formed. A coronoid process and condyle, as well as the mandibular and mental foramina, were present in the mandible of the parasitic twin. The maxilla and zygomatic bones were prominent and rudimentary remains of frontal bone were observed in the parasite. The orbita of the parasitic twin was fully formed and filled with fat tissue (Figure 1 E).

The cranial cavity was revealed when the parasitic twin was cut along the median line. The cavity was covered with a white, hard and a relatively thick layer of dura mater. Under the dura mater, cerebrum and cerebellum with a whitish lobular structure were surrounded by transparent mucinous liquid (Figure 1 F). No other abnormalities were observed in the body of the autosite twin.

Histopathological examinations of the parasitic twin revealed normal histological structures of the skin, oral mucosa, hard palate, and soft palate. The orbitas were filled with fatty tissue. The bone tissues of the skull and mandibles were formed by immature bone structure composed of trabeculae with calcified cartilage areas (Figure 2 B). Periosteum and endosteum were well developed in both bone tissues although osteoblasts were present on the trabeculae; osteoclasts were not observed (Figure 2 A, B). There was no bone marrow in the space between the trabeculae (Figure 2 A, B). The dura mater covering the internal surface of the cranial cavity was thickened. In the brain, the layers of the gray matter were not fully formed, the white matter was hypomyelinated, and the ependymal cells were arranged in multifocal rosette formations (Figure 2 C, D). Choroid plexus-like structures were present in some of the sections. In the cerebellum, the molecular layer was relatively thin with the presence of a few Purkinje cells. The granular layer and white matter have not been formed (Figure 2 E, F).

Congenital duplication anomalies are uncommon in humans and animals (4). Symmetric conjoined twins can be encountered in ruminants. In cattle, symmetric conjoined twins are commonly connected in the cranial region while connections in the caudal region are more common in sheep and pigs (2, 4). The symmetrical conjoined twins in the calf are usually parapagus (doubling of all cranial structures) cases in the cranial region in the form of diprosopus (facial duplication) (1, 8) or dicephalus (cranial duplication) (5, 12).

Asymmetric conjoined twins are extremely rare in animals (10). There are monochorionic and monoamniotic twins, consisting of an incomplete twin attached to the fully developed body of the co-twin (11). Abnormal conjoined twins are seen in different forms such as an externally attached parasitic twin, an enclosed fetus in a fetus, an internal teratoma or an acardiac twin connected via the placenta (10). A parasitic twin does not have its own circulatory system, thus embryonic death occurs due to the inadequacy of vascularization. The nutritional supply of parasitic twin is only provided via the veins of the autosite's cardiovascular system (9). In the case presented in this report, the vascularization of the parasitic twin was ensured by the branches emerging from the autosite's linguofacial trunk.

Although the exact mechanism of conjoined twinning is not clear, fission and fusion theories are proposed (9). However, the fission theory cannot explain the location of the connection between the conjoined twins in the vertebrate embryos. The hypothesis behind the development of conjoined twins which is predominantly accepted is the secondary local fusion of two separate embryonic discs formed in the early embryonic period (9). There are also studies suggesting that twins may arise from fusions following fission of a single embryonic disc (12).

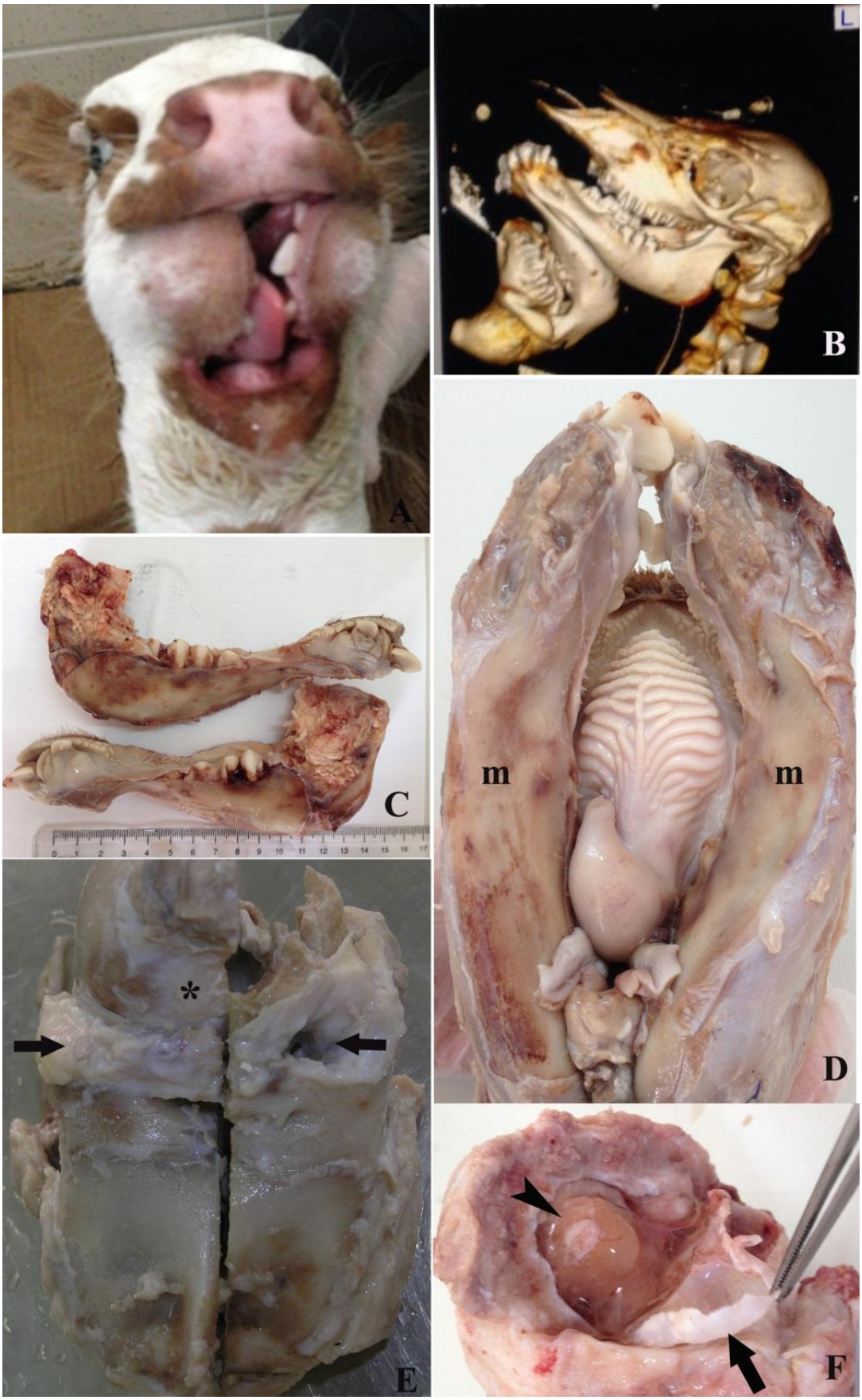

Figure 1. Macroanatomic views of the asymmetric conjoined twins.
The overview rostral (A), radiologic (B), mandibles (C), ventral view of the parasitic twin: m- mandible (D), dorsal view of the parasitic twin: calvarium (*), orbita (arrow) (E), the cavity of cranium of the parasitic twin: dura matter (arrow), brain (arrowhead) (F).

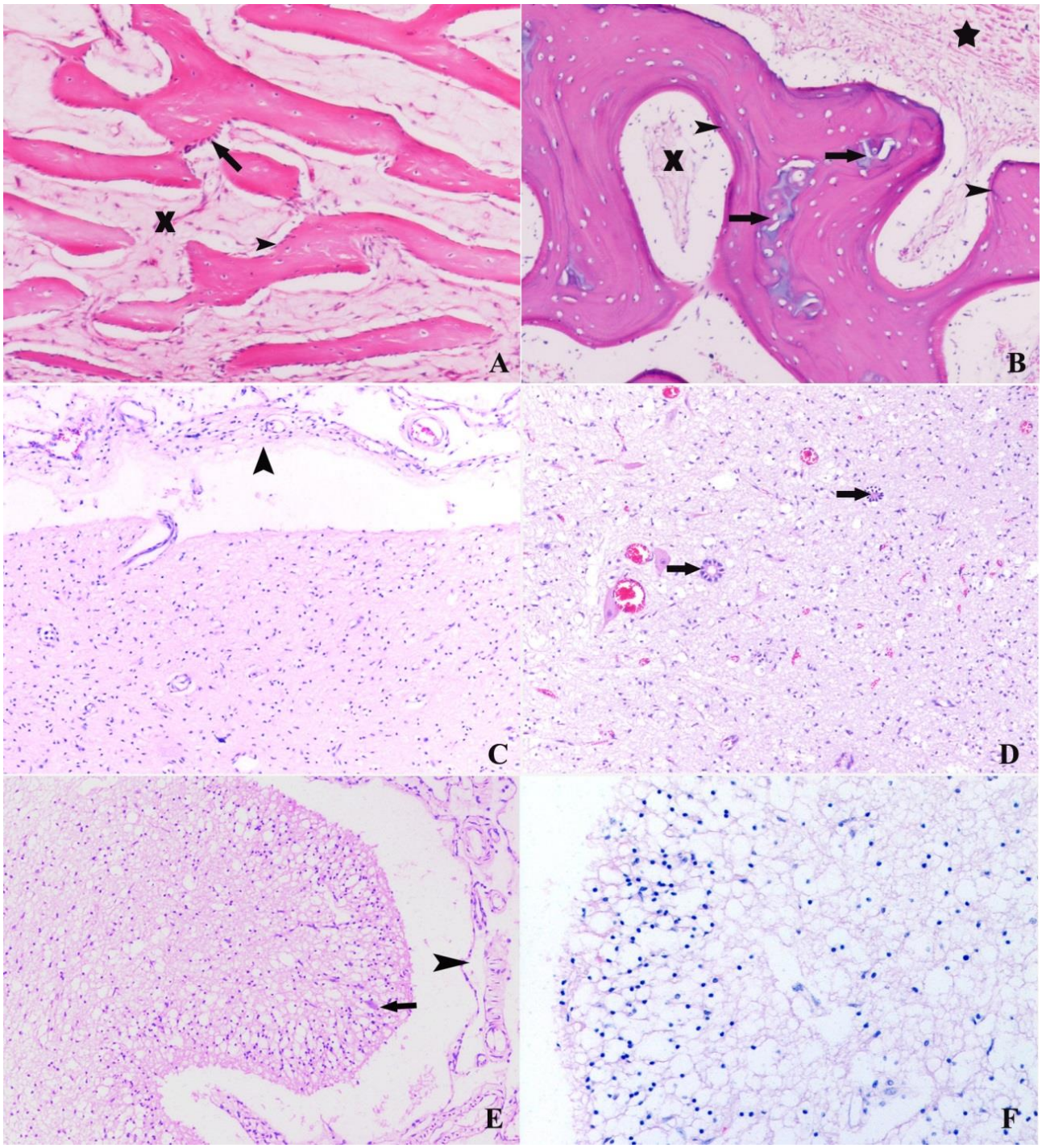

Figure 2. Histopathological sections of the parasitic twin.
Skull, osteoblast (arrow), endosteum (arrowhead), marrow space (X) (A), HE, 10X. Mandible, calcified cartilage in trabeculae (arrows), endosteum (arrowheads), periosteum (asterisk), marrow space (X) (B) HE, 10X. Cerebral cortex, pia mater (arrowhead) (C), HE, 10X. Cerebrum, rosette formations (arrows) (D), HE, 10X. Cerebellum, Purkinje cell (arrow), pia mater (arrowhead) (E), HE, 4X. Cellular disorganization in cerebellum, (F), HE, 20X.

The diagnosis of asymmetric conjoined twins is difficult due to the ectopic localization and poor organization (2). In the present case, radiological examination confirmed the localization of the parasitic twin which was attached to the mandible of the autosite.

Conjoined twins, including those connected through the mandible, were reported in human medicine. In these cases, it has been reported that symmetrical conjoined twins (gnatho-thoracopagus) were connected by an adhesion zone which started from the mandible and

extended into the thoracic region (6, 7). However, the present case reveals asymmetric conjoined twins in which autosite and parasite were connected to each other only through the mandibular regions.

In the present case, it was not possible to analyze a specific cause of the development of such abnormality in the family type small animal husbandry enterprise. In this type of farms, it is not possible to get accurate information about animal feeds or grazing pastures. Furthermore, these types of farms also generally do not keep the pedigree of animals. These calves were born in the first pregnancy of the mother, but there is no information about the inseminating bull. Due to the lack of information on the etiopathogenesis of congenital duplication anomalies, it was not possible to determine the underlying cause of the abnormality in this newborn calf. We hope that this study will contribute to determining the incidence of asymmetric conjoined twins in cattle.

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Ethical Statement

This study does not present any ethical concerns.

Conflict of Interest

The authors declared that there is no conflict of interest.

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