World Journal of Surgery

Journal Homepage: surgeryworld.org

Original Article

A Single-Center Prospective Comparison of Natal Cleft Depth of 200 Patients with and without Pilonidal Sinus Disease

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ARTICLE INFO

Article history:
Received: 26 August, 2023
Accepted: 14 September, 2023
Published: 4 October, 2023

Keywords:
Intergluteal fold depth, natal cleft, pilonidal sinus, mechanism of disease, PSD

ABSTRACT

The etiology of primary pilonidal sinus disease (PSD) remains unclear. Current understanding suggests that sharp hair fragments from the occiput contribute to the formation of PSD. In 2009, Akinci *et al.* reported a correlation between PSD and a deeper natal cleft. We investigated the association between intergluteal fold (IGF) depth and PSD risk using a standardized five-step measuring protocol. Our study included 95 PSD patients and 105 non-PSD individuals, and measurements were taken from the glabella sacralis to the anus in a controlled in-house setting after obtaining informed consent from voluntary participants of a northern German population. The mean (± SD) intergluteal depth progressively increased from the intergluteal opening at the glabella sacralis at 9.1 (± 3.4) mm to a maximum of 62.6 (± 10.4) mm. Notably the deepest point was consistently observed at the anus, where PSD occurence is rare. No significant difference in intergluteal fold depth between PSD and non-PSD patients was found. Additionally, PSD predominantly developed in the proximal (cranial) third of the intergluteal fold, despite the maximum depth being in the distal region. These findings suggest that intergluteal fold depth is not a risk factor for PSD.

1. Introduction

The mechanism of primary pilonidal sinus disease (PSD) has been a concern for medical professionals since 1833. Numerous theories about risk factors and prevention have emerged due to PSD's midline appearance in the lumbar region, glabella sacralis, and cranial opening of the intergluteal fold (IGF). Embryologic origins, such as remnants of the preen gland [1], residual human tail [2], neuro-cutaneous traction [3], faulty ectodermal closure [4], and gluteal muscle involvement [5] have been discussed. During World War II, the US American military was the first army to have large motorized unit, where even the common soldiers had their cars. When more than 17.000 soldiers succumbed to PSD, Buie proposed that the disease was acquired from driving in hard seats on bumpy roads, coining the term "Jeeps disease" [6]. Subsequent research disproved the connection to automotive factors [7].

This resulted in speculations about the acquired reasons for PSD: higher BMI, faulty hygiene, enhanced sweating and hormonal imbalances [8] have not been definitively proven or ruled out. The folliculitis theory [9], ingrowing gluteal hair theory or ruptured hair from elsewhere piercing into the skin had to be discarded due to lack of supporting evidence. Recent studies have shown that sharp hair fragments constitute the primary component of the pilonidal sinus nest [10]; particularly from the occiput [11]. Electron microscopy images [12] have captured hair strands erecting themselves and piercing the skin of the upper (cranial) IGF when a sharp edge is in proximity and hair scales point away from the skin. A hairy IGF seems to hold hair longer into position, explaining that hairy individuals appear to have a higher susceptibility to PSD.

In 2009, Akinci *et al.* reported a correlation between PSD and deeper natal clefts, suggesting implications for the choice of the surgical

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DOI: 10.60123/j.WJS.2023.20.05

procedure [13]. Understanding that a steeper upper natal cleft would erect hair more forcefully, we wondered why a deeper natal cleft could add major risk to PSD. To investigate further we developed a standardized measuring protocol to asses natal cleft depth in both PSD and non PSD individuals.

Our study aims to address the key questions: the most reliable method for measuring the IGF depth, the presence of one, or multiple "deepest points" within the IGF, and potential differences in maximum IGF depth between genders and PSD status. Ultimately, our goal is to determine if IGF depth is an independent risk factor in pilonial sinus disease. Our null hypothesis: There is no association between IGF depth and PSD risk, and IGF depth does not serve as an independent risk factor for PSD.

2. Methods

2.1. Patients

A total of 200 participants from a normal population in northern Germany were included in this study. Participants were required to provide informed consent and sign the study protocol before being included in this study. Among the participants, 95 individuals (47.5%) were diagnosed with PSD, while 105 individuals (52.5%) were non PSD-patients.

The study population consisted of 125 males (62.5%) and 75 females (37.5%). The age of the participants ranged from 16 to 81 years, with a mean (±SD) age of 37.7 (±15.5) years. The participants' body mass index (BMI) had a mean (±SD) value of 27.2 (±5.1) kg/m^2. The range of BMI values was 17.8 - 50.6 kg/m^2. Figure 1 presents histograms depicting the distribution of age and BMI in the study population.

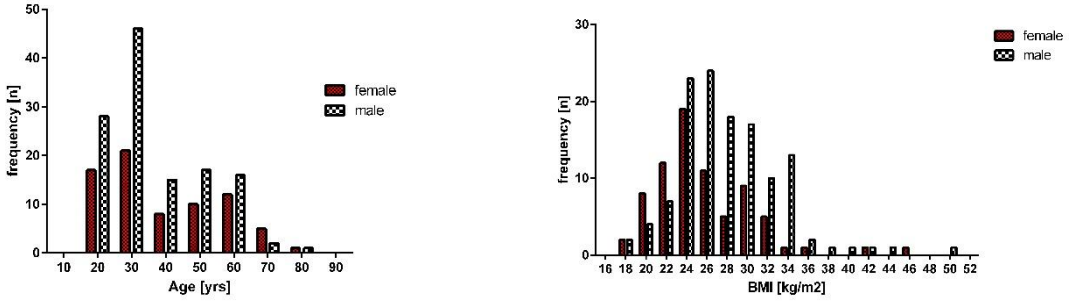

FIGURE 1: Age (left side) and BMI (right side) distribution in n=200 patients*.
(*One woman with normal BMI declined to give her age, so age graph was drawn with n=199 patients).

2.2. Measuring Tool

A measuring tool was developed based on Akincis research. The tool consists of a carbon electronic measuring scale, securely attached rectangularly to a lightweight alloy plate with a contact area of 20 × 3 cm, weighing 118 g in total.

2.3. Measuring Procedure

To gauge IGF depth, the distance from the upper opening of the intergluteal fold (with a sub-3 mm diameter) to the IGF's end at the anus was evaluated. This span was split into four equal parts, resulting in five measuring points. These were indicated using water soluble pen marks. Measurements were taken at positions "a" (cranial opening), position "e" (end of IGF/anus), and the three marked positions (Figure 2). The procedure involved gently placing the tool's thin alloy plate over both buttocks without pressure, as patients lay prone on their belly. The plastic lever was then gently lowered until the midline's IGF depth was visually identified. For precision, each IGF depth at positions "a" to "e" underwent five measurements, recorded, and the mean was used for analysis. The instrument was always positioned perpendicular to the sacral bone to measure skin-to-midline distance, as depicted in (Figure 2).

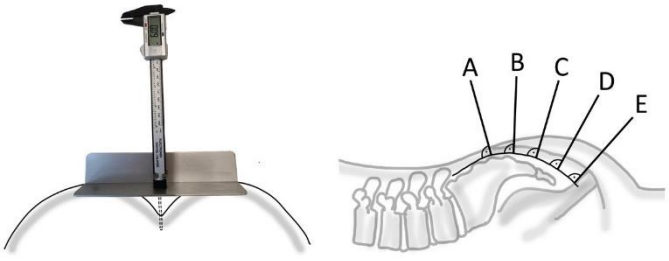

FIGURE 2: The left side of the figure shows a photo of the lightweight measuring tool used in the study. Please note that the numbers on the tool in the illustration are not correct and are for illustrative purposes only.
On the right side of the figure, there is an illustration demonstrating the measuring points. The measuring tool was positioned at a 90-degree angle to the sacral bone. The distance from the upper opening of the intergluteal fold (point A, with a diameter of less than 3 mm) to the end of the intergluteal fold at the anus (point E) was measured. This length was then divided into four equal parts, resulting in the identification of three additional points: B, C, and D.

2.4. Statistical Analysis

The study data was recorded in an excel-sheet (Excel 2016, Microsoft Corporation, Redmond, WA, USA). Continuous variables are presented as mean ± standard deviation. Categorical variables are expressed as proportions and analyzed using fishers t-test. All statistical analyses were performed using SPSS version 26.0 (IBM Corp., Armonk, NY, USA).

2.5. Ethics

Ethical approval for this study was obtained from two separate ethics committees. The study received approval from the Ethics Committee of Saarland University Homburg/Saar (Approval No. 59/22) on July 11, 2022, chaired by Prof. Dr. Grundmann. Additionally, approval was granted by the Ethics Committee of the County Ethics Chamber Lower Saxony in Hannover (Approval No. GRAE/151/2022) on August 19, 2022, chaired by Prof. Dr. Creutzig.

While the study methods did not involve any interventions that could potentially harm human participants, obtaining ethical approval was necessary to ensure adherence to relevant guidelines and regulations governing research involving human subjects. The study was conducted in compliance with these guidelines and regulations, prioritizing the welfare and rights of the participants [14].

3. Results

The mean (±SD) length of the intergluteal fold from glabella sacralis to anus was 16.3 (±2.5) mm in males and 15.3 (±2.5) mm in females. The mean IGF depth at different positions was as follows: 9.1 (±3.3) mm at position a; 21.1 (±8.1) mm at position b; 32.4 (±10.1) mm at position c; 45.6 (±10.0) mm at position d, and 61.8 (±10.8) mm at position e (Figure 3). The largest IGF depth was observed between the gluteal muscles at the level of the anal crest. There was no statistically significant differences in IGF depth between males and females (p=0.816; t-test), or between PSD- and non-PSD patients (p=0.833; t-test; (Figure 4)). The effect size of our study was calculated to be a Cohen's d value of 0.0858. The calculated power of the study was 5%.

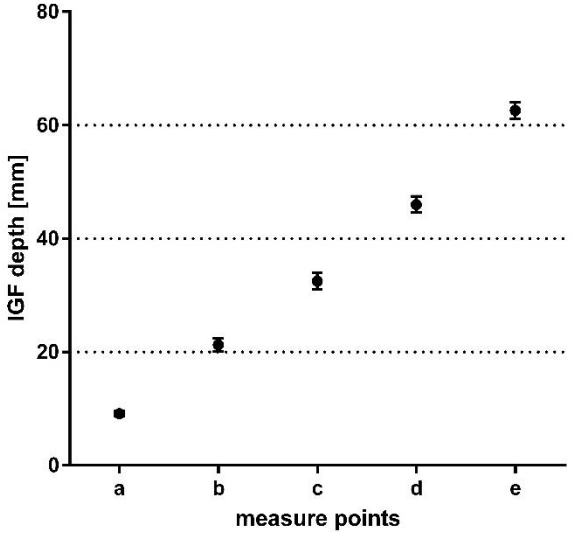

FIGURE 3: Intergluteal fold depth between intergluteal opening cranially down to the distal end of the intergluteal fold at the anus in n=200 patients. Measuring points: (a) cranial opening of intergluteal fold; (b) proximal intergluteal fold; (c) mid intergluteal fold; (d) distal intergluteal fold; (e) caudal end of intergluteal fold close to anus.

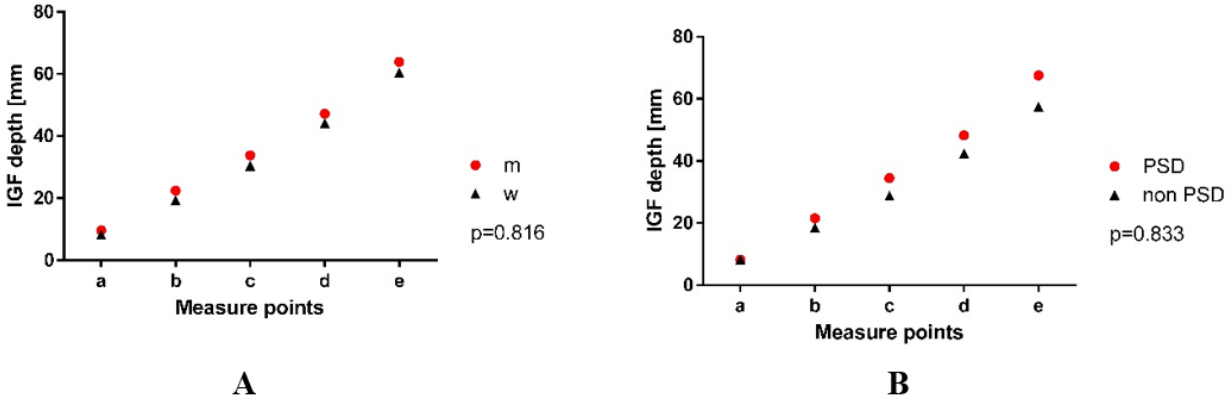

FIGURE 4a & 4b: a) IGF depth for men and women; **b)** further IGF depth for PSD versus non-PSD patients. Measuring points: **a)** cranial opening of intergluteal fold; **b)** proximal intergluteal fold.

A relationship between BMI and IGF depth was examined. Figure 5 illustrates BMI versus the intergluteal depth [in mm], showing a moderate increase in IGF depth with increasing BMI (slope 95% CI: 0.01050 to 0.5088, R square = 0,004163, p=0.041). The data indicates that the IGF depth moderately increases with BMI in our cohort measured with this method at this time.

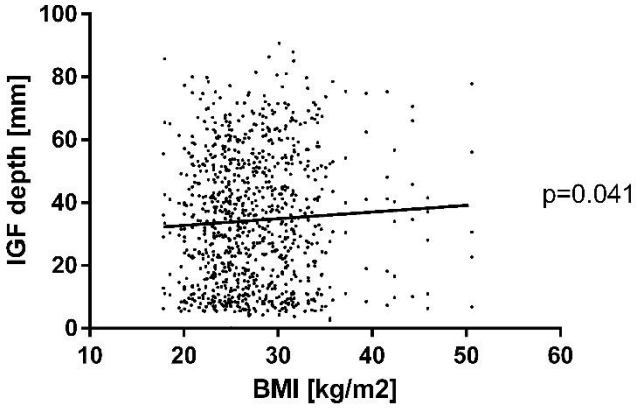

FIGURE 5: Intergluteal fold depth and BMI (n=1000 measurements).

Age and its effect on body composition was found to affect BMI in the cohort, with a mean (±SD) increase in BMI over age, from the second decade of life (25.0 ± 3.9 kg/m^2) to the 8th decade (28.5 ± 5.3 kg/m^2) (Figure 6). The effect of age on IGF depth was analyzed between the second and eighth decades (Table 1). It was observed that IGF depth decreased at all five measuring points; with the largest decrease (by a factor of 1.7) observed in the mid region of the intergluteal fold (position c) (Table 1). This indicates a decrease in IGF depth with age at all measured points while BMI slightly increases, suggesting a less muscular contour effect. Overall, these findings suggest that IGF depth is not significantly influenced by gender or the presence of PSD, but it shows a moderate association with BMI and a decrease with advancing age.

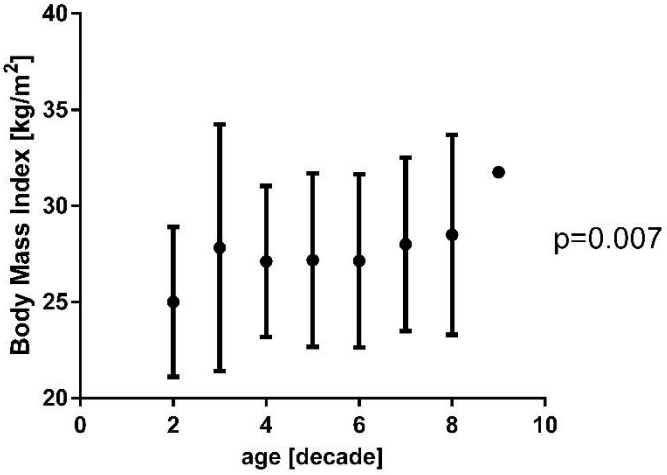

FIGURE 6: Age and Body Mass Index in n=200 patients.

TABLE 1: Intergluteal fold depth [in mm] at five measuring points a-e, versus age decades 2-8. Measuring points: **a)** cranial opening of intergluteal fold; **b)** proximal intergluteal fold; **c)** mid intergluteal fold; **d)** distal intergluteal fold; **e)** caudal end of intergluteal fold close to anus.

age [decade]	a	b	c	d	e
2	1.55	2.91	4.27	5.73	7.00
3	1.48	2.78	4.03	5.55	7.10
4	1.33	2.82	3.98	5.20	7.00
5	1.21	2.33	3.29	4.75	6.42
6	1.28	2.64	3.56	5.04	6.40
7	1.05	1.89	3.00	4.42	6.16
8	1.00	2.25	2.50	3.50	5.75

4. Discussion

The mechanism of sharp hair fragment insertion pilonidal sinus disease is an intriguing research topic. While certain co-factors like higher axial hair force (positive) and sweating (negative) have been identified as contributing factors, there are still some areas that require further exploration. Exploring the concept of anatomical variations as a contributing factor to the insertion of hair fragments is a straightforward avenue of investigation. Considering the publication in 2009 that proposed IGF depth as a potential confounding factor, it became necessary to reevaluate this concept due to contrasting experiences observed in clinical practice which led us to our thesis, that there is no association between IGF depth and PSD risk, and IGF depth does not serve as an independent risk factor for PSD.

In our study, we conducted measurements using a standardized 5-point-method using a light weight tool in a larger cohort and found no significant difference in depth of the natal cleft between PSD patients and the normal population. This raises doubts about the overall influence of IGF depth on the development of PSD due to several reasons:

i. The maximum IGF depth is observed at the level of the anus, where PSD occurrence is rare and worth case reports [15]. Most cases of PSD are found at the cranial opening of the IGF near the glabella sacralis. This questions the importance of a region unaffected by PSD in the disease's genesis.
ii. The proximal IGF depth at the cranial opening, where PSD primarily originates, is only a few millimeters. The depth does not appear to influence the occurrence of PSD in this region.
iii. Previous research indicates that sharp hair fragments slide down the back, and the initial contact point between the hair and a change in hair position (other than parallel to the skin surface) may occur at the cranial opening of the IGF.

Hair tends to stand upright at the cranial opening of the IGF and can be held in place by existing hair, potentially being driven into the skin by the walking movements of the gluteal muscles. The amount of hair reaching the IGF from the occiput to the lumbar region and the anatomical shape of the upper intergluteal opening may affect the presence of hair in the IGF. The discrepancy between our study's findings and the previous study from Turkey regarding the association of IGF depth with PSD may be attributed to several factors. It is important to consider the following possible explanations.

4.1. Variations in Study Populations

Our study involved a different population from a normal population from northern Germany, while the previous study was conducted in Turkey. Genetic, environmental, and lifestyle differences among populations can contribute to variations in disease prevalence and risk factors. The collected data of either study does not provide conclusive evidence to draw a satisfactory conclusion regarding the differences between the study cohorts.

4.2. Sample Size and Statistical Power

The sample sizes in the two studies differ, which can influence the statistical power to detect associations. A larger sample size yields more robust and reliable results.

4.3. Random Chance

It is also possible that the discrepancy is due to random chance. In scientific research, there is always a possibility of obtaining conflicting results purely by chance, especially when dealing with complex biological phenomena.

4.4. Methodological Differences

Differences in the methodology used to measure IGF depth between the two studies could contribute to contradictory results. Variations in measuring tools, techniques, and protocols can impact the accuracy and reliability of the measurements.

4.5. To Address this Possible Explanation

Using a measuring tool that applies pressure on the gluteal muscles, the weight of the tool can compress the soft contour of one of the largest muscles in the body, leading to smaller IGF depth measurements compared to a lightweight tool. Intriguingly, three out of five standard position measurements in our study exceeded the maximum measurements by Akinci *et al.* Body weight in the Turkish cohort could theoretically also explain a shallower IGF. We showed that a reduction of the BMI from 50 kg/m^2 to 20 km/m^2 reduces the IGF depths by 25% (Figure 4). So, to reduce the IGF depth by a factor of 2 (alias 100%), the BMI of the turkish cohort must have been below 0 kg/m^2 in average, which is impossible. This excludes the BMI effect as a significant denominator for very low numbers in the cohort of Akinci *et al.*

Age- related muscle atrophy is also unlikely to account for this discrepancy, since Akinci´s cohort was significantly younger (mean age of 27 years, while our cohort median age was 38 years). Since Akinci´s cohort was significantly younger, the intergluteal depth should have been more prominent than in our cohort. Adding another aspect, his female patient share was 13/91 (14%), while our cohort aimed to measure as much females (n=75; 37.5%) as males (n=125, 62.5%) Thus we had twice as much females within our cohort, which showed a trend - albeit not significant - onto lower BMI and smaller IGF depths than men. But the effect of lesser female numbers within his cohort does not explain the extent of deviation in the measurements. As neither differences in age nor BMI nor gender composition of the cohort can explain Akinci´s exceptionally lower measurement values, there must be other reasons not mentioned before.

Hence, we propose that the deviation in measurements may be attributed to compression of the gluteal muscle onto the sacrum during measurement, possibly due to the use of a heavy-weight tool or manual compression. Such measurement settings may not provide reliable information about the anatomical shapes of delicate soft tissues like the intergluteal fold and the gluteal muscle.

Limitation

i. Position during measurement: The IGF depth was measured in supine patients, representing just one of the positions individuals adopt daily. Different positions might affect measurements, introducing variability. The prone was position technically necessary for measurements and to compare with the prior Acinci *et al.* study.
ii. Population cohort: The study focused on a northern German population, recognizing that anatomical characteristics and predispositions can vary across populations and ethnicities. Findings may not be directly applicable to other countries or populations.
iii. Generalizability: While the study suggests similarities in PSD genesis between developed countries like Germany and Turkey, it is essential to acknowledge each country's unique contributing factors. Caution is needed when extrapolating findings to other regions.
iv. Anatomical variety: The presence of migrants within the German population was noted, potentially contributing to anatomical diversity. However, the extent to which this diversity is represented in the study cohort is not explicitly specified.

Further research is required to validate and reconcile these opposing findings. Additional studies involving diverse populations, rigorous methodologies, and larger sample sizes could offer a clearer understanding of the link between IGF depth and PSD risk.

The study identified a small effect size with a Cohen's-d value of 0.0858. Although drawing definitive conclusions from a small effect size presents challenges, it is crucial to note that this small effect provides insightful information about the connection between IGF depth and PSD risk. Moreover, the study's power was calculated at 5%, indicating the necessity for larger sample sizes and stringent methodologies in future research to bolster statistical power and enhance findings.

This study reveals that the deepest point of the IGF consistently resides distant from areas where PSD primarily occurs. Consequently, it can be inferred, that IGF depth does not correlate with PSD development and does not impact PSD therapy. Given that most sharp hair fragments measure between 5 and 15 mm short, any structure within this range could elevate hair fragments, causing the sharp end to potentially penetrate the skin irrespective of IGF's anatomical features. Our study's findings provide evidence that IGF depth lacks association with PSD risk. IGF depth does not emerge as an independent risk factor for PSD.

In conclusion, this study conducted on a northern German cohort offers compelling evidence that IGF depth is not linked to PSD development and does not act as an independent risk factor. The findings suggest other factors beyond IGF depth contribute to PSD's origins. This underscores the importance of considering factors beyond IGF depth in PSD therapy. To validate and expand these insights, future research should involve larger and diverse populations, robust methodologies, and thorough exploration of potential risk factors.

Ethical Approval

The analysis done in this study did not contain any interventions that could potentially cause harm to human participants. Nevertheless, Ethic approval was given by the Ethics Committee of the county Ethics chamber of the Saarland University Homburg / Saar 59/22 from 11th of July 2022 (Chair Prof. Dr. Grundmann) and by the Ethics Committee Hannover GRAE/151/2022 from 19th of August 2022 (Head Prof. Dr. Creutzig).

Conflicts of Interest

None.

Funding

We acknowledge financial support by Deutsche Forschungsgemeinschaft and Friedrich-Alexander-Universität Erlangen-Nürnberg within the funding programme "Open Access Publication Funding".

Author Contributions

Main manuscript draft by Matthias Maak, Philipp Mörsdorf, and Dietrich Doll. Reviewing of the main manuscript and final approvel by Matthias Maak, Philipp Mörsdorf, Layla Bari, Myriam Braun-Münker, Maximilian Scharonow, Marcel Orth, and Dietrich Doll. Study concept by Dietrich Doll. Data acquisition by Layla Bari and Dietrich Doll. Figures preparation by Matthias Maak, Layla Bari, and Dietrich Doll. Data analysis and interpretation by Matthias Maak, Philipp Mörsdorf, Layla Bari, Myriam Braun-Münker, Maximilian Scharonow, Marcel Orth, and Dietrich Doll.

Data Availability

The datasets generated and/or analyzed during the current study are not publicly available because they are not fully published yet but are available in due time from the corresponding author on reasonable request.

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