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CASE REPORT

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Multiple coexisting variations of skull and cervical spine anatomy in a symptomatic patient — uncommon or uncommonly noticed? A CT-based case study

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Background: *Among many anatomical variations of the skull and cervical spine, some may be an underlying cause of a disease, while others remain clinically silent. The estimated individual prevalences of them differ, but given the available data it is impossible to clarify how frequently they coexist. The following study presents an example of 7 anomalies, amongst which at least few manifested clinically in the examined patient.*

Case report: *A 35-year-old Polish woman who suffered from chronic sinusitis was subjected to a computed tomography scan. Mild thickening of the anterior ethmoidal cells' mucosa, bilateral concha bullosa, paradoxical right middle turbinate, bilateral uncinate process pneumatisation, and arrested pneumatisation of sphenoid sinus were found and addressed in the context of the reported symptoms. Simultaneously, other, clinically silent anatomical anomalies were found — namely ossification of the anterior petroclinoid ligament, incomplete medial basal canal, and bilateral arcuate foramen.*

Conclusions: *To the best knowledge of the authors, this case report is the first to present such a coexistence of this many anatomical anomalies, some of which played a crucial role in the chronic sinusitis experienced by the patient. Concurrence of multiple variations in the same anatomical area or functional unit may exacerbate clinical presentation of a patient. Identifying a single anomaly ought to warrant a thorough investigation into any other potentially existing variants.*
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Keywords: chronic sinusitis, concha bullosa, paradoxical middle turbinate, arrested pneumatization, anatomy, radiology

INTRODUCTION

There are numerous anatomical variations that can obstruct air flow in the nasal cavity and outflow of discharge from the paranasal sinuses (PNS), causing symptoms which can be attributed to chronic sinusitis. In the long-term it can produce favourable conditions for chronic inflammation and can be a causal factor for rhinosinusitis [8, 26].

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The osteomeatal complex (OMC) is defined as part of the nasal cavity in which ostia of the frontal sinus, maxillary sinus, and ethmoidal infundibulum drain into the middle nasal meatus, laterally to the middle nasal concha (MNC). The destination for their openings is called the hiatus semilunaris (HS). It is a crescent-shaped groove bound by the ethmoid bulla, ethmoidal process of the inferior nasal concha, and uncinate process of the ethmoid bone (UP). Aberrant construction of the OMC components may lead to impairment of PNS ventilation and obstruction of the middle nasal meatus [12].

Concha bullosa (CB) is a pneumatised turbinate, most commonly involving the MNC, and its prevalence varies between 9% in non-rhinosinusitis patients [10] and up to 76% in patients with chronic rhinosinusitis [37]. It can be divided into 6 types based on the localisation and expansion of its aeration [11]. The other common anomaly of the MNC is the so-called paradoxical middle turbinate (PMT). In most cases, the MNC is medially convex. When the curvature is reversed and the concavity of the meatus is facing the nasal septum, the term PMT can be used. In this instance, the convexity of the turbinate is facing the HS and therefore may also contribute towards narrowing of the OMC [12].

Deformation, hypertrophy, hypotrophy, or pneumatisation of the anteroinferior boundary of the HS, namely the uncinate process, can lead to obstruction of the ethmoidal infundibulum and therefore the anterior ethmoidal cells. UP pneumatisation may be present in approximately 2.5% of the population [8].

During the ontogeny of the nasal cavity and paranasal sinuses, bones undergo an early fatty marrow conversion prior to their pneumatisation. When respiratory mucosa covers the area before the aeration process is complete, a persistent compartment filled with the fatty marrow is created, termed as arrested pneumatisation (AP). It most commonly occurs in the sphenoid sinus. The reasons behind this phenomenon are elusive and are still debated in the literature [16, 40].

Petroclinoid ligaments are folds of the dura mater connecting the anterior clinoid process with the apex of the petrous pyramid. The posterior clinoid process divides the fold into the anterior and posterior ligament, both of them limiting the hypophyseal area and cavernous sinus [33]. A recent meta-analysis estimated the prevalence of petroclinoid ligaments calcification to be 10% [30].

The medial basal canal (MBC), also known as the (medial) clival canal, is a term describing various forms of midline basiocciput communications involving either one or both surfaces of the midline portion of the clivus. It can be divided into 6 groups: 3 complete forms (including all types of a complete canal with at least 2 openings on one or two sides of basiocciput) or incomplete (in a form of recess on superior or inferior surface of clivus) [13]. It needs to be distinguished from the craniopharyngeal canal (conversely the persistent hypophyseal canal), which communicates the ventral surface of the clivus with the floor of the pituitary fossa and is most probably a result of incomplete closure of Rathke's pouch [1].

The ossified inferior margin of the posterior atlanto-occipital membrane is called the ponticulus posticus, which can reshape the groove for the vertebral artery into the arcuate foramen (FA). The contemporary classification divides the FA into complete (when the ponticulus posticus converts the groove into the foramen) and incomplete (when the groove is covered only superiorly, and the foramen is opened posteriorly). A recent meta-analysis estimated the prevalence of the complete FA to be 9.1%, but only in about one-third of those cases was complete FA found bilaterally. The incomplete anomaly was present in approximately 13.6% of the population [28].

The purpose of this case report was to present a unique description of numerous anatomical anomalies of the PNS found in a single patient, whose clinical symptoms could result from their synergistic restrictive effect on the OMC.

CASE REPORT

The following study presents multiple co-existing anatomical variants of skull and cervical spine encountered during a computed tomography (CT) scan of a Polish 35 y.o. woman. She was examined due to chronic sinusitis, nasal obstruction, and headaches in February 2023. The examination showed mild thickening of the anterior ethmoidal cells' mucosa, bilateral concha bullosa (type 2 with anteromedial aeration), paradoxical right middle turbinate (in the form of a recess on the medial surface of the concha), and bilateral uncinate process pneumatisation. All the above may have contributed to the aforementioned symptoms due to their restrictive effect upon the HS (Fig. 1–3).

After further examination, other anomalies unrelated to the presented symptoms were spotted. The first of them was ossified right anterior petroclinoid

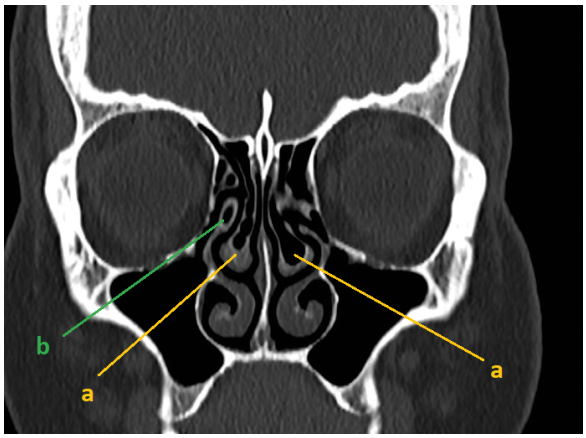

Figure 1. A CT scan of the skull, frontal plane; a — bilateral concha bullosa; b — pneumatisation of the right uncinate process.

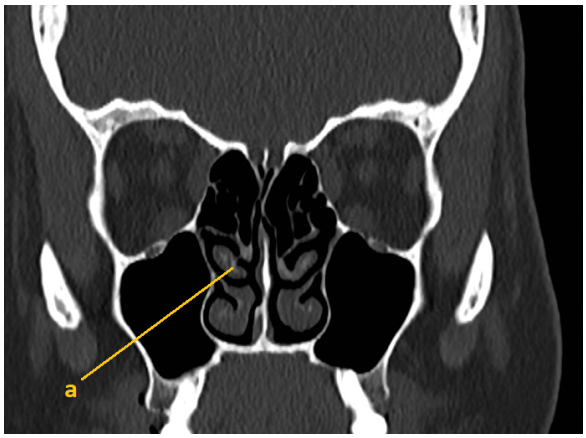

Figure 2. A CT scan of the skull, frontal plane; a — paradoxical right middle turbinate.

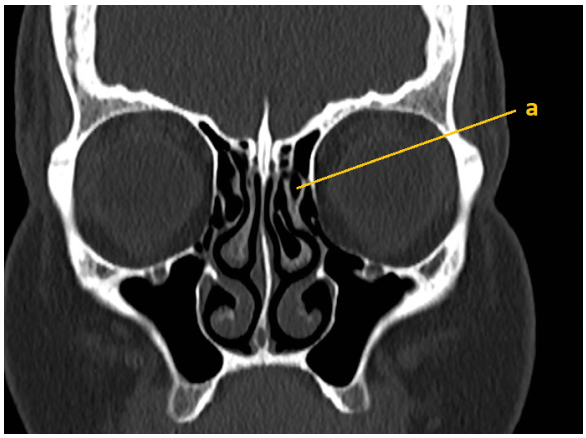

Figure 3. A CT scan of the skull, frontal plane; a — pneumatisation of the left uncinate process.

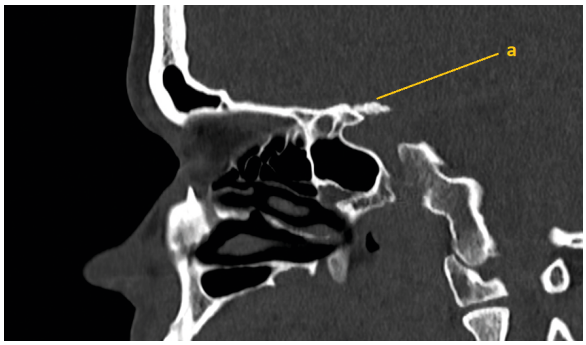

Figure 4. A CT scan of the skull, sagittal plane; a — ossification of the right anterior petroclinoid ligament.

ligament (11 mm length, 5 mm in width), reaching the plane of dorsum sellae, adjacent to the right posterior clinoid process. Its solid structure made it look like a prolonged anterior clinoid process (Fig. 4, 5).

The second finding was the incomplete MBC in the form of inferior recess in the basiocciput. Its tubular structure inclined us not to classify it as pharyngeal fossa, although the choice of description is arbitrary (Fig. 6).

The third aberration in the anatomical structure of the skull was incomplete AP of the right sphenoid sinus. Two compartments of fatty marrow were present: the first at the base of the right pterygoid process, and the second in the posterior part of the right sphenoid sinus (Fig. 7, 8).

Lastly, the fourth anomaly, concerning first cervical vertebra (C1, the atlas) was bilateral, complete arcuate foramen (Fig. 9).

No further variations from the so-called anatomical norm were noted upon the CT examination.

DISCUSSION

It has been proposed that in the case of recurring or refractory rhinosinusitis an underlying cause is usually stenosis of the OMC [36]. Few prospective studies of patients with chronic sinusitis noted increased incidence of the OMC deviations. Sonone et al. [35] reported that among their patients over 75% showed abnormalities in the uncinate process structure, while Tiwari et al. [37] showed a similar prevalence of concha bullosa. Although incidences of particular anomalies were different in both the aforementioned studies, their limited sample size in combination with various possible structural abnormalities may explain the inaccuracies between them. Nevertheless, their common conclusion is increased

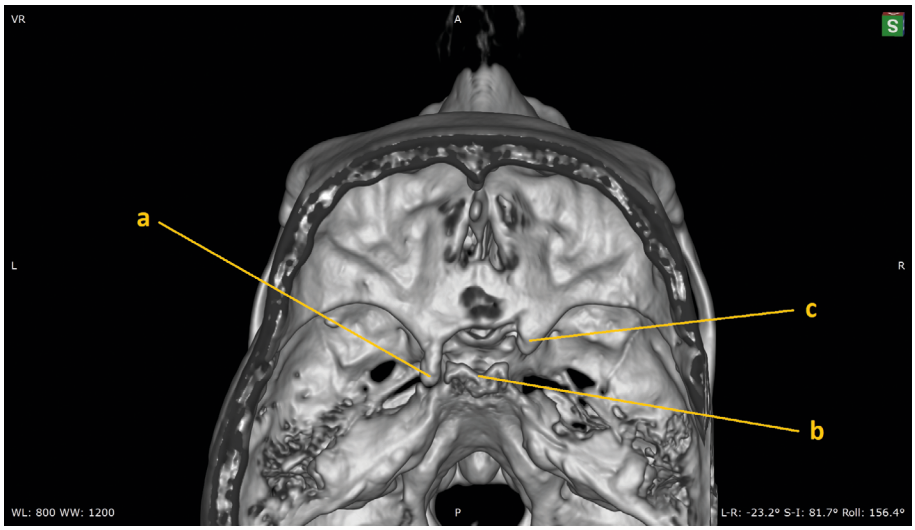

Figure 5. A 3D reconstruction of the skull, superior view; a — ossification of the anterior petroclinoid ligament; b — dorsum sellae; c — left anterior clinoid process.

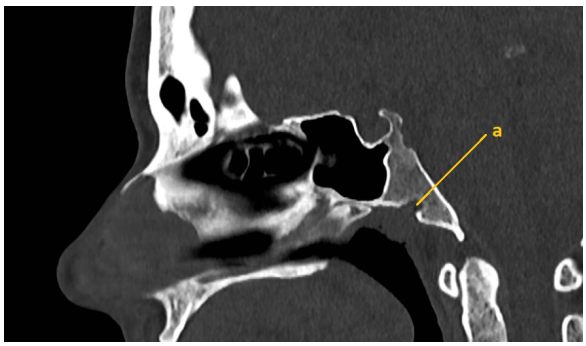

Figure 6. A CT scan of the skull, sagittal plane; a — median basilar canal.

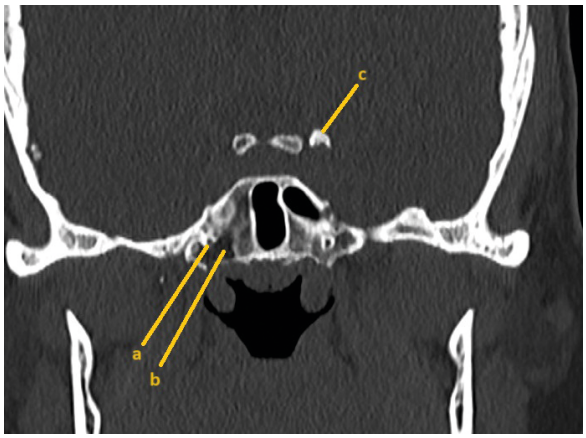

Figure 7. A CT scan of the skull, frontal plane; a — right Vidian canal; b — arrested pneumatisation in the right pterygoid process; c — ossification of the left anterior petroclinoid ligament.

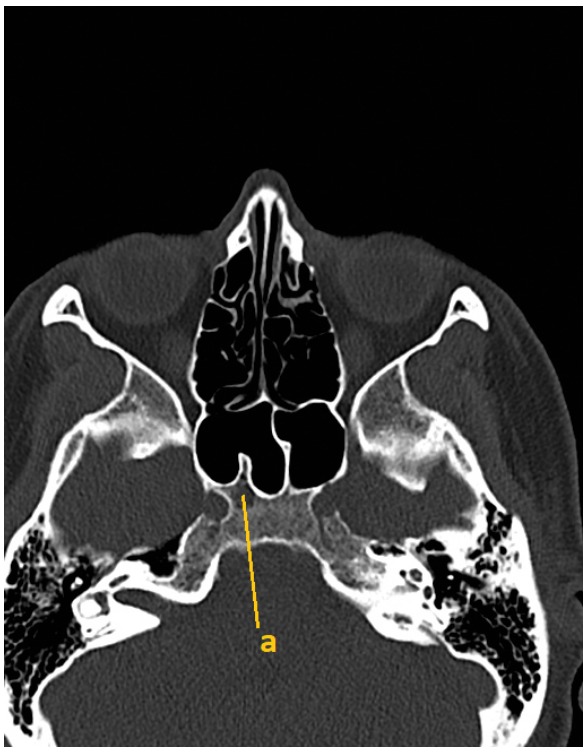

Figure 8. A CT scan of the skull, transverse plane; a — arrested pneumatisation in the posterior part of the right sphenoid sinus.

frequency of variants in the structure of the OMC that seems to be linked to disease development. It is worth remembering that in most of the cases it is impossible to point towards one definitive causative factor

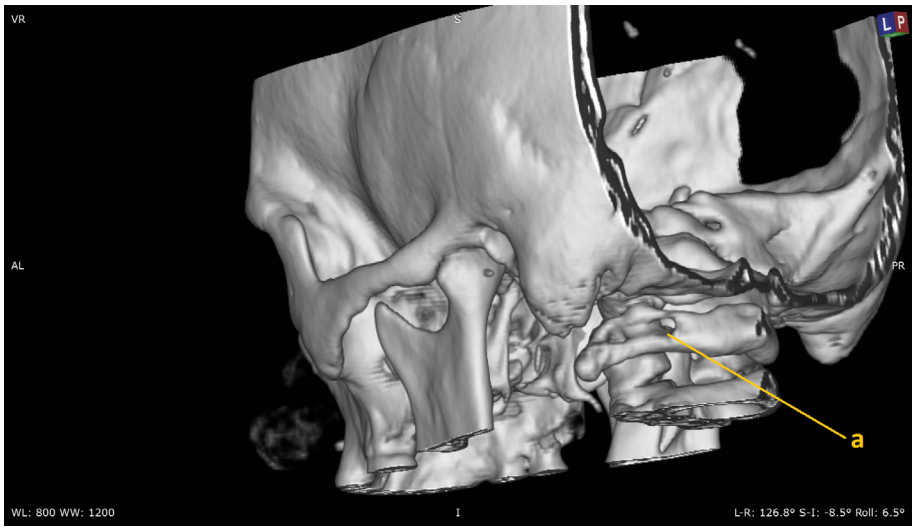

Figure 9. A 3D reconstruction of the skull and cervical spine, left posterolateral view; a — left arcuate foramen.

leading to sinusitis. Apart from stenotic disruption of sinus drainage, abnormal mucociliary transport, allergic hypersecretion, immune deficiency, or local dysbiosis may be a significant comorbidity influencing the clinical picture [17].

In the present case, chronic sinusitis and nose obstruction may have been caused or aggravated by the bilateral CB with PMT and pneumatised UP. Due to their synergistic effect on impairing ventilation and drainage of the maxillary sinus, frontal sinus, and anterior sphenoid cells, those 3 could be called a "tandem anomaly", as the authors have proposed in another paper. It is crucial to remember that the clinical importance of individual variation depends on its correlation with the overall anatomy and coexisting anomalies, and therefore single variation should often not be considered as a causal factor but a risk factor.

Even though the CB seems to be more commonly found in patients with sinusitis than healthy individuals [5, 34], some authors did not find a statistically significant correlation between the CB and sinusitis, unless the turbinate is large, unilateral, and associated with septal defect [3, 21]. In this context, it is worth mentioning that a higher prevalence of the CB was found among patients with septal deviation [42]. In this case another MNC anomaly, the PMT, may have exacerbated the potential effect of the CB because its paradoxical convexity was facing the HS.

The UP contributes to creating the HS, into which the ethmoidal infundibulum, and maxillary and frontal sinus drain. Therefore, its aberrant anatomy, including pneumatisation, may contribute to their impaired drainage, and ultimately to inflammation [5, 8]. An anatomical study of surgical accessibility of maxillary sinus ostium showed that in 20% of specimens the uncinate process or concha bullosa made maxillary sinus cannulation impossible, which proves their significance [39]. Among possible techniques of functional endoscopic sinus surgery (FESS), excision or crushing of concha bullosa and uncinectomy are used to improve the OMC patency [2, 4].

The clinical importance of the AP can be described in 2 ways. Firstly, it needs to be differentiated with both malignant and benign skull base tumours, such as chondrosarcoma, chondroma, lipoma, haemangioma, or metastases [14]. Important markers of an invasive disease are the adjacent structures, like the Vidian (pterygoid) canal and inferior orbital fissure. When these anatomical entities are disrupted in any way, they are a strong predictive factor for an aggressive lesion, while if unaffected, they can point towards the AP. Other characteristic features include well-defined sclerotic margins and fatty content with curvilinear calcifications. The AP must be limited to areas of normal or accessory pneumatisation, and it is never expansive. The above-mentioned features were gathered in Welker's criteria and are widely accepted [16, 18, 27, 40].

Secondly, according to some studies, the prevalence of AP is significantly higher among patients with some haematological conditions, namely sickle cell anaemia and thalassaemia [6, 7, 31]. This supports the hypothesis that the driving cause for AP is regional disturbance of blood perfusion, which can be

attributed to the abovementioned diseases. The other considered mechanisms involve the timing of the sinus aeration and ratio of cortical and trabecular bone [7].

The clinical significance of the MBC depends vastly on its type and co-existing pathologies. A case study presenting cerebrospinal fluid leakage through a complete MBC following elective septoplasty provides proof that in unfavourable circumstances this anomaly may cause a life-threatening condition [22]. Incidental cases of recurrent meningitis due to basioccipital meningocele as a complication of a complete MBC were also reported [19]. Nevertheless, incomplete forms of the MBC are considered completely benign, [13] although they might be of some importance in cases of traumatic skull base fracture.

The anterior petroclinoid ligament is a fold of dura mater limiting the basin of the hypophyseal region and laterally the superior wall of the cavernous sinus [33]. Due to its proximity to the oculomotor nerve running through the superior portion of the lateral wall of the cavernous sinus, the calcified ligament may predispose to nerve damage. It was proposed that anterior petroclinoid ligament resection is a sufficient way to gain access to low-lying internal carotid–posterior communicating artery aneurysms and allow for decompression of the oculomotor nerve following the procedure [9, 23]. In case of the fold's ossification, the third cranial nerve may be more prone to injury due to compression and surgical access may pose more difficulties. Most of the available literature is focused on the posterior petroclinoid ligament (also termed as the petroclival or Gruber's ligament) calcification due to its proximity to both the oculomotor and abducens nerves [15]. Some case studies also point out the clinical significance of anatomical variations of its attachment. Żytkowski et al. [44] reported a bifid origin of the petroclival ligament, with one bundle attached to the cavernous internal carotid artery, while Wysiadecki et al. [41] reported a case of Gruber's ligament duplication into 2 bundles with abducens nerve and dorsal meningeal artery between them. Significantly less attention has been given to the anterior petroclinoid ligament anomalies. Nevertheless, its ossification may be of some significance in cases of cavernous thrombosis, increased intracranial pressure, skull base injuries, or neurosurgical procedures concerning the hypophyseal area and the cavernous sinus proximity.

The FA in most cases remains clinically silent. Nevertheless, symptoms such as cervicogenic headache, migraine, or vertigo were attributed to this anomaly — some available data confirm this hypothesis by showing that resection of the ponticulus posticus can bring relief from those problems [24, 25].

Evidence-based data from meta-analysis confirmed the relationship between the presence of the FA and headaches. Interestingly, such a relationship was observed in both complete and incomplete FA. Of note, bilateral FA did not significantly increase the incidence of headache compared to the unilateral anomaly [29].

Those observations were also reflected in the results of the cadaveric study by Tubbs et al. [38], which confirmed possible compression of the bony structure of the FA on the vertebral artery. Existence of the FA must be acknowledged by orthopaedic surgeons performing C-1 lateral mass screw, because it may cause misevaluation of the posterior arch anatomy leading to its misalignment that could put the vertebral artery at risk of an iatrogenic injury [43].

The main limitation of the present study was its study type (case report), retrospective character, and incomplete knowledge about the patient's full medical history (especially the subsequent treatment). Future studies ought to pay more attention to understanding the possible consequences of multiple coexisting anatomical variants in the clinical context. Moreover, large studies would be required to assess how often anatomical anomalies are omitted in radiological and clinical evaluation due to their mild character and lack of relation to patients' complaints. Lastly, further prospective studies on a large population of patients with chronic sinusitis are required to assess the incidence of individual anomalies and how frequently they coexist compared to the healthy population. The concept of tandem anomalies may also explain why available data on the influence of particular anomalies on the occurrence and severity of chronic sinusitis is not entirely coherent. Henceforth, further studies should focus more on the entirety of the OMC patency rather than just assessing anomalies of its components. However, selection bias of symptomatic patients might distort the true prevalence of coexisting variants [32], although the existence of the so-called "anatomical norm" is disputable [45].

CONCLUSIONS

To the best knowledge of the authors, this is the first case report to present such a variety of anatomical variations coexisting in one patient. Most

of them are clinically silent but need to be differentiated from pathological findings that may require further examination and possible subsequent treatment. Identifying a single anomaly ought to warrant a thorough investigation into any other potentially existing variants.

The reported case raises an imminent question: Are multiple coexisting variations of head and neck anatomy uncommon, or only uncommonly noticed? The authors believe that even seemingly unimportant anomalies should be assessed in the clinical context, and that the medical professionals should bear in mind their potential synergistic effect.

ARTICLE INFORMATION AND DECLARATIONS

Ethics statement

This type of study did not require approval from the local Ethics Committee. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Guidelines set by Henry et al. [20] applied.

Author contributions

Conceptualisation — JJT, BK; project administration — IZ, BK; methodology — IZ, JJT, BK, MPZ; investigation — IZ, JJT, BK; data curation — IZ, JJT, BK; formal analysis — JJT, BK; visualisation — JJT; writing (original draft) — IZ, JJT, BK, MPZ, PP, JAW, JZ, ML; writing (review & editing) — IZ, JJT, BK, MPZ, PP, JAW, JZ, ML; supervision — MPZ, PP, JAW.

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Conflict of interest

The authors declare no conflicting interests.

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