

Diplomarbeit

**Anatomy of the Le Fort I Segment:
Are arterial variations a potential risk factor for avascular bone
necrosis in Le Fort I osteotomies?**

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unter der Anleitung von

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Graz, 25.07.2018

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Graz, am 25.07.2018

Simon Bruneder eh

Vorwort

Die vorliegende Publikation befasst sich mit der arteriellen Gefäßversorgung des Le Fort I Segmentes sowie möglichen Auswirkungen variabler Gefäßvarianten.

Die Le Fort I Osteotomie ist eine Routine Operation in der Mund-, Kiefer und Gesichtschirurgie, welche zur Umstellung dento-fazialer Missbildungen zum Einsatz kommt. In dieser Publikation wird die Standard Variante der arteriellen Gefäßversorgung des Le Fort I Segmentes mit einer speziellen, vorher noch nicht beschriebenen Variante verglichen.

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Die Entstehung dieses Artikels ist eng mit meiner Tätigkeit am Institut für Anatomie der Medizinischen Universität Graz verbunden. Von Anfang an meiner dortigen Tätigkeit faszinierte mich besonders die Anatomie der Kopf- Hals Region.

Die am Institut für Anatomie Graz entwickelte Konservierungsmethode und die Injektion der arteriellen Gefäße mit einer speziellen röntgenopaken Latex-Lösung bieten einzigartige Möglichkeiten zur wissenschaftlichen Untersuchung des Gefäßsystems.

Mein Humanmedizin Studium schloss ich 2015 mit dem Diplomarbeitsthema

„*Die vaskuläre Versorgung des Oberkiefers, im Hinblick auf die Le Fort I Osteotomie*“ ab.

Im Zuge dieser ersten Diplomarbeit beschäftigte ich mich eingehend mit der vaskulären Gefäßsituation des Oberkiefers und der Le Fort I Osteotomie.

Aus dieser ersten Diplomarbeit ergaben sich Grundlagen und Erkenntnisse, genauso wie offene Fragestellungen als Ausgangspunkt für den vorliegenden Artikel. Vor allem aber das Interesse zum Weiterforschen auf dem Gebiet der arteriellen Anatomie des Oberkiefers war geweckt.

Dank meinem Vorgesetzten Prof. Anderhuber hatte ich große Freiheit bezüglich meiner Forschungstätigkeit und konnte ausgiebig Daten sammeln, Methoden probieren und Erkenntnisse gewinnen, was letztendlich zur vorliegenden Publikation führte.

Herausfordernd waren beim Erstellen der vorliegenden Arbeit die Durchführung und das Finden einer passenden, wissenschaftlich validierten Messmethode zur Evaluierung der arteriellen Gefäßdurchmesser des Le Fort I Segmentes.

Verschiedene Ansätze wurden ausprobiert. Schließlich wurde mit der von *B. Hillen* in seiner Dissertation „*Form and haemodynamics of the circulus arteriosus wilisi*“ beschriebenen

Methode eine in der Durchführung zwar etwas umständliche, aber wissenschaftlich anerkannte Methode gefunden.

Schlussendlich habe ich die von *Anderhuber et. Weiglein* modifizierte Variante dieser Messmethode gewählt.

An der Medizinischen Universität Graz werden Basisfertigkeiten des wissenschaftlichen Arbeitens unterrichtet. Meine derartigen Kenntnisse konnte ich durch die praktischen Anwendungen im Zuge des Erstellens dieses Manuskriptes erweitern.

Lehrreich war die Auseinandersetzung mit dem wissenschaftlichem Schreibstil, den formalen Anforderungen einer wissenschaftlichen Publikation sowie den Vorgaben einschlägiger Fachzeitschriften.

Das Verfassen einer wissenschaftlichen Arbeit in Englisch war auch eine Herausforderung, die mir einiges an Mühe abverlangt hat.

Darüber hinaus konnte ich meine Fähigkeiten mit verschiedenen Computerprogrammen wie IBM SPSS, Excel und Randomizer ausbauen.

Auch die Erstellung von Grafiken und Schemata stellten zeitintensive Aufgaben dar. Erste handschriftliche anatomische Skizzen und selbstgezeichnete detailreiche Darstellung mussten auf die wesentlichen Punkte reduziert und vereinfacht werden. Zur Übertragung der Handskizzen war die Erarbeitung grundlegender Fähigkeiten mit modernen Bildverarbeitungsprogrammen notwendig.

Das Erstellung der gesamten Arbeit, vom Finden einer adäquaten Messmethode, Erheben der Daten, Verfassen des Artikels einschließlich des Review Prozesses, war umfangmäßig aufwendiger, aber auch lehrreicher als eine herkömmliche Diplomarbeit. Auf jeden Fall konnte ich durch neu gewonnenen Kenntnisse und Erfahrungen im Zuge dieser Arbeit profitieren.

Danksagungen

Ein besonderer Dank gilt meinen Betreuern und Coautoren Priv. Doz. DDr. Wolfgang Zemann und Ao. Univ. Prof. Dr. Andreas Weiglein welche mir bei der Erstellung dieser Publikation mit ihrer Erfahrung mit Rat und Tat zur Seite gestanden sind.

Ein weiterer Dank gilt dem Elsevier Verlag der großzügigerweise die Verwendung des Manuskriptes (preprint) des bereits publizierten Artikels für die folgende Diplomarbeit gestattet hat.

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Anatomy of the Le Fort I segment: Are arterial variations a potential risk factor for avascular bone necrosis in Le Fort I osteotomies?

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Abstract

Background and objectives: Osteotomies of the Le Fort I segment are routine operations in orthognathic surgery with low complication rates. In this context ischemic complications are, with an occurrence of about 1% rare, but can have severe consequences. These ischemic complications range from gingival retraction and/or loss of tooth vitality to avascular bone necrosis of the mobilized Le Fort I segment. Anatomically the blood supply of the Le Fort I segment is mainly based on the ascending palatine, the ascending pharyngeal, the descending palatine artery and smaller arteries that reach the alveolar process from the buccal side. The aim of this study was to investigate the blood supply and special arterial variants of the Le Fort I segment responsible for arterial hypoperfusion or ischemic avascular necrosis after surgery.

Material and methods: A total of 30 halved human cadaver head specimens, preserved with a high-quality embalming method were selected according to defined inclusion and exclusion criteria. All specimens were subjected to arterial latex injection for improved visibility. The arterial anatomy of the Le Fort I segment was analyzed after the ascending palatine and ascending pharyngeal arteries were completely dissected until the submicroscopic level by clinical and anatomical specialists. Dissection was performed using surgical standard methods and a dissection microscope. In all specimens the arterial variants of the Le Fort I segment as also the arterial diameters measured at two points (D1: origin and D2: palate-entry of artery) were evaluated to categorize the occurring types of arterial variations.

Results: The typical known palatine vascularization pattern was apparent in 90% of all specimens, in which the ascending palatine artery (D1: 1,2mm \pm 0,34mm; D2: 0,8mm \pm 0,34mm) and ascending pharyngeal artery (D1: 1,3mm \pm 0,58mm; D2: <0,4mm) were both supplying the Le Fort I segment. However in 10% of all specimens, the Le Fort I segment was dependent on the ascending pharyngeal artery alone. In these special cases the missing ascending palatine artery was replaced with the anterior branch of the ascending pharyngeal artery (D1: 1,9mm \pm 0,32; D2: 1,0mm \pm 0,3mm).

Conclusion: This study is the first description of a special type of arterial variation of the Le Fort I segment which occurred in 10% of the investigated cases. The type of this arterial variation, potential risk factors and resulting clinical consequences accompanied with the variation are explained. Individuals with this special variant of arterial anatomy may clinically be at a high risk of hypoperfusion and avascular necrosis of the Le Fort I segment due to surgical procedures and segment

mobilization. An individualized operation plan that considers the arterial anatomy may prevent ischemic avascular complications in at-risk patients.

Abstract (deutsch):

Die Le Fort I Osteotomie ist eine Routine Operation mit einer insgesamt geringen Komplikationsrate. Ischämische Komplikationen treten selten auf, können aber zu schwerwiegenden Konsequenzen wie Osteonekrose des Le Fort I Segmentes führen. Ziel dieser Arbeit ist es, die Blutversorgung des Le Fort I Segmentes sowie spezielle anatomische Varianten der arteriellen Blutversorgung des Le Fort I Segmentes welche zu Osteonekrose führen könnten, zu untersuchen.

Material und Methoden: Die arterielle Anatomie des Le Fort I Segmentes wurde anhand anatomischer Methoden und Vermessung an 30 Schädelpräparaten untersucht. Die arteriellen Versorgungstypen sowie die arteriellen Durchmesser wurden an vorher definierten Messpunkten (D1, D2) analysiert.

Resultate: Beim typischen Versorgungsmuster (90%) tragen A. paltina ascendens (D1: 1,2mm ± 0,34mm; D2: 0,8mm ± 0,34mm) und A. pharyngea ascendens (D1: 1,3mm ± 0,58mm; D2: <0,4mm) zur Blutversorgung bei. Das Le Fort I Segment wird in 10% der untersuchten Fälle von der A. pharyngea ascendens (D1: 1,3mm ± 0,58mm; D2: <0,4mm) alleine versorgt.

Konklusion: Die beschriebenen arteriellen Varianten sowie die Auswirkungen der gemessenen Gefäßdurchmesser auf das Le Fort I Segment werden diskutiert. Eine beschriebene Variante könnte klinisch eine Osteonekrose des Le Fort I Segmentes begünstigen.

Introduction

Osteotomies of the Le Fort I segment are routine operations in orthognatic surgery and are clinically used to correct skeletal deformities of the mandibulo-maxillary complex. As with any surgical procedure, general and specific complications are associated with this operation (Kim and Park, 2007). With a complication rate of 6.4% the osteotomy of the Le Fort I segment is considered to be a safe surgical procedure (Tung et al., 1995; Kramer et al., 2004).

However one rare-reported complication is the ischemic avascular bone necrosis of the palate (Westwood and Tilson, 1975; Lanigan et al., 1990; Lanigan, 1997; Singh et al., 2008; Pereira et al., 2010; Teemul et al., 2017). The clinical appearance of ischemic avascular necrosis ranges from the loss of tooth vitality or the presence of mucosal or periodontal defects and gingival retractions (Gardner, 2007) to severe necrosis of the bony segment or the palate (Bell et al., 1995). Kramer et al. reported that the incidence of ischemic complications is about 1%. 0,2% of these ischemic complications are further associated with very severe forms such as the maxillary bone necrosis. (Kramer et al., 2004).

Practically, postoperative blood flow diminution after Le Fort I osteotomy in animals can be measured in the osteotomized segment. In order, minimal bone necroses are likely to be actually quite common during the surgical Le Fort I mobilization in clinical practice (Lanigan et al., 1990).

Anatomically the blood supply of the Le Fort I segment is mainly based on the ascending palatine and ascending pharyngeal arteries, which reach the Le Fort I segment from the posterior (Bell, 1969; Siebert et al., 1997). Despite their small diameters, the arteries that reach the Le Fort I segment from the buccal side also contribute to the segment's blood supply (Westwood and Tilson, 1975; Piehslinger et al., 1991; Siebert et al., 1997). Further the descending palatine arteries also reach the Le Fort I segment, but their ligation may often be necessary during the Le Fort I segment mobilization in the surgical routine.

In that context, the importance to maintain the descending palatine arteries for the blood supply of the Le Fort I segment is discussed controversially in the literature (Dodson et al., 1997; Lanigan, 1997; Omura et al., 2015). Lanigan et al. reported about ischemic avascular bone necrosis after ligation of these arteries in some cases (Lanigan et al., 1990). In contrast, the ligation of the palatine descending arteries during surgery can stop bleeding or prevents thrombosis without postoperative

complications. Therefore Dodson et al. suggested the ligation of these arteries to avoid such complications (Dodson et al., 1997).

However, excessive blood flow diminution to the palate has been observed after intraoperative ligation of the descending palatine artery (Meyer and Cavanaugh, 1976; Nelson et al., 1977; Nelson et al., 1978; El Deeb et al., 1981). Further excessive Le Fort I segment mobilization results in susceptibility to arterial damage, because of bending or kinking forces that affect the arteries. Due to altered hemodynamics with diminished blood perfusion, surgical segment mobilization can cause arterial thrombosis and/or avascular bone necrosis of the palate (Lanigan et al., 1990; Singh et al., 2008).

Beneath these intraoperatively occurring surgical complications some other reasons for severe avascular necrosis have been discussed. Some authors suspected anatomical variations as potential risk factors for avascular bone necrosis of the Le Fort I segment. More precisely they found that patients with an altered arterial anatomy of the palate due to previous operations or due to congenital deformities may be more prone to avascular complications or necrosis in that region than others (Maher, 1977; Lanigan et al., 1990).

Siebert et al. already described the arterial anatomy of the palate which is commonly responsible for the Le Fort I segment's blood supply (Siebert et al., 1997). Gauthier et al. partially confirmed these findings but also claimed for larger case series to investigate the arterial blood support and further potential arterial variations of the Le Fort I segment more precisely (Gauthier et al., 2002).

In order, the aim of this study was to examine the blood supply the Le Fort I segment using modern embalming techniques and sample series sufficiently enough to identify specific arterial variations. The hypothesis of this study was that the Le Fort I segment is anatomically not always supported by the commonly known standard type of arterial blood supply. Hypothetically a special kind of arterial variation may clinically lead to a reduced blood supply (hypoperfusion) or to vascular complications such as ischemic avascular necrosis of the Le Fort I segment, especially after osteotomy or surgical mobilization of the palate.

Materials and Methods

In this prospective randomized study, the arteries of the Le Fort I segment and the anatomy of the palatine region were examined. The study was carried out using human cadaver specimens that were obtained from the Department of Anatomy of the Medical University of Graz, Austria.

Within its special donation program approved by the Medical university of Graz all human specimens utilized in this study have been donated to the Institute of Anatomy for scientific research and training purposes¹. Within this Donation Program all specimens were anonymized before their usage.

All cadaver specimens were preserved according to Thiel's embalming method (Thiel, 1992b) (**Fig. 2, 3, 4, 6, 7,**), which is commonly used for high quality surgical training (Okada et al., 2012; Cabello et al., 2015). The arteries of all specimens were filled with a bright red latex solution, for an improved visibility. Due to the characteristics of Thiel's method the arteries can be qualitatively dissected down to the level of 0,2mm. In order the Le Fort I segment's blood supply was assessed by the anatomical detection of the filled arteries down to the submicroscopic level.

A total of 128 halves of human cadaver heads² have been assessed for eligibility in this study. From these 128 cadaver specimens 45 were randomly selected using a computer program³. From these 45 specimens 15 were secondly sorted out, by anatomical and maxillofacial specialists according to defined inclusion and exclusion criteria.

These inclusion criteria were: 1.) complete soft and hard tissue preservation, 2.) mature specimens, 3.) anatomical integrity of the pharyngo-palatine region, 4.) successful arterial latex injection.

The exclusion criteria were: 1.) obvious damage to the ascending palatine or ascending pharyngeal arteries, 2.) obvious damage of external carotid branches, 3.) previous palatine operations or surgical training, 4.) malformations or deformities, 5.) unsuccessful latex injection of terminal arterial branches.

Finally a total of 30 halves of high quality-preserved cadaver heads meeting the selection criteria (males: 17, females: 13; age: mean=77,6 ± 10,5 years, min=59 years, max=94 years,) were examined. All specimens (n=30) were further allocated to the dissection procedure. Each dissection

¹ Within this donation program no further approval by an ethical board is required.

² Cadaver heads have been separated by professional employees of the Institute of Anatomy in the median-sagittal plane using a circular saw (blade thickness <1mm).

³ Randomizer® <https://www.randomizer.at>; randomization for clinical and non-clinical trials; Medical University of Graz, Austria

was done by both an anatomical specialist and a maxillofacial specialist with experience of more than 10 years. The dissection procedure was performed with an operation microscope⁴ using surgical standard methods until the submicroscopic level.

Firstly, the common carotid artery as well as the internal and the external carotid artery were completely dissected with respect to arterial variations. All external carotid branches were dissected completely until a submicroscopic level. The internal carotid artery was dissected up to the cranial base. Pharyngeal ascending and palatine ascending artery were followed to the palatine region. The facial artery was dissected up to the facial region. The maxillary artery was dissected until it's final branches in the infratemporal fossa. The palatine canal within the descending palatine arteries was opened with an osteotom⁵.

When dissection procedures were completed each specimen was finally examined by an anatomical and maxillofacial specialist to identify the types of arterial variations in the Le Fort I segment.

A further assessment of the Le Fort I segment's blood supply was done by the standardized method according to Hillen (Hillen, 1986b) (**Fig. 1**). This method is a commonly used standard for the measurement of arterial diameters and was originally developed for the diameter assessment of the cerebral arteries (Hillen, 1986a; Hillen, 1986b; Anderhuber et al., 1990; van der Zwan et al., 1993). In our study the arterial diameters of the ascending palatine artery and the ascending pharyngeal artery as well as the anterior branch of the ascending pharyngeal artery were measured at two measuring points on each artery. For an objective comparison between the diameters these measuring points (D1, D2) were defined prior to the measurement process as follows: D1 was set to be the arteries' origin in the carotid triangle and D2 was set to be the arteries' entry into the soft palate (level of the soft palate), which is located directly next to the Le Fort I segment. (**Tab. 1**) D1 is the arterial diameter measured at the offspring of the artery, before giving rise to further arterial branches. The arterial branches which are given rise between D1 and D2 partially contribute to the Le Fort I segment's blood supply through thin branches (pharyngeal capillary net) and partially contribute to other anatomical structures. D2 is the reduced diameter of the same artery where D1 was already measured, located directly next to the Le Fort I segment at the level of the soft palate. D1 was chosen under the hypothetical adoption that this diameter plays an additional role for the Le

⁴ Carl Zeiss OPMi-1, Carl Zeiss Medical Products AG, Oberkochen, Germany

⁵ Blade thickness <1mm

Fort I segment's blood supply through the capillary network. The diameter D2 which is more important for the blood supply of the palate than D1 was chosen to be the very last endpoint-diameter before the artery enters the Le Fort I segment through the soft palate.

D1 was measured to get an overview about the arterial diameter at the origin of the Le Fort I segment's vascularization pattern, but not directly for assessment reasons of the segment's arterial perfusion. Therefore D2 was considered to be the measurement point that is responsible for the arterial capacity of the Le Fort I segment due to its localization.

The arterial diameter measurement was performed according to Hillen's method (**Fig. 1**) by carefully removing a section of approximately 0,2-0,3cm of each artery from the specimen at the defined measuring point (D1, D2). These removed arterial sections were pressed with a pair of standard anatomical tweezers and the hemi-circumference ($= \frac{c}{2}$) was measured using a conventional Vernier caliper⁶. In order the arterial diameter could be calculated using the standardized method according to Hillen ($d = \frac{c}{\pi}$) (Hillen, 1986a). Arterial diameter measurements were limited to a minimum diameter of 0,4mm, due to increasing subjectivity in the measuring process the smaller the diameter gets. Therefore diameters <0,4mm were adopted to be measured not accurately enough (**Tab. 1**)

For an according assessment of the measurement results statistical calculations consisted of descriptive methods, that were used to analyze minimum, maximum, mean values and standard deviations. Difference values of the arterial diameters of the main arteries supplying the Le Fort I segment were calculated comparatively at the measurement point D2 to assess a potential topography-based hypoperfusion due to an altered arterial anatomy.

An objective comparison of the measured results was done with a professional statistical computer program⁷. To further demonstrate the variation of the origin of the palatine ascending and pharyngeal ascending arteries Pie-chart diagrams were created. (**Tab 2, 3**) Moreover a flow-chart diagram based on the Consort statement for clinical trials was created to report the study enrollment. (Schulz et al., 2011) (**Tab. 4**)

⁶ Vernier Caliper Atom CI 902061; Hahn & Kolb Werkzeuge GmbH, Ludwigsburg, Germany

⁷ R-project ©; Austrian Association for Statistical Computing; R-Foundation for Statistical Computing, Vienna, Austria

Results

In all included specimens (n=30) the arterial topographic anatomy of the dissected palatine ascending and pharyngeal ascending artery was completely analyzed by an anatomical and maxillofacial specialist down to the submicroscopic level. The arterial diameters were measured at two points (D1, D2) for each artery including minimum, maximum, mean values and standard deviations. **(Tab. 1)**

Moreover, the origins of these arteries were systematically investigated: In 17 specimens (57%) the ascending palatine artery branched off from the facial artery, in 7 specimens (23%) from the external carotid artery, in 2 cases (7%) from the ascending pharyngeal artery and in 1 case (3%) from the lingual artery. **(Tab. 2)**

The ascending pharyngeal artery originated from the external carotid artery in 24 specimens (80%), in 3 specimens (10%) from the occipital artery, in 2 cases (7%) from the carotid bifurcation and in 1 specimen (3%) from the internal carotid artery. **(Tab. 3)**

In order the origin of the ascending palatine artery was more variable, than of the ascending pharyngeal artery. The origin from the ascending palatine artery was found at the facial artery in a little more than in half of all cases **(Tab. 2)**, in contrast the ascending pharyngeal artery originated directly from the external carotid artery nearly in most of the cases. **(Tab. 3)**

In 27 specimens (90%) a similar palatine blood supply for the Le Fort I segment was apparent. **(Fig. 2, 3, 4, 5) (Tab. 5)** In these cases the palatine artery (D1: 1,2mm ± 0,34mm; 0,5mm - 1,7mm) emits irregular pharyngeal branches to the pharyngeal constrictor muscles and a variable tonsillar branch to the palatine tonsils. Through these branches, the palatine artery connects to the ascending pharyngeal artery. **(Fig. 5)** The ascending palatine artery reaches the level of the soft palate (D2: 0,8mm ± 0,34mm; 0,4mm - 1,6mm) between the levator veli palatine muscle and the tensor veli palatini muscles. **(Fig. 2)** Covered by the palatine aponeurosis, the ascending palatine artery spreads to smaller branches and curves downward to reach the uvular muscle. Several branches of the ascending palatine artery run ventral to the osseous palate to penetrate it or to continue into connective tissue below the palatine plate. **(Fig. 4)**

Beneath the levator veli palatini muscle, a bundle of small branches arises from the ascending palatine artery and passes upward to the auditory tube and the pharyngeal roof. **(Fig. 2, 5)** These

branches usually communicate with the anterior branch of the ascending pharyngeal artery. The ascending pharyngeal artery (D1: 1,3mm ± 0,58mm; 0,4mm - 2,3mm) supplies the pharyngeal muscles and mucosa. **(Fig. 4, 5) (Tab. 5)** In all specimens several thin arterial branches (in the majority of specimens D2 <0,4mm) from the pharyngeal wall reach the soft palate. In the parapharyngeal spatium, the ascending pharyngeal artery spreads to its terminal branches. The most anterior terminal branch reaches the cranial base, where it is fixed through connective tissue and turns downward to reach the auditory tube. **(Fig. 5)** The anterior branch reaches the soft palate via the tubal anastomotic net or communicates with the ascending palatine artery through thin branches (D2 <0.4mm). The diameter of the branches of the ascending pharyngeal artery that reach the palate are smaller than of the branches of the ascending palatine artery.

In the cases were a similar palatine blood supply for the Le Fort I segment could be identified (27 specimens, 90%) most of the branches of the ascending pharyngeal artery, the arterial net of the pharyngeal wall (D2 <0.4mm) and the anterior branch all contributed together to the blood supply of the Le Fort I segment.

However in 3 specimens (10%) the ascending palatine artery was rudimentary or absent. More precisely a special constellation of the arterial blood supply in which the missing ascending palatine artery was replaced with the ascending pharyngeal artery was found in these 3 specimens (10%). **(Fig. 6, 7, 8) (Tab. 6)** In these cases, only a rudimentary ascending palatine artery which terminated at the level of the styloid muscles, branched off from the facial artery. The missing ascending palatine artery was replaced by a well-developed anterior branch of the ascending pharyngeal artery (D1: 1,9 mm ± 0,32mm; 1,5mm - 2,1mm), which branched off from the external carotid artery.

In this special constellation, the comparatively prominent ascending pharyngeal artery spreads to its terminal branches, located adjacent to the pharyngeal muscles. The most anterior terminal branch ascends just below the cranial base, where it is fixed to the cranial base with connective tissue. The anterior branch bends downward once more and reaches the palate (D2: 1,0mm; ± 0,3mm; 0,6mm - 1,2mm) adjacent to the levator veli palatini muscle, where it spreads in the same manner as the ascending palatine artery. **(Fig. 7)**

In 10% of all cases this special type of arterial variation was found, in which the route of the anterior branch (from its origin up to the cranial base and down again to the soft palate) can be described as a more or less S-shaped curve formed by the artery, that turns about 275° degrees. **(Fig. 6, 7)**

The difference values of the arterial diameters of the main arteries supplying the Le Fort I segment were calculated comparatively at the measurement point D2 at the level of the soft palate, directly next to the Le Fort I segment. According to the categorized arterial vascularization pattern **(Tab. 5, 6)** the difference values were calculated to be D2: 0,2mm ± 0,4mm; 0,2mm - 0,4mm. **(Tab. 7)**

Discussion

The aim of this study was to evaluate the Le Fort I segment's blood supply, since the anatomy of specific arterial variants that may clinically lead to a reduced blood circulation (hypoperfusion) or vascular complications such as ischemic avascular necrosis especially after surgery hasn't been described yet.

In this study we considered the use of Thiel's embalming method for high quality cadaver preservation to be superior to the widely used Formalin preservation. This embalming method preserves soft and hard tissues in a life-like condition concerning natural color, tissue flexibility and consistency (Peuker et al., 2001; Benkhadra et al., 2009). Therefore the tissue's consistence can be compared to the clinical situation (Peuker et al., 2001; Benkhadra et al., 2009). Arterial dissection down to the submicroscopic level becomes also feasible within the use of this method (Thiel, 1992b; Thiel, 1992a).

We evaluated the arterial anatomy of the Le Fort I segment by dissecting the arteries responsible for the segment's blood supply and measured the diameters at two points using Hillen's method. (Hillen, 1986a)

As known from literature, there is a wide range of variability in the external carotid branches (Lippert and Papst, 1985). The external carotid branches can give rise to each other, share a common origin, exchange their supplying territory or even be absent (Hafferl and Thiel, 1969). This was also found in our study. We hypothesized that some of these arterial variants might have consequences for a reduced blood supply of the Le Fort I segment. Accordingly, to respect these anatomical variations the dissection process was performed to the entirety of external carotid branches in the carotid triangle, in the parapharyngeal spatium and the temporal fossa. We also considered variable or aberrant arteries through the examination of all external carotid branches performed by an anatomical and a maxillofacial specialist.

Siebert et al. already described a commonly-known type of the Le Fort I segment's arterial blood supply. (Siebert et al., 1997; Gauthier et al., 2002) Gauthier et al partially confirmed these findings but however claimed for further investigations in this field due to their guess of more existing types of arterial blood supply of the palate (Gauthier et al., 2002).

Siebert et al.(Siebert et al., 1997) examined the Le Fort I segment's arteries using 10 cadaver phantoms. Their findings were similar to ours when they described a commonly-occurring type of

arterial blood supply for the Le Fort I segment. In our study this common standard type was found in the majority of all cases in 90%. The results described by Gauthier et al. (Gauthier et al., 2002) who examined 11 cadaver phantoms partially contradict the findings described by Siebert et al. (Siebert et al., 1997) and also those reported herein. Gauthier et al. described the blood supply of the Le Fort I segment as occurring through the descending palatine artery, the ascending palatine artery and a so-called pharyngeal branch of the ascending pharyngeal artery (Gauthier et al., 2002). This pharyngeal branch appears to be identical to the arteries in the pharyngeal wall described in our study. However, Gauthier et al. denied the existence of an anterior branch of the ascending pharyngeal artery. This could be explained because they harvested the palates from the cranium and the anterior branch is attached to the cranial base.

In their reports neither Siebert et al. nor Gauthier et al. described an arterial variant of the predominant ascending pharyngeal artery, which could be identified amongst others in this study, as a special type of the Le Fort I segment's arterial blood supply. When comparing our findings to the literature, a close relationship was already found between the ascending palatine and ascending pharyngeal arteries (Hafferl and Thiel, 1969), but no exact explanation is reported concerning their variable arterial topography. In that context several authors mentioned the possibility of anatomical variants in the Le Fort I segment's arterial blood supply as a potential risk factor for ischemic complications after surgical treatment such as the Le Fort I osteotomy (Maher, 1977; Lanigan et al., 1990).

Clinically, patients undergoing Le Fort I osteotomies are usually young and healthy adults with a more or less high potential for a fast postoperative recovery. However, patients who have already undergone previous palatine operations and/or those with congenital facial deformities such as cleft lip palates (Maher, 1977; Drommer, 1979) are at greater risk for ischemic complications such as avascular bone necrosis (Lanigan et al., 1990; Bell et al., 1995).

As described in this study, different arterial variations of the ascending palatine and ascending pharyngeal arteries exist. In order, unilateral absence of the ascending palatine artery was found in 10% of the cases and in a similar percentage in the study conducted by Gauthier et al (Gauthier et al., 2002). In fact, a missing ascending palatine artery would be compensated by the contralateral ascending palatine artery or as seen in the 10% of our investigated cases, via the predominant ascending pharyngeal artery (special type arterial blood supply).

However when a Le Fort I osteotomy with an additional surgical segmentation (i.e. a sagittal osteotomy) is performed in a patient with this special type of arterial variation, a segment without a sufficient arterial blood supply may result. In order an arterial compensation cannot be achieved through the contralateral artery after sagittal osteotomy. Additionally in this special type of arterial variation, no evidence exists whether the blood supply through the anterior branch of the ascending pharyngeal artery alone is sufficient enough for an adequate arterial circulation in the Le Fort I segment.

Therefore clinically important may be the S-shaped form of the anterior branch of the ascending pharyngeal artery. **(Fig. 6, 7, 8)** Moreover, when the descending palatine artery is ligated, which may be necessary during an operation, and the ascending palatine artery is absent the blood supply of the whole bone segment becomes dependent only on the S-shaped anterior branch.

More precisely in the commonly occurring standard type of the Le Fort I segment's blood supply, which was found in this study in 90% of the cases, the ascending palatine artery turns approximately 90° to reach the palate. However, in the special type of the arterial blood supply, which occurred in 10% of the cases, the S-shaped anterior branch of the ascending pharyngeal artery is much more curved than the ascending palatine artery in the commonly occurring standard type. In order the anterior branch of the ascending pharyngeal artery turns extensively with about 275° to reach the palate. **(Fig. 7, 8)**

Apart from this extensive change in direction, the anterior branch releases a few smaller branches at its peak below the cranial base. These branches continue upwards to supply cranial structures. The volumes of these smaller branches leading upwards do not contribute to the supply of the Le Fort I segment. Again no evidence exists, whether this S-shaped curve of about 275° leads to more fluid turbulences, arterial thrombosis and/or reduced arterial flow to the periphery, which theoretically could occur in these special cases. If so, this special type of arterial variation would be a potential risk factor for vascular complications such as ischemic avascular necrosis and others. Moreover the curved anterior branch is likely to be more vulnerable to bending forces compared with the arterial standard topography that commonly occurs. This is especially true when the curved anterior branch is mobilized due to surgical procedures as it is the case during a Le Fort I segment osteotomy.

Clinical experience and previous studies have shown that most patients tolerate additional sagittal osteotomy of the Le Fort I segment, without clinical evidence of ischemic avascular necrosis (Quejada et al., 1986; Bell et al., 1995). Moreover, ischemic complications due to Le Fort I osteotomies are reported to be rare with about 1% (Kramer et al., 2004). However, cases of avascular necrosis can clinically still occur and are still reported following an additional segmentation of the Le Fort I segment (Lanigan et al., 1990). This contradiction may be explained by additional factors such as arterial thrombosis or circulation disorders that decompensate an already physiologically limited blood supply, especially in cases with aberrant and compensating arteries that exist in special types of Le Fort I arterial variants. In these cases a surgical induced Le Fort I mobilization leads to a strongly reduced arterial circulation, limited blood flow or hypoperfusion of the palate due to an anatomically altered arterial topography.

Since severe vascular complications in association with Le Fort I segment osteotomies are disastrous for the patient (Kramer et al., 2004), several authors have reported the variable arterial anatomy of the Le Fort I segment as a potential risk factor for vascular complications. (Maher, 1977; Drommer, 1979; Lanigan et al., 1990; Pereira et al., 2010).

On the background of the findings in this investigation the hypothesis of this study that the Le Fort I segment is anatomically not always supported by the commonly known standard type of arterial blood supply can be confirmed for the following reasons: 1) The special type of arterial variation identified in this study and described as a special type of the Le Fort I segment's blood supply is not only an anatomical variation that occurs just in some cases. It is further likely clinically not favorable for the blood supply of the Le Fort I segment especially after surgical interventions. 2) In this special type the pharyngeal arterial system is physiologically reduced to just one artery, instead of three (i.e. due to the absence of the ascending palatine artery and due to possible ligation of the descending palatine artery). 3) The architecture of the supplying artery (S-shaped curve) is more vulnerable to bending and torqueing forces during a therapeutic Le Fort I mobilization. 4) Fluid turbulence and arterial thrombosis which lead to vascular complications are more probable, as the only remaining important artery turns anatomically 275° instead of commonly 90°. These fluid turbulence and arterial thrombosis would probably occur more often when extensive surgical segment mobilizations are performed.

Regarding the comparison of the main arterial diameters between the categorized types of arterial vascularization pattern at the level of the Le Fort I segment (D2) (**Tab. 5, 6**), no direct evidence for a potential topography-based arterial hypoperfusion or diameter related arterial hypoplasia could be found (**Tab. 7**). In order no systemically occurring reduced arterial diameter could be clearly identified in the described special type of the Le Fort I segment's blood supply at the localization of the measurement point D2. This may be the reason because only slightly occurring difference values of the arterial diameters between the two types could be identified. (**Tab. 5, 6, 7**) But due to the number of cases and the used assessment method these findings cannot be completely clarified in this investigation.

In order, we are aware of some limitations in our study. 1) For the reason of high costs and the technique sensitive embalming procedure Thiel's method is only used by a few anatomical institutions (Hayashi et al., 2016). For that reason our dissection results are probably visually less comparable to other investigations when different preservation techniques are used. 2) As a result of the strictly defined inclusion and exclusion criteria a certain number of specimens could not be used in this examination due to their drop out in the selection process. However in this study these inclusion and exclusion criteria were defined to create a homogenous and comparable sample collective. 3) Regarding to the increasing inaccuracy and the missing objectivity in the arterial measurement process the more the measurement is performed in the submicroscopic level, branches <0,4mm were considered to be clinically less important in the Le Fort I segment's blood supply than branches >0,4mm. In order 0,4mm was set to be the lowest level of the measured diameters because of objectivity reasons. 4) In this study the described special type of the Le Fort I segment's blood supply was systemically only found in the minority of the investigated cases. However, although this arterial variant may occur just in some cases, it is still relevant for both the patient and the surgeon due to the potential triggering of severe postoperative complications. 5) For statistical comparison analysis, the use of a larger case series would probably be more effective. However, the analysis of 30 cases, as it was done in this study, is larger than the case series of any other already existing investigation in this field. (Bell et al., 1995; Siebert et al., 1997; Gauthier et al., 2002; Apinhasmit et al., 2005)

In summary, two types of arterial blood supply were categorized in this study. One type was found in most of the cases and is more or less commonly occurring in the Le Fort I blood supply (**Fig. 5**) (**Tab. 5**). This common standard type was already described in the literature by Siebert et al. or Gauthier et al. (Bell et al., 1995; Siebert et al., 1997; Gauthier et al., 2002; Apinhasmit et al., 2005) However, a special altered arterial anatomy that occurred systematically in some cases could be identified. In these cases the absence of the palatine ascending artery and instead a compensatory well developed anterior branch of ascending palatine artery was found. To the best of the author's knowledge this altered arterial anatomy is the first description of a special type of the blood supply of the Le Fort I segment (**Fig. 6, 7, 8**) (**Tab.6**).

Still, clinically ischemic avascular complications following Le Fort I osteotomy are rare (Westwood and Tilson, 1975; Lanigan et al., 1990; Lanigan, 1997; Singh et al., 2008; Pereira et al., 2010; Teemul et al., 2017) and severe complications are very rare, but however the results of this study and the identification of a special type of arterial variation in the Le Fort I segment's blood supply are for the following reasons clinically of high relevance. 1) If such severe vascular complications occur, they are disastrous for the patient (Kramer et al., 2004). 2) The results of this study most likely show the first causal approach to systematically explain the triggered occurrence of severe ischaemic segment necrosis after Le Fort I surgery that were also described by Lanigan et al. (Lanigan et al., 1990) or others (Westwood and Tilson, 1975; Lanigan et al., 1990; Lanigan, 1997; Singh et al., 2008; Pereira et al., 2010; Teemul et al., 2017).

In order in cases with a special type of arterial variation a combination of unfavorable circumstances may decompensate an already limited circulation system and may lead to severe vascular complications and avascular ischemia after surgical Le Fort I segment mobilization. Although no arterial diameter related evidence of hypoperfusion could clearly be verified in this investigation, ischemic complications due to Le Fort I osteotomies (Kramer et al., 2004) may be associated with anatomical variants of a special arterial blood supply. Therefore awareness in Le Fort I related surgical procedures such as osteotomies or segment mobilization procedures is necessary in patients with special types of the Le Fort I segment's blood supply, since these patients are more prone to vascular and ischemic complications than others. This is because of a strong limited arterial potential due to missing arteries that usually additionally supply the palate. In these cases the integrity of the descending palatine artery, which may be ligated during the surgical procedure,

should be ensured since this artery may be a safety factor for the blood supply of the Le Fort I segment.

Unawareness of an individual's arterial anatomy that is associated with special arterial variants is a potential risk factor for avascular necrosis of the Le Fort I segment. This is especially true for orthognathic surgery where a topography-based altered vascular pattern of the Le Fort I segment is more vulnerable to osteotomies or surgical segment mobilization due to an already limited compensatory blood supply mechanism. In order high-risk patients, such as those with cleft lip palates, previous maxillary surgical interventions, thrombotic tendencies, or other congenital facial deformities could benefit from both a particularly indicated preoperative radiological imaging such as magnetic resonance or computer tomography based angiography and an exact evaluation of the operation technique before undergoing orthognathic surgery. Individualized operation planning that takes the patient's arterial anatomy into consideration may help to prevent ischemic vascular complications of the Le Fort I segment and increase the operation outcome in at-risk patients. Therefore additional investigations with larger case series are needed to confirm the findings of this study and further investigate arterial variants of the Le Fort I segment or potential risk factors for vascular complications more precisely.

Ethical approval

Not needed

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Tables

Tab. 1

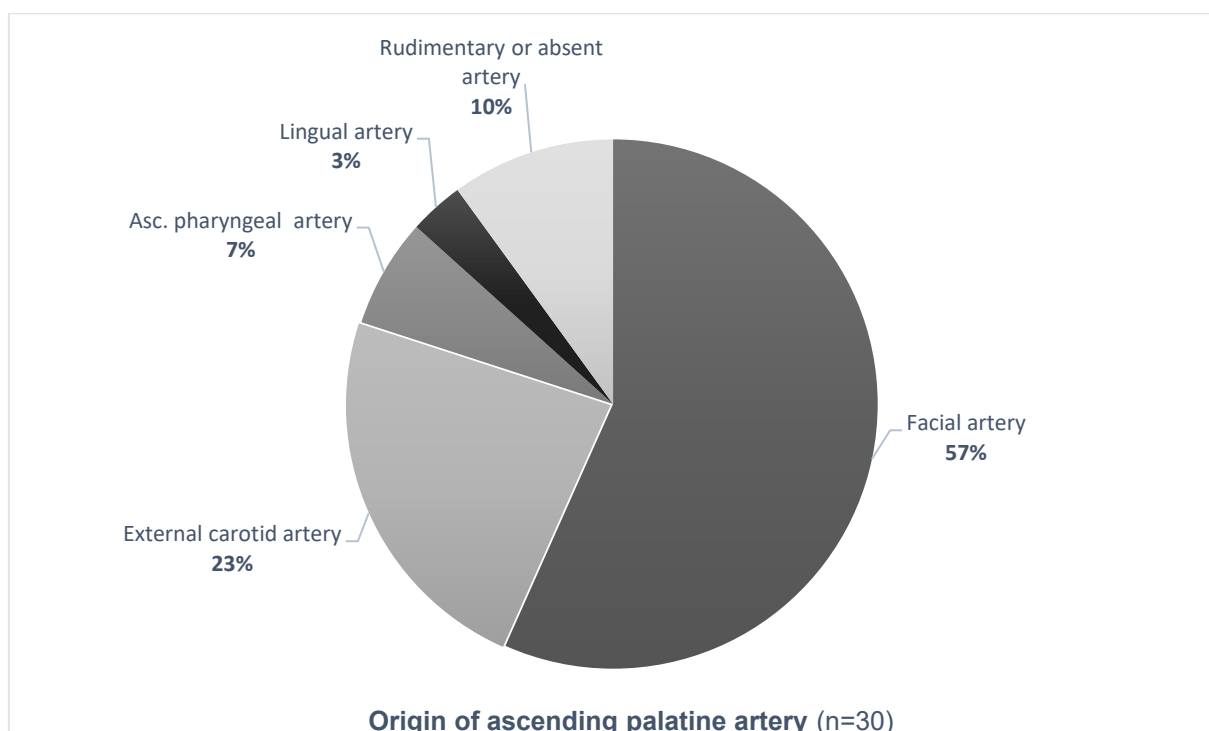
Specimen*/Side*	APA	APA D1	APA D2	APHA	APHA D1	APHA D2	Ra D1	Ra D2
	Origin	Diameter (mm)	Diameter (mm)	Origin	Diameter (mm)	Diameter (mm)	Diameter (mm)	Diameter (mm)
1 l	-	-	-	E	1,9	<0,4	1,3	1,06
2 r	F	1,5	0,9	I	1,0	<0,4	0,7	<0,4
3 l	F	1,4	1,1	O	0,94	<0,4	0,6	<0,4
4 r	F	1,7	1,6	E	1,1	<0,4	<0,4	<0,4
5 l	-	-	-	Bif	1,5	<0,4	0,9	0,6
6 r	F	1,1	0,5	E	0,9	<0,4	0,7	<0,4
7 l	F	1,4	1,3	E	1,6	0,98	0,7	<0,4
8 r	APHA	1,0	0,8	E	0,8	<0,4	<0,4	<0,4
					trippled			
					0,5			
					0,6			
					0,4			
10 r	L	1,1	0,5	E	1,1	<0,4	0,8	<0,4
11 r	F	0,6	0,4	E	1,0	<0,4	0,8	<0,4
12 r	E	1,1	0,9	E	2,2	0,9	<0,4	<0,4
13 l	F	0,8	0,5	E	0,9	0,6	0,6	<0,4
14 l	E	1,0	0,6	E	1,2	<0,4	0,5	<0,4
15 r	E	1,0	1,0	E	1,2	<0,4	0,5	<0,4
16 l	E	1,3	1,2	E	1,1	<0,4	<0,4	<0,4
17 r	APHA	1,6	1,5	E	2,1	<0,4	<0,4	<0,4
18 r	F	1,0	0,9	Bif	0,8	<0,4	0,7	<0,4
19 l	F	1,3	0,5	E	2,0	0,68	0,5	<0,4
		doupled						
		0,5						
		0,5						
21 l	-	-	-	E	2,1	<0,4	1,6	1,24
22 l	E	1,6	1,1	E	1,52	0,6	<0,4	<0,4
23 l	F	1,0	0,4	E	1,14	<0,4	0,5	<0,4
24 l	F	1,6	0,9	E	2,0	0,7	0,6	<0,4
25 r	F	0,9	0,8	E	0,7	<0,4	<0,4	<0,4
26 r	F	1,7	1,1	E	1,7	0,5	<0,4	<0,4
27 l	F	1,2	1,0	E	2,0	0,7	<0,4	<0,4
28 l	F	1,4	1,0	E	1,0	<0,4	0,5	<0,4
29 r	E	1,3	0,8	E	1,1	<0,4	0,5	<0,4
30 r	F	0,8	0,5	O	0,89	<0,4	<0,4	<0,4

Categorization of arterial variants in the Le Fort I segment

According to the dissection process arteries supplying the palate were categorized to identify types of arterial variation. In every case the dissection side of the specimen, the origin of the artery and the inner arterial diameter measured at two points (**D1**, **D2**) were determined. D1 was measured at the origin of the artery, D2 was measured at the arteries' entry into the soft palate (level of the soft palate), which is located directly next to the Le Fort I segment to be the very last endpoint-diameter before the artery enters the Le Fort I segment through the soft palate. *Note: Arterial diameter measurements were limited to a minimum diameter of 0,4mm, due to increasing subjectivity in the measuring process the smaller the diameter gets. Therefore diameters <0,4mm were adopted to be measured not accurately enough*

*Halves of human cadaver heads; **l** = left, **r** = right; **APA** = ascending palatine artery; **APHA** = ascending pharyngeal artery; **Ra** = anterior branch of the APHA; **Bif** = carotid bifurcation; **E** = external carotid artery; **F** = facial artery; **I** = internal carotid artery; **O** = occipital artery; **D1** = inner arterial diameter, at the level of the arterial origin, in the carotid triangle; **D2** = inner arterial diameter, at the level of the soft palate, next to the Le Fort I segment; **gray shading: 1 l; 5 l; 21 l** = cases found with the special type arterial anatomy

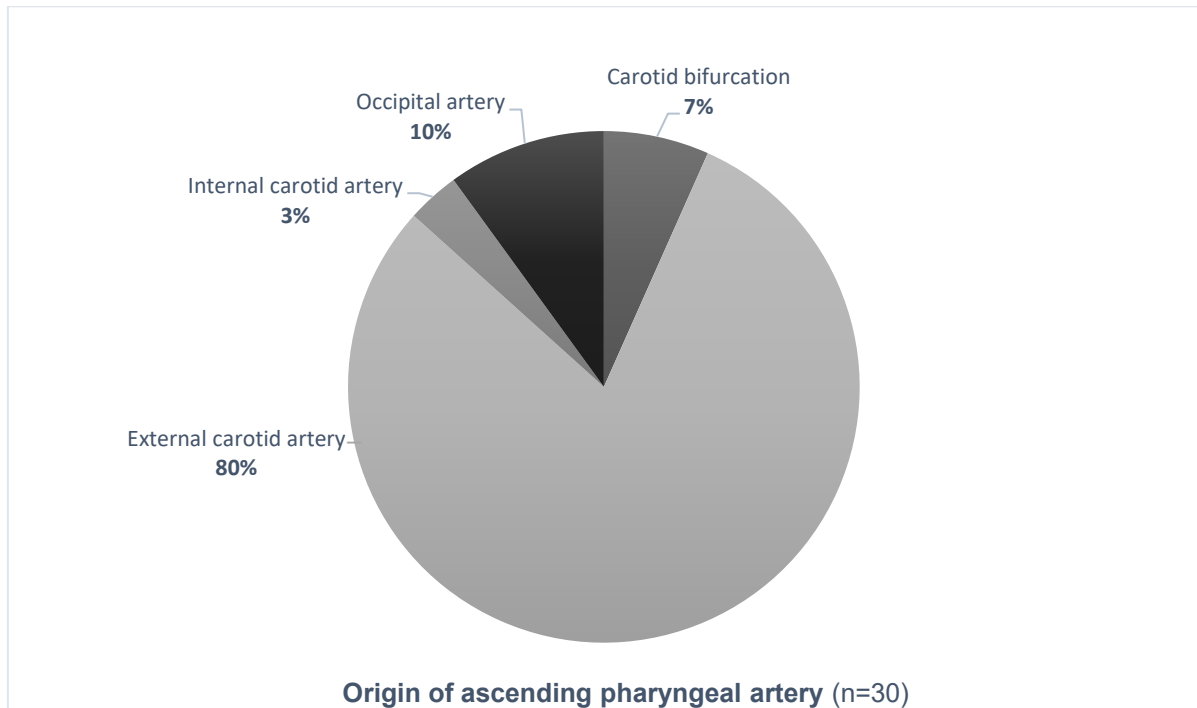
Tab. 2



Origin of ascending pharyngeal artery (n=30)

The origins of the ascending palatine artery were analyzed in order to clearly identify occurring anatomical variations of the Le Fort I segment. Arteries can be identified by their supplying territories and by their offsprings. Several arterial variations of the ascending palatine artery are known from anatomical literature. The ascending pharyngeal artery can be given rise from different external carotid branches or even can be absent. In this study the palatine artery branched off from facial artery in about two thirds, followed by external carotid artery, ascending pharyngeal artery, lingual artery and was absent in the specimens with the anatomy of the special type. **Asc.:** ascending

Tab. 3



Origin of ascending pharyngeal artery (n=30)

The origins of the ascending pharyngeal artery were analyzed in order to clearly identify occurring anatomical variations. It is known from anatomical literature, that the ascending pharyngeal artery can be given rise from different external carotid branches. In the majority of all specimens the ascending pharyngeal artery branched off from external carotid artery, followed by occipital artery, the carotid bifurcation and the internal carotid artery.

**Tab. 4 (see original article: *Journal of Cranio-Maxillo Facial Surgery*
DOI <https://doi.org/10.1016/j.jcms.2018.04.023>)**

Work flow sequence

The trial protocol is shown in a modified CONSORT flow diagram. *Note:* Human cadaver head specimens were selected by the defined inclusion and exclusion criteria. **Asc.:** ascending

Tab. 5

	APA D1	APA D2	APHA D1	APHA D2
	Diameter (mm)	Diameter (mm)	Diameter (mm)	Diameter (mm)
Min	0,5	0,4	0,4	<0,4
Max	1,7	1,6	2,3	<0,4
Mean	1,2	0,8	1,3	<0,4
SD	0,34	0,34	0,58	-

Arterial diameters in the common standard type of the arterial variation in the Le Fort I segment (n=27)

A common standard type of arterial blood supply for the Le Fort I segment was identified in most of the cases (n=27). The arterial diameter in cases with a commonly-known arterial blood supply (common type) was measured at two points (D1,D2). Minimum (Min), maximum (Max), mean values and standard deviations (SD) are shown. For comparison and objectivity reasons in the measurement process diameter measurements were limited to 0.4mm. **APA** = ascending palatine artery; **APHA** = ascending pharyngeal artery

Tab. 6

	APHA D1	Ra D2
	Diameter (mm)	Diameter (mm)
Min	1,5	0,6
Max	2,1	1,2
Mean	1,9	1,0
SD	0,32	0,3

Arterial diameters in the special type of the arterial variation in the Le Fort I segment (n=3)

A special type of arterial blood supply for the Le Fort I segment was identified in some few of the cases (n=3). The arterial diameter in cases with a special variation of arterial blood supply (special type) was measured at two points (D1,D2). Minimum (Min), maximum (Max), mean values and standard deviations (SD) are shown. The ascending pharyngeal artery (APHA) gives rise to neuromeningeal branches (posterior meningeal artery, inferior tympanic artery), pharyngeal branches (smaller branches to the pharyngeal constrictors) and an anterior branch (Ra) which supports to the blood supply of the Le Fort I segment

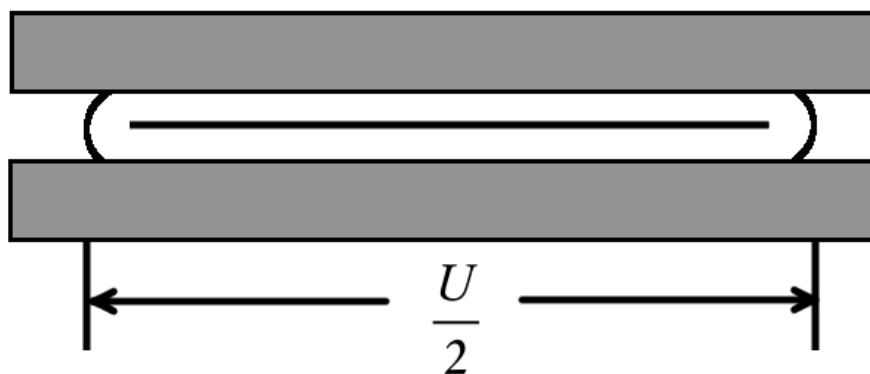
Tab. 7

	APA D2	Ra D2	APA D2 - Ra D2
	Standard- type	Special- type	Difference
	Diameter (mm)	Diameter (mm)	Diameter (mm)
Min	0,4	0,6	0,2
Max	1,6	1,2	0,4
Mean	0,8	1,0	0,2
SD	0,34	0,3	0,04

Difference between standard and special type of the arterial variation in the Le Fort I segment

The main arteries of the Le Fort I Segment are the ascending palatine artery (**APA**) in the standard type of palatine blood supply and the anterior branch of the ascending pharyngeal artery (**Ra**) in the special type of palatine blood supply. The comparison between these main arteries of the special type and the standard type (**APA - Ra**) is expressed by the difference values of the arterial diameter at the level of the soft palate (**D2**). *Note:* D2 is the point of the artery that is located directly next to the Le Fort I segment at the level of the soft palate. The diameter D2 which is most important for the blood supply of the palate and was chosen to be the very last endpoint-diameter before the artery enters the Le Fort I segment through the soft palate.

Illustrations



$$d = \frac{U}{\pi}$$

Figure 1. Cross-section of the measurement process that was used to determine the arterial diameter (modified Hillen's method (Anderhuber et al., 1990)). The artery is pressed with standard anatomical tweezers and the arterial hemi-circumference ($= \frac{c}{2}$) is measured with a Vernier caliper. In order the arterial diameter could be calculated using the standardized method according to Hillen ($d = \frac{c}{\pi}$) (Hillen, 1986a))



Figure 2. Medial view of the palate: The ascending palatine artery (**thin arrow**) divides into smaller branches, and curves downwards to reach the uvular muscle. *Note:* One thin branch is extending upwards to the auditory tube (**triangle**), which communicates with the ascending pharyngeal artery. The canal of the descending palatine artery (**large arrow**) was artificially opened.



Figure 3. Medial view of the palate showing the ascending palatine artery running to the osseous palate.



Figure 4. Posterior view of the pharynx showing the branches of the ascending pharyngeal artery on the pharyngeal wall.

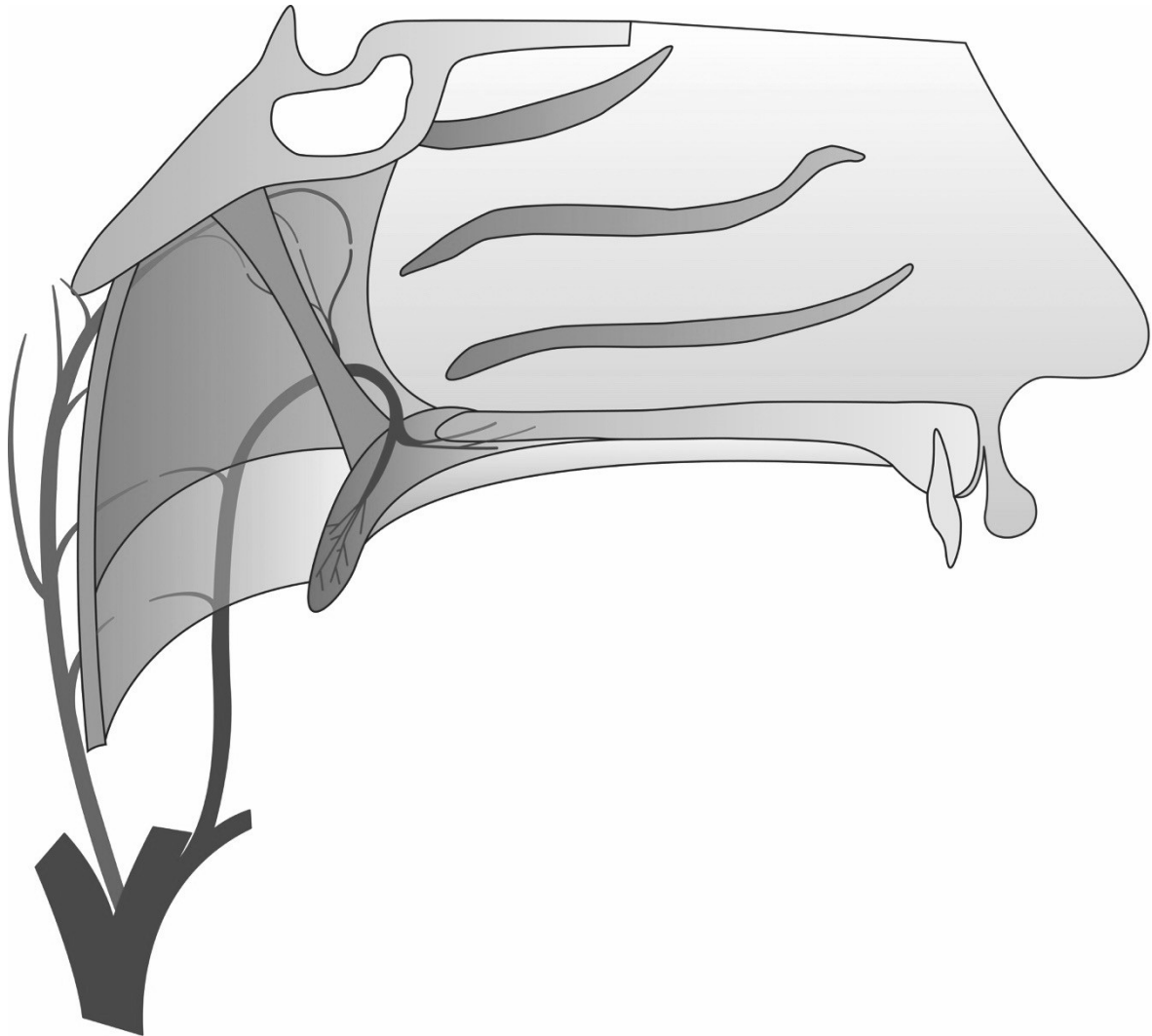


Figure 5. Anatomy of the commonly-known arterial blood supply of the Le Fort I segment (**standard type of arterial variation**), showing the ascending palatine (**gray**) and pharyngeal (**light gray**) arteries. This type occurs in most of the cases. Commonly occurring there is a connection between these two arteries through the pharyngeal arterial net and in the region of the auditory tube.

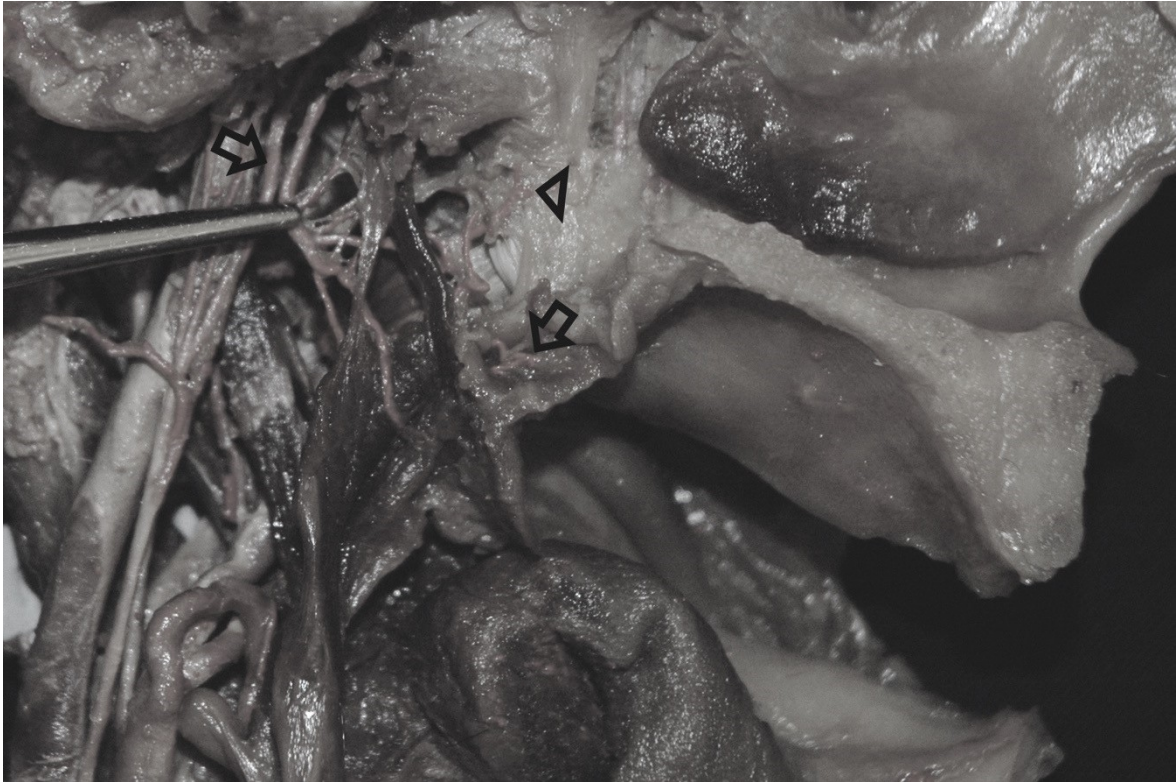


Figure 6. Medial view of the palatine and pharyngeal region with the anatomy of the special type of arterial variation of the Le Fort I segment's blood supply (**special type of arterial variation**). The missing palatine ascending artery is replaced by a well-developed anterior branch of the pharyngeal ascending artery (**open arrows**). This anterior branch reaches the palate next to the levator veli palatine muscle where it branches in the same manner as the ascending palatine artery (i.e. there is a branch up to the auditory tube (**open triangle**))



Figure 7. Inferior-medial view of the palatine and pharyngeal region showing the special type of arterial variation for the Le Fort I segment's blood supply. The anterior branch of the ascending pharyngeal artery (**open arrow**) rises up just below the cranial base, where it is fixed with connective tissue and then curves down to the soft palate. In this special type of arterial variation the S-shaped curve of the anterior branch of the ascending pharyngeal artery, turns about 275°

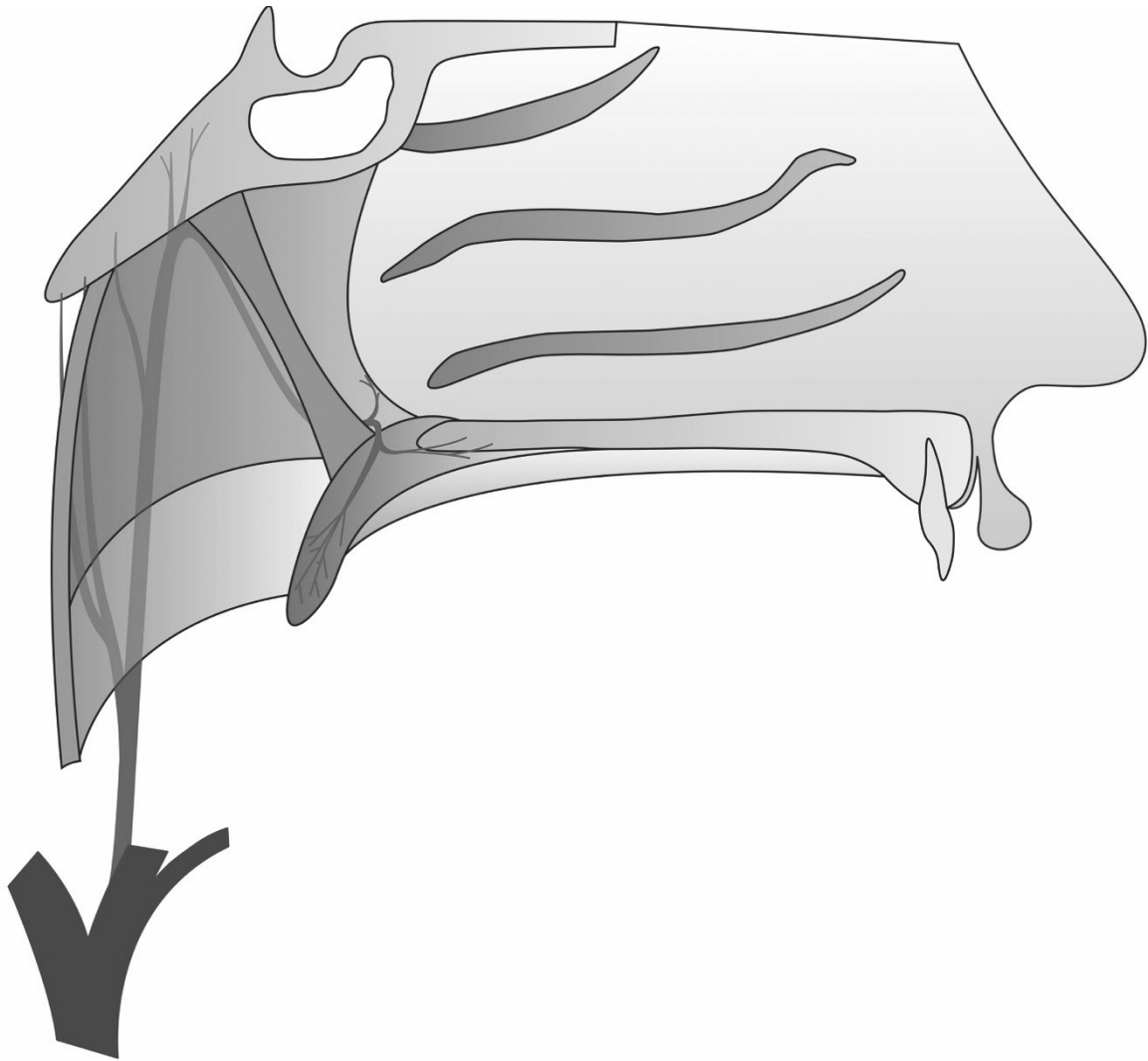


Figure 8. Anatomy of the special type of arterial blood supply of the Le Fort I segment, which was found in this study and only occurred in some cases. The missing palatine ascending artery is replaced by a well-developed anterior branch that branches off from the pharyngeal ascending artery. The pharyngeal ascending artery branches off in the commonly-known manner from the external carotid artery. *Note:* The branches at the apex of the artery continue upwards to the cranial base.

