

The epidemiology, healthcare and societal burden of basal cell carcinoma in Wales 2000–2018: a retrospective nationwide analysis

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Abstract

Background Basal cell carcinoma (BCC) represents the most commonly occurring cancer worldwide within the white population. Reports predict 298 308 cases of BCC in the UK by 2025, at a cost of £265–366 million to the National Health Service (NHS). Despite the morbidity, societal and healthcare pressures brought about by BCC, routinely collected healthcare data and global registration remain limited.

Objectives To calculate the incidence of BCC in Wales between 2000 and 2018 and to establish the related healthcare utilization and estimated cost of care.

Methods The Secure Anonymised Information Linkage (SAIL) databank is one of the largest and most robust health and social care data repositories in the UK. Cancer registry data were linked to routinely collected healthcare databases between 2000 and 2018. Pathological data from Swansea Bay University Health Board (SBUHB) were used for internal validation.

Results A total of 61 404 histologically proven BCCs were identified within the SAIL Databank during the study period. The European age-standardized incidence for BCC in 2018 was 224.6 per 100 000 person-years. Based on validated regional data, a 45% greater incidence was noted within SBUHB pathology vs. matched regions within SAIL between 2016 and 2018. A negative association between deprivation and incidence was noted with a higher incidence in the least socially deprived and rural dwellers. Approximately 2% travelled 25–50 miles for dermatological services compared with 37% for plastic surgery. Estimated NHS costs of surgically managed lesions for 2002–2019 equated to £119.2–164.4 million.

Conclusions Robust epidemiological data that are internationally comparable and representative are scarce for nonmelanoma skin cancer. The rising global incidence coupled with struggling healthcare systems in the post-COVID-19 recovery period serve to intensify the societal and healthcare impact. This study is the first to demonstrate the incidence of BCC in Wales and is one of a small number in the UK using internally validated large cohort datasets. Furthermore, our findings demonstrate one of the highest published incidences within the UK and Europe.

What is already known about this topic?

- The incidence of basal cell carcinoma (BCC) is recognized to be increasing.
- Epidemiological studies in Wales are limited, and no significant large cohort BCC research has been performed.

What does this study add?

- This study identifies the incidence of BCC in Wales and recognizes it to be one of the highest published in Europe.
- This study identifies geographical and social variations, while integrating routinely collected healthcare data to assess distances travelled for specialist input.
- This study provides an assessment of the significant financial implication of care in Wales, and a projection of future burden of care to inform service planning.

Accepted: 5 November 2022

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Basal cell carcinoma (BCC) is the most common malignancy within the White population worldwide, with an incidence rate that continues to rise.¹ While BCCs have a low mortality, they are associated with a significant degree of physical and psychological morbidity, in addition to societal and healthcare costs. Despite this significant burden, BCC primary data collection is relatively poor in comparison with data collection for other common malignancies, which negatively impedes research and policymaking. Routinely collected electronic health record (EHR) data and registration for many cancer registries remains less than fully representative, largely because of the associated workload, synchronicity and metachronicity of lesions, and nonintegration of clinically diagnosed (but not histologically confirmed) and treated lesions.² The resulting impact of this current data collection strategy is an underestimation of the true burden of disease.^{3–8} UK registries also only consider the first diagnosed BCC per patient as stipulated by the UK and Ireland Association of Cancer Registries, meaning that the true workload associated with the many patients who present with multiple lesions at once or in succession is underestimated. However, this practice is not isolated to the UK.¹

In order to combat poor data collection, novel methods have been employed, the results of which suggest an annual UK growth in incidence of 5%.⁷ Historically the epidemiology of BCC in Wales has been poorly represented in the literature, in a country in which 96% of its population identify as White with a high proportion of Fitzpatrick skin types I and II.⁹ Regional population-based studies of nonmelanoma skin cancer (NMSC) that have been carried out in Wales, although small, have highlighted a growth in registration of 66% in a 10-year interval.¹⁰ These rising rates are predicted to result in 380 000 cases of NMSC (80% of which would be BCCs) in the UK by 2025, with a projected economic impact on the NHS of £338–465 million.¹¹

The purpose of this study is to use population-scale data to curate an electronic cohort (e-cohort) of anonymized healthcare data to determine the epidemiology, utilization and societal and financial implications of BCC care across Wales over a period of 18 years. This represents the largest epidemiological study of its kind reinforced by validated pathological data and advanced data linkage to cross-interrogate various data sources in Wales.

Materials and methods

Study design and data sources

This population-based retrospective cohort study was conducted in accordance with the REporting of studies Conducted using Observational Routinely collected Data (RECORD)¹² statement.

The Secure Anonymised Information Linkage (SAIL) Databank, a world-leading trusted research environment with population-scale anonymized healthcare data in Wales,^{13,14} was the primary source, and was supplemented by robust pathological data from Swansea Bay University Health Board (SBUHB).

The SAIL Databank holds billions of deidentified person-based records from multiple data sources, integrated through privacy-protecting data linkage.¹⁵ Routinely collected

EHRs are assigned a unique identifier referred to as an anonymized linking field (ALF), generated by Digital Health and Care Wales, which undergoes two further encryptions prior to project access within the SAIL Databank.¹⁵

The e-cohort was defined within the Welsh Cancer Intelligence Surveillance Unit (WCISU) and linked with primary care data covering the vast majority of the general practice population, and secondary care inpatient and outpatient data covering 100% of the population (Table 1). These data were linked between the years 2000 and 2018 and were analysed to investigate the epidemiology, healthcare utilization and cost of BCCs.

Pitfalls within the coding of NMSC arise owing to generic coding,² such as the International Classification of Diseases 10th revision (ICD-10) C44 code. Therefore, the most reliable method of identification of a BCC involves identifying those that are histologically proven as such within cancer registries and linked using the unique patient identifier to various other databases where coding is less specific.

Study population

All patients with a diagnosis of BCC registered within the WCISU between 2000 and 2018 were considered in the study. An e-cohort of patients was defined using the ICD-10 code C44 and International Classification of Diseases for Oncology 02 (ICD-O2) morphology codes 8090–8095, 8097 and behavioural code 3 (invasive). *In situ* disease was not included. Analysis took place using the first registered BCC per patient during the 18-year study period. Table 1 summarizes the data sources used within SAIL.

Outcomes

Patient demographics

Demographics, lesion locality and morphology were identified using the date of histological lesion diagnosis. The Welsh Index of Multiple Deprivation (WIMD, 2011 version) was used, which is a tool approved by the Welsh Government to assess the socioeconomic status of small geographical regions, known as Lower-layer Super Output Areas (LSOAs), of which there are 1909 within Wales.¹⁶

Table 1 Definitions of accessed data sources

| Database name | Description |
|---|--|
| Patient Episode Database for Wales (PEDW) | Attendance and clinical information for all hospital admission including operations performed in Wales |
| Outpatient Dataset for Wales (OPDW) | Attendance information for all hospital outpatient appointments in Wales |
| Welsh Cancer Intelligence and Surveillance Unit (WCISU) | Cancer registry for Wales, used automated systems to integrate information from multiple sources to include histopathology, multidisciplinary teams data, etc. |
| Welsh Longitudinal General Practice Dataset (WLGP) | Attendance and clinical information for all general practice interactions |
| Welsh Demographic Service Dataset (WDSD) | Population spine for Wales, used to identify anonymized changes in general practice registration and residency over time |

Each LSOA is assigned a quintile of socioeconomic status from one (the lowest) to five (the highest).

Incidence

Incidence is described as the crude rate, European age-standardized rate (EASR)^{17,18} and World age-standardized rate (WASR)¹⁹ to permit comparison with worldwide data. Mid-year population estimates for specified age groups and regions were obtained from Stats Wales published by the Welsh Government (<https://statswales.gov.wales/>).

Healthcare utilization

Primary care attendances were accessed via the Welsh Longitudinal General Practice Dataset (WLGP), secondary care outpatient appointments via the Outpatient Dataset for Wales (OPDW) and inpatient or day case procedures via the Patient Episode Database for Wales (PEDW). Duplicate records were removed within each data source to permit a single ALF per patient.

Access to health

Centroid coordinates of each patient's LSOA were used to calculate geodesic distances travelled for specialist treatment. A distance formula based on the earth's radius was used to give an approximation of distances travelled as the crow flies.

Costing

The financial burden of BCC care on NHS Wales is given based on research by Vallejo-Torres *et al.* using a bottom-up and top-down cost model (specific methodology is provided in their study), and subsequently used by Goon *et al.* as a framework to estimate cost projection for the UK in 2025.^{11,20} The top-down approach combined health service utilization data with unit cost of services data, whereas the bottom-up approach constructed a simplified patient pathway that was subsequently populated with probabilities based on published literature and associated unit cost of each element.

Data validation

Pathological data from SBUHB were extracted from a prospective pathology database by a consultant histopathologist (N.W.) between 2003 and 2019 using SNOMED Clinical Terms codes for BCC (Table S1; see [Supporting Information](#)). Two districts reliably encompassed within the jurisdiction of SBUHB (Swansea and Neath Port Talbot) were isolated; 4 years of data were selected and validated (2014–2017). Initial validation of pathological data took place via a filtering algorithm to highlight any BCC excision or biopsy within 3 months of another excision or biopsy with a matching NHS number to avoid double counting of lesions (i.e. those undergoing a biopsy prior to excision or those that were incompletely excised and required further surgery). We opted to use 3 months as the upper boundary cutoff as this represents the urgent suspected cancer pathway and referral to treatment timeframe.²¹ Histopathology reports that

occurred within 3 months of one another were then individually searched by a single plastic surgeon (N.I.). Incorrectly coded tumours were eliminated, lesions that were biopsied and subsequently excised were counted once, and incompletely excised lesions were counted once. Electronic documents were consulted if details from the histopathology report were insufficient. An adjustment factor was applied to all remaining pathological data based on the validation. Comparison in case burden was then made between adjusted pathology data and SAIL data for matched regions and years.

Statistical analysis

SAIL data were extracted by a SAIL data analyst using SQL developer IBM DB2 (IBM Corp, Armonk, NY, USA). Microsoft Excel 2016 (Microsoft Corp, Redmond, WA, USA) and R i386 Version 3.5.0 (R Foundation for Statistical Computing, Vienna, Austria) were used for statistical analysis. Mathematica Version 12.3 (Wolfram Research, Inc., Champaign, IL, USA) was used for data forecasting. A statistical forecasting model was constructed within SPSS (IBM) based on linear and quadratic equations to estimate three case burden scenarios extrapolated to 2030 for Wales.

Results

Overall, 61 404 patients with a histologically proven BCC for the duration of the study period were identified between 2000 and 2018 in Wales. The population of Wales increased by 7.9% during the study period to 3 138 631.

Data acquisition from the WCISU is illustrated in Figure 1 and population demographics are summarized in Table 2. The median age of the first histologically proven BCC for both male and female patients for the duration of the study period was 72 years [95% confidence interval (CI) 70.5–70.7 (men); 95% CI 70.3–70.6 (women)] with a male preponderance (1 : 0.83).

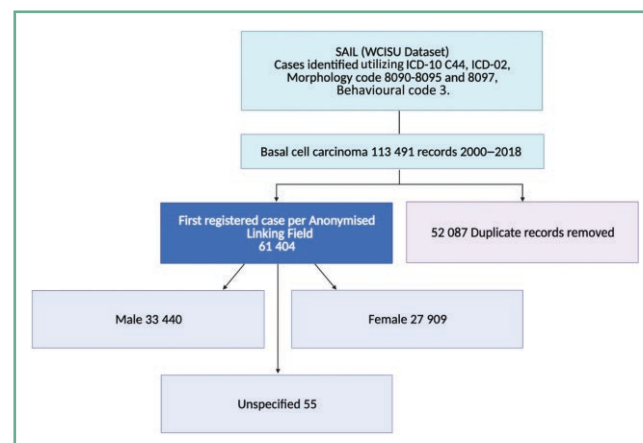


Figure 1 Data preparation for the electronic cohort. SAIL, Secure Anonymised Information Linkage; WCISU, Welsh Cancer Intelligence and Surveillance Unit; ICD-10, International Classification of Disease 10th revision.

Table 2 Patient demographics 2000–2018

| | Male patients, n=33 440 (54.5) | Female patients, n=27 909 (45.5) |
|--------------------|-----------------------------------|-------------------------------------|
| Age group, years | | |
| 5–19 ^a | 15 | 12 |
| 20–24 | 33 (0.1) | 29 (0.1) |
| 25–29 | 64 (0.2) | 82 (0.3) |
| 30–34 | 148 (0.4) | 184 (0.7) |
| 35–39 | 275 (0.8) | 380 (1.4) |
| 40–44 | 539 (1.6) | 643 (2.3) |
| 45–49 | 903 (2.7) | 1064 (3.8) |
| 50–54 | 1542 (4.6) | 1555 (5.6) |
| 55–59 | 2288 (6.8) | 1984 (7.1) |
| 60–64 | 3309 (9.9) | 2540(9.1) |
| 65–69 | 4766 (14.3) | 3456 (12.4) |
| 70–74 | 5544 (16.6) | 3867 (13.9) |
| 75–79 | 5742 (17.2) | 4014 (14.4) |
| 80–84 | 4673 (14.0) | 3842(13.8) |
| 85–90 | 2874 (8.6) | 3100 (11.1) |
| > 90 | 725 (2.2) | 1157 (4.1) |
| Total | 33 440 | 27 909 |
| WIMD 2011 | | |
| 1 (Most deprived) | 4732 (14.2) | 4155 (14.9) |
| 2 | 5710 (17.1) | 4891 (17.5) |
| 3 | 6608 (19.8) | 5571 (20) |
| 4 | 7464 (22.3) | 6229 (22.3) |
| 5 (Least deprived) | 8846 (26.5) | 6990 (25.1) |
| NA | 80 (0.2) | 73 (0.3) |
| Site | | |
| Lip | 342 (1.0) | 650 (2.3) |
| Eyelid/canthus | 2432 (7.3) | 2617 (9.4) |
| Ear | 2687 (8.0) | 449 (1.6) |
| Face | 11 683 (34.9) | 10 884 (39) |
| Scalp/neck | 1808 (5.4) | 1366 (4.9) |
| Trunk | 2811 (8.4) | 1508 (5.4) |
| Upper limb | 1228 (3.7) | 782 (2.8) |
| Lower limb | 729 (2.2) | 1829 (6.6) |
| Overlapping lesion | 40 (0.1) | 32 (0.1) |
| Unspecified | 9680 (28.9) | 7792 (27.9) |

WIMD, Welsh Index of Multiple Deprivation; NA, not available. ^aAge groups merged to avoid risk of unmasking when *n* < 5, as per Secure Anonymised Information Linkage disclosure control policies.

Incidence

As confirmed through direct correspondence with the WCISU, data collection for NMSC was made mandatory from 2016. This accounts for the large apparent increase

in recorded incidence in 2016. The mean crude incidence from 2016 to 2018 was 242.5 per 100 000 person-years (PYs) (95% CI 222.2–262.8), the EASR was 235.3 (95% CI 209–261.5) and the WASR was 117.2 per 100 000 PYs (95% CI 104.2–130.1) for the first registered BCC per patient. For the period 2016–2018 a mean of 7579 cases were diagnosed per year. Inferences on incidence change are unlikely to be representative as data capture was incomplete prior to 2016, as exemplified in Figure 2. Age-specific incidence per age group (by decade) between 2016 and 2018 is represented in Figure 3. All forms of standardization methodology are represented in Table S2 (see Supporting Information).

Pre-2016 projection

Prior to 2016, data collection for NMSC was nonmandatory within the WCISU, resulting in under-reporting of incidence. Comprehensive pathology data from SBUHB for the districts of Swansea and Neath Port Talbot have been consistently collected since 2003. Overall, 4 years of this dataset were validated, and an adjustment factor was applied to the remaining cohort (Table S3; see Supporting Information). These data were compared directly to matched regions within the SAIL Databank and used to backward-project the pre-2016 data to give a more accurate reflection of pan-Wales incidence (Figure 4).

Body site

The face was the most affected anatomical region in both male and female patients (34.9% in male patients and 39% in female patients). BCCs of the face, lower limb, lip and eyelid were more common in female patients and BCCs of the ear/external auditory canal, scalp/neck, trunk, and upper limb were more common in male patients (Table 2). Pearson’s χ^2 -test demonstrated a clear statistical significance between site and sex (*P* < 0.05).

Socioeconomic and geographical distribution

Crude incidence based on urban/rural locality is demonstrated in Figure 5. Despite the diagnostic caseload within urban regions being three times higher than that recorded

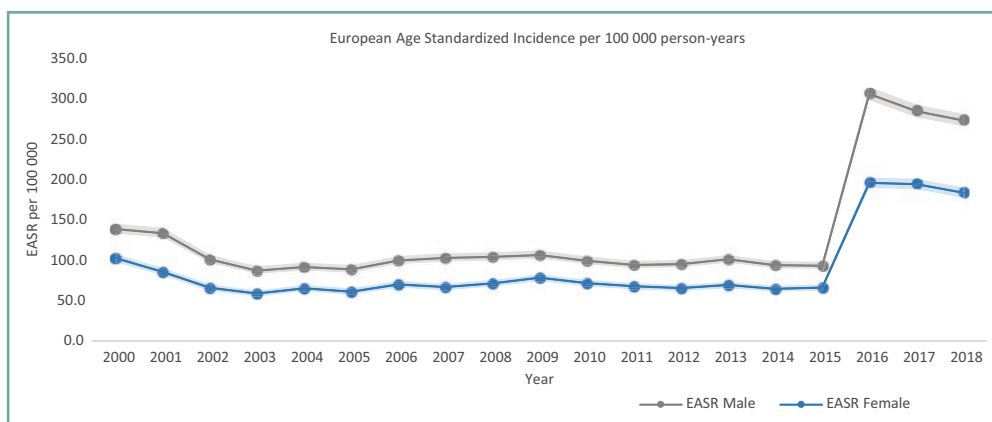


Figure 2 European age-standardized incidence (per 100 000 person-years) of basal cell carcinoma in male and female patients 2000–2018. Shading denotes upper and lower 95% confidence intervals. EASR, European age-standardized rate.

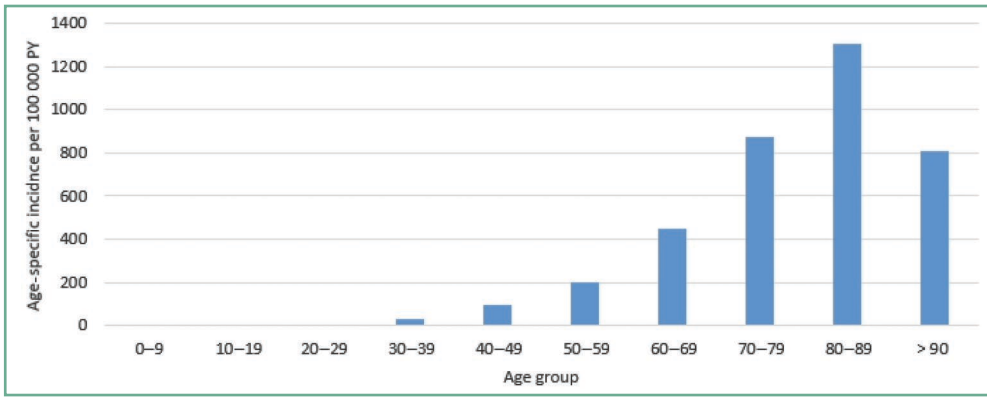


Figure 3 Age-specific incidence (per 100 000 person-years) per decade age group 2016–2018.

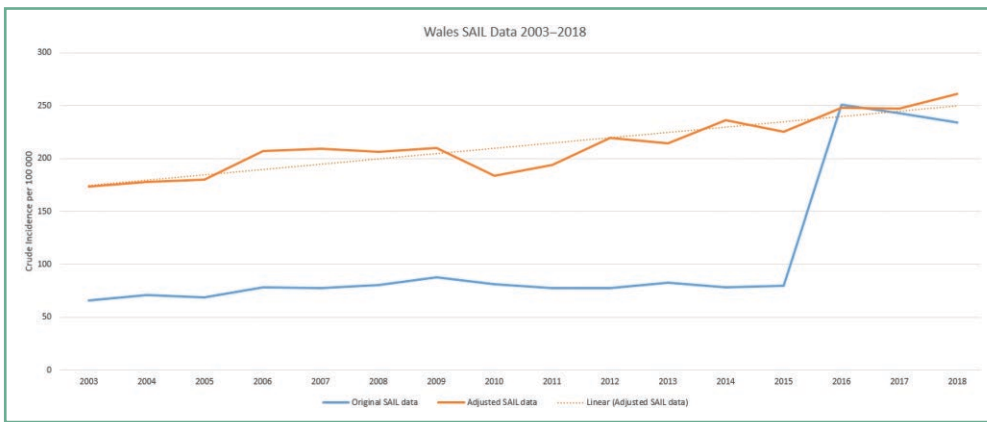


Figure 4 Original and adjusted Secure Anonymised Information Linkage (SAIL) crude incidence for Wales 2003–2018. Original SAIL data depicted in blue; the blue shading outlines the period of nonmandatory data collection within Welsh Cancer Intelligence Surveillance Unit followed by an increase in 2016–2018 when mandatory collection was introduced. The adjusted crude incidence (solid orange line) has been calculated by determining the relationship between regional pathological data and pan-Wales SAIL data from 2016 to 2018. This has been backward-projected by multiplying this relationship by the adjusted pathological data to give the adjusted SAIL data, i.e. a more accurate reflection of pan-Wales incidence during the nonmandatory skin cancer registration period (regional data are based on histopathological reports and hence were not affected by cancer registry practice). The orange dashed line represents the line of best fit through the forecasted SAIL data.

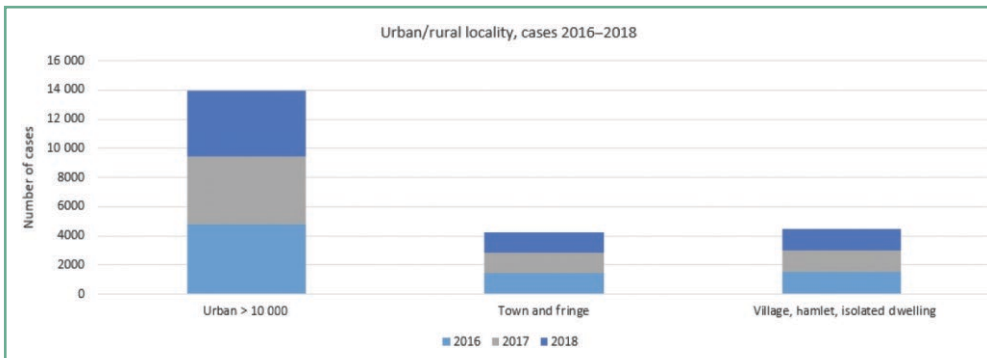


Figure 5 Basal cell carcinoma crude incidence per 100 000 person-years based on urban/rural locality (morphology codes 1–3) 2016–2018.

for rural regions (Figure 6), relative to the population, crude incidence was demonstrated to be higher within a rural locality. The mean crude incidence was found to be statistically significant using one-way anova. The Tukey’s honest significant difference test was used to further delineate

statistical significance between each of the three categories ($P < 0.05$).

A greater proportion of the study group was found to be within the least socially deprived socioeconomic group (WIMD 5) for both male and female patients (Table 2). No

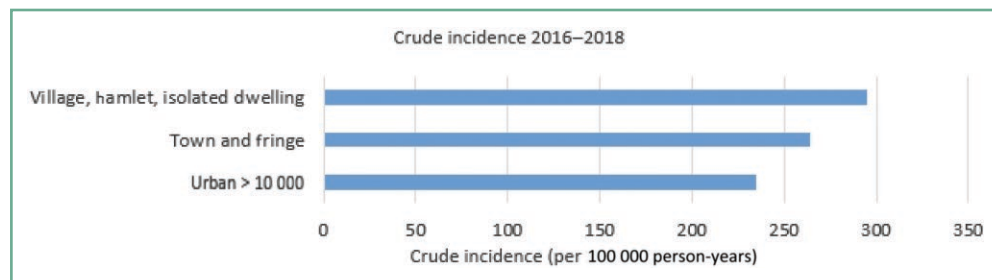


Figure 6 Basal cell carcinoma caseload based on urban/rural locality 2016–2018.

statistical difference was noted between the median WIMD of male patients vs. female patients using the Mood median test ($P > 0.05$).

Healthcare utilization: outpatient appointments

In total, 16 016 records were identified as BCC-related outpatient appointments between 2004 and 2018, 11 686 were squamous cell carcinoma and the remaining 14 306 were coded generically as C44, which encompasses all NMSC. The mean number of follow-ups for BCC-coded outpatient appointments (based on linked morphology codes) was 2.42 [median 1, interquartile range (IQR) 1–3]. Overall, 84% of referrals were from general practitioners, 9% were referred from other consultants (excluding Accident & Emergency), 4% from other sources and 3% from the consultant responsible for the OPDW episode. The proportions of outpatient appointments by specialty are illustrated in Table 3.

Healthcare utilization: inpatient/day case treatment

Overall, 61 284 inpatient/day case procedures were coded as BCC-related and histologically proven as such, which represents 39 803 patients. The mean spell duration was 0.5 days (median 0, IQR 0–0). The mean number of episodes per patient was 1.5 (median 1, IQR 1–2) during the study period. The cumulative number of bed days was 28 682. The distribution among inpatient/day case specialty is outlined in Table 4 and the distance travelled to obtain specialist treatment is provided in Table 5.

Geographical variation

The EASR of each principal region within Wales based on incidence data for 2016–2018 is represented in the choropleth map (Figure 7 and Table S4; see [Supporting](#)

Table 3 Proportion of appointments per specialty using both generically coded C44 data and data linkage from Outpatient Dataset for Wales to Welsh Cancer Intelligence Surveillance Unit 2004–2018

| Specialty | Proportion of outpatient appointments with a diagnosis of basal cell carcinoma (%) | Proportion of outpatient appointments with C44 code (%) |
|-------------------|--|---|
| Dermatology | 92.9 | 89.5 |
| Plastic surgery | 3.0 | 5.8 |
| Clinical oncology | 3.7 | 4.0 |

Note that the sum does not total 100% as the smaller contributions from allied specialities have been removed.

Table 4 Proportion of inpatient/day case episodes by specialty 2000–2018 (Patient Episode Database for Wales database)

| Specialty | Proportion of inpatient/day case spell (%) |
|----------------------|--|
| Dermatology | 40.5 |
| Plastic surgery | 23.4 |
| Oral surgery | 11.8 |
| Clinical oncology | 8.0 |
| Ophthalmology | 7.7 |
| Ear, nose and throat | 5.0 |
| General surgery | 3.0 |

Note that the sum does not total 100% as the smaller contributions from allied specialities have been removed.

Table 5 Access to dermatology, plastic surgery and oncological services 2000–2018

| Distance travelled | Dermatology (%) | Plastic surgery (%) | Oncology (%) |
|--------------------|-----------------|---------------------|--------------|
| < 25 miles | 97.9 | 57.1 | 91.6 |
| ≥ 25–50 miles | 1.8 | 36.9 | 7.4 |
| ≥ 50 miles | 0.3 | 6 | 1 |

Geographical reference points: Wales spans 130 miles from north to south and the widest point spans 100 miles from east to west.

[Information](#)). The EASR was noted to be highest in urban Swansea (318.9 per 100 000 PYs, 95% CI 296.1–341.6) with a population of 245 000, almost twice as high as that noted in rural Ceredigion (172.6 per 100 000 PYs, 95% CI 144.8–200.5), which has a population of 73 000 (stated populations based on 2018).

Projecting future cases

Analysis of validated SBUHB pathology data shows a 45% greater incidence compared with matched regions within the SAIL Databank for the period from 2016 to 2018 (based on Swansea and Neath Port Talbot). Linear and quadratic projections of the adjusted and scaled SBUHB pathology data to all regions within Wales and adjusted SAIL data are shown in Figure 8. Three scenarios are illustrated for each dataset based on projections of growth. Projections of the adjusted pathology dataset estimate between 18 874 and 20 370 cases in Wales by 2030.

Costing

Based on previous research by Vallejo-Torres *et al.*, estimated cost for diagnosis and treatment of cases identified histologically through SAIL for 2018 alone, would amount to £6.5–9.0

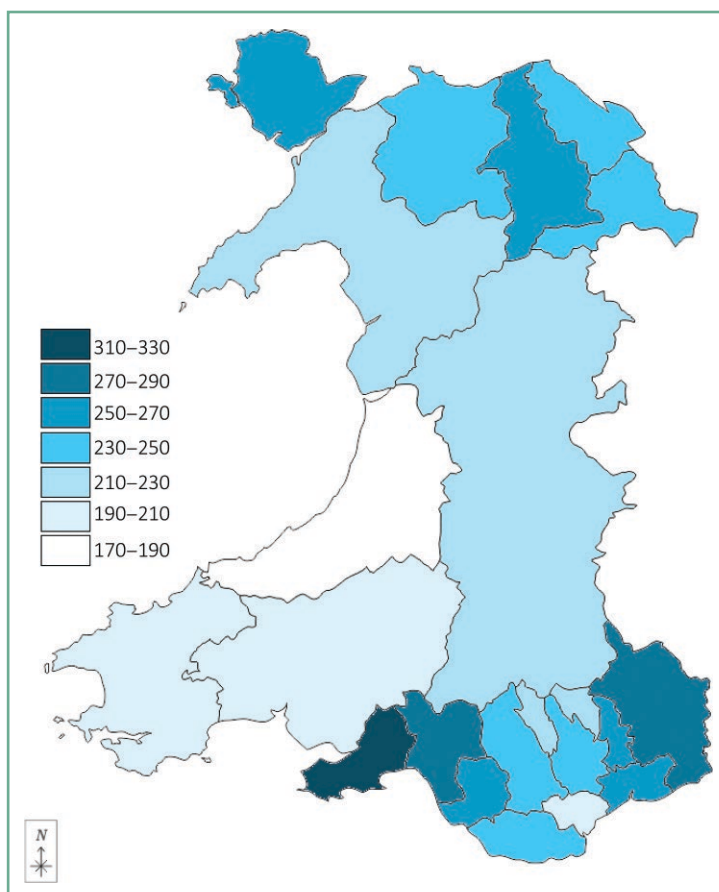


Figure 7 European age-standardized rate of basal cell carcinoma in each principal region within Wales 2016–2018 (per 100 000 person-years).

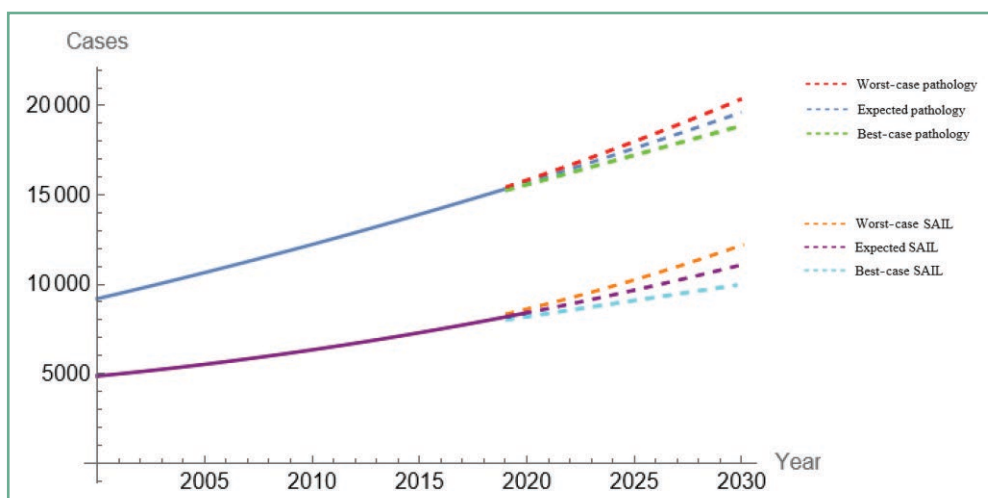


Figure 8 Projection of adjusted Swansea Bay University Health Board (SBUHB) cases (Swansea and Neath Port Talbot) to the population of Wales to 2030 (blue). Three potential projections, based on utilization of linear (green dashed line), quadratic (red dashed line) and an ‘expected’ scenario derived by taking an average of both models (blue dashed line). Projection of adjusted Secure Anonymised Information Linkage (SAIL) data to 2030 (purple). Three potential projections, linear (light blue dashed line), quadratic (orange dashed line) and ‘expected’, demonstrated an average of both models (purple dashed line). Multiple projection models were tested in SPSS (IBM, Armonk, NY, USA); we concluded the best two models were that of a linear and nonlinear (quadratic) equation. The linear model with a constant gradient was used to model the best-case scenario, i.e. that the cases do not change by a significant amount year-on-year. The quadratic model with the x^2 term clearly has a nonuniform gradient and the predicted cases can differ significantly year-on-year; therefore it was used to model the worst-case scenario. From this, an expected model was formulated, defined as the average of the worst-case and best-case models.

million (single case per histological type per patient). When the 45% difference calculated between pathology and SAIL data for matched regions (which represents multiple lesions data) is incorporated, this figure rises to £9.5–13.1 million. For the period 2003–2018 the total estimated cost to NHS Wales based on projected pathology data is £92–127 million. Based on the predictive model in Figure 8, the case burden in 2030 alone would equate to £18.1–24.9 million to NHS Wales as a conservative estimate (this does not account for inflation and lesions treated through nonsurgical methods).

Discussion

There is a paucity of robust epidemiological data in the international scientific literature related to NMSC. European guidelines (ENCR²²) mandate that only a single case per histological type per patient is recorded within cancer registries – despite a 44% subsequent risk of further BCC.²³ This method of recording underestimates case burden by up to 45%, as evidenced by the discrepancy demonstrated between validated pathological data (accounting for all BCCs) and SAIL data (representing cancer registry data, i.e. single lesion per patient) matched to the same region and year. This under-representation has clear implications for planning and resource allocation for all health services involved in the skin cancer treatment pathway. The rising global incidence of NMSC,¹ coupled with struggling healthcare systems in the post-COVID-19 recovery period, only serves to intensify the likely societal and healthcare impact. Studies have predicted that the likely case burden for surgically managed BCCs alone in the UK will rise to 298 305 by 2025, amounting to an estimated cost of £365 million.^{11,20}

Epidemiological data within Wales have been poorly represented with no previous large studies linking primary and secondary healthcare data. Recent publications proposing novel methods of data presentation have also omitted Welsh data on the basis of incomplete coverage.⁷ This study represents the largest internally validated Welsh cohort examining the epidemiology of BCCs using advanced data linkage techniques to extract data from multiple routine healthcare datasets.

Based on SAIL data from the 2018 cohort, the crude incidence of BCC in Wales was 234.3 per 100 000 PYs (EASR 224.6, WASR 111.8 per 100 000 PYs) with 7353 first recorded histologically confirmed diagnoses costing an estimated £6.5–9.0 million.

Using The Health Improvement Network database, a primary care repository, Musah *et al.* reported crude rates of 196.4 per 100 000 PYs (EASR 114.4, WASR 78.1 per 100 000 PYs, $n=2822$) between 2004 and 2010.²⁴ A regional study performed by Roberts⁵ and Holme *et al.*¹⁰ highlighted a crude incidence of 224.3 per 100 000 PYs (WASR 114.2 per 100 000 PYs, $n=414$) in 1998; they noted a 66% increase in BCC incidence compared with a study carried out 10 years earlier. Based on the validated pathological data, an increase in case burden of 105% in a 16-year period between 2003 and 2019 was reported (average annual growth of 6.5%). Our study demonstrates one of the highest published incidences of BCCs in Europe with a higher than average rate of growth.¹ Recent studies in the Netherlands have demonstrated comparable incidences for

2018 with crude incidences rates of approximately 284 per 100 000 PYs when counting only the first registered BCC.²⁵ The incidence in Wales is partially attributed to the higher proportions of Fitzpatrick skin types I and II in Wales. In the 2011 census, 96% of Welsh residents identified themselves as White.⁹ Comparably there was also less inward migration of individuals with a lower susceptibility to skin cancer in comparison with other parts of the UK. Similar demographics to those for Wales can be found in regions such as the Southwest and Northern Ireland, but such incidences have not been reported. However, this remains significantly lower than published rates in Australia; the 2002 national NMSC survey published incidences of WASR 883.7 per 100 000 PYs (95% CI 816.4–956.6),²⁶ with an estimated expenditure of \$1 315 140 072²⁷ in 2019/2020.

In line with previous studies, we demonstrate a correlation between case burden and those within the least socially deprived socioeconomic group. This is partially attributable to a greater disposable income permitting international travel and outdoor recreational activities (hence greater exposure to intense intermittent ultraviolet radiation exposure).^{28–30} This may also be confounded by a recognized higher rate of referral to specialist services among the least socially deprived.³¹

Blaenau Gwent, Merthyr Tydfil, Rhondda Cynon Taf and Swansea contain the highest proportions of deprived LSOAs; however, the second most populous principal region in Wales, Swansea, displayed the highest regional incidences between 2016 and 2018, and Blaenau Gwent reported one of the lowest incidences, suggesting that regional variations in BCC incidence cannot be explained by socioeconomic status alone.³²

A behavioural study commissioned by the Welsh Government published in 2009 highlighted that 8.2% of 11–17-year-olds had used a sunbed, as had 22% of all respondents to the Welsh omnibus survey in 2017.^{33,34} Sunbed use is significantly associated with increased risk of BCC especially in those under the age of 25 years.³⁵

Wales presents a unique challenge as it has a relatively small population situated over a large geographical region, with one in three people living rurally. The geodesic distance (shortest path) to access specialist services was calculated to give an insight into distances travelled for treatment. Access to dermatological, oncological and plastic surgery services (single journey and single lesion per patient) were considered. Over 90% of the patients were found to live within a 25-mile radius of dermatological and oncological services; however, this was the case for only 57% of patients seeking plastic surgery treatment, with a further 37% travelling between 25 and 50 miles and 6% travelling farther than 50 miles. A systematic review of distance to healthcare services and associated implications concluded that 77% of sampled studies ($n=108$) displayed a distance decay association, i.e. patients living closer to healthcare facilities displayed better outcomes.³⁶ Considering the demographic of patients who are commonly affected, this places pressure on patients, carers and family members through missed employment – the 'Greater Patient' concept.³⁷

In terms of social deprivation vs. access to plastic surgery tertiary care, the study revealed that the least socially deprived travelled the greatest distances. This is partially

attributed to the locality of the plastic surgery centre within a deprived principal region. However, utilization of healthcare services is a complex and multifaceted issue, which is not addressed by distance travelled alone.

Limitations of this study include specific identification of BCCs from the generic ICD-10 code of C44 encompassing all NMSCs. Only lesions that were histologically proven to be BCC were considered using morphology coding within the WICISU dataset. However, the disadvantage of this approach is that it only considers biopsy-proven lesions or those that have been surgically excised. Hence, this is likely to represent an underestimation of true burden as lesions managed through topical and destructive methods were not considered. This is confounded by the recognized underestimation of using the first diagnosed BCC per patient. This effect is carried forward when linked to the OPDW and PEDW databases; however, without linkage techniques, coding would be limited to generic ICD-10 coding and therefore specific BCC delineation would not be possible from the umbrella of NMSC. This was partially circumvented by the integration of pathological data scaled to the Welsh population, as this approach considered all histologically confirmed BCC, not just the first registered BCC. As the historic records were limited, reporting the first diagnosed BCC per patient may falsely represent their first lesion within a year that does not actually represent the first diagnosis of BCC in their lifetime. Other limitations include missing, incomplete, and misclassified datasets. Geographic information systems calculations were based on geodesic distances from LSOA to treatment centre. The modifiable areal unit problem, a form of statistical bias that can arise when using spatial analysis, must be recognized. This occurs when varying configurations of spatial units cause modified aggregation of individuals.^{38,39} Standardized populations within WASR are less comparable to the population of Wales, as they are based on a younger population structure; hence, the noted disparity between the EASR and WASR.

This study is the first to present the burden of BCC in Wales using the largest population cohort to date, supported by regional pathological data for validation and forecasting. This study demonstrates one of the highest age-standardized incidences in Europe and forecasts a conservative growth in case burden of approximately 25% in the next 10 years, in addition to a threefold increase in the expected expenditure between 2018 and 2030. Although there have been published improvements in registration technique in recent years, we cannot rely on historical coding to predict future service needs as this will result in chronic under-resourcing. If left unchecked the burden of BCC within the umbrella of NMSC is likely to overwhelm future services, in addition to the strain placed on the healthcare system during the post-COVID-19 recovery, resulting in a significant impact on waiting lists for both assessment and treatment of this disease.

Acknowledgments

The authors would like to thank Rhys Whelan, Library Services Manager, Swansea Bay University Health Board, Morriston Hospital, Swansea for his support with the literature search. The authors would also like to thank Octavian Parkes for his support as Research Programme Manager.

Funding sources

This research was supported via the RESECT project, which is part of the Scar Free Foundation Programme of Regenerative Research at the Reconstructive Surgery & Regenerative Medicine Research Centre (ReconRegen) in partnership with Health and Care Research Wales. This work was supported by Health Data Research UK, which receives its funding from HDR UK Ltd (HDR-9006) funded by the UK Medical Research Council, Engineering and Physical Sciences Research Council, Economic and Social Research Council, Department of Health and Social Care (England), Chief Scientist Office of the Scottish Government Health and Social Care Directorates, Health and Social Care Research and Development Division (Welsh Government), Public Health Agency (Northern Ireland), British Heart Foundation and the Wellcome Trust; ADR Wales is part of the Economic and Social Research Council (part of UK Research and Innovation) funded ADR UK (grant ES/S007393/1).

Conflicts of interest

The authors declare they have no conflicts of interest.

Data availability

The data were acquired from the SAIL Databank at Swansea University, Swansea, UK, but as restrictions apply, they are not publicly available.

Ethics statement

Approval for the use of anonymized data in this study, provisioned within the SAIL Databank, was granted by an independent Information Governance Review Panel (IGRP) under project 0593. The IGRP has a membership comprising senior representatives from the British Medical Association, the National Research Ethics Service, Public Health Wales and the NHS Wales Informatics Service. Usage of additional data was granted by the data owner. The SAIL Databank complies with General Data Protection Regulations and the UK Data Protection Act.

References

- 1 Lomas A, Leonardi-Bee J, Bath-Hextall F. A systematic review of worldwide incidence of non-melanoma skin cancer. *Br J Dermatol* 2012; **166**:1069–80.
- 2 Ibrahim N, Gibson J, Ali S *et al.* Is poor quality non-melanoma skin cancer data affecting high quality research and patient care? *J Plast Reconstr Aesthet Surg* 2021; **74**:1355–401.
- 3 Lucke TW, Hole DJ, Mackie RM. An audit of the completeness of non-melanoma skin cancer registration in Greater Glasgow. *Br J Dermatol* 1997; **137**:761–3.
- 4 Beadle PC, Bullock D, Bedford G *et al.* Accuracy of skin cancer incidence data in the United Kingdom. *Clin Exp Dermatol* 1982; **7**:255–60.
- 5 Roberts DL. Incidence of non-melanoma skin cancer in West Glamorgan, South Wales. *Br J Dermatol* 1990; **122**:399–403.
- 6 de Vries E. Population-based estimates of the occurrence of multiple vs first primary basal cell carcinomas in 4 European regions. *Arch Dermatol* 2012; **148**:347–54.

- 7 Venables ZC, Nijsten T, Wong KF *et al.* Epidemiology of basal and cutaneous squamous cell carcinoma in the U.K. 2013–15: a cohort study. *Br J Dermatol* 2019; **181**:474–82.
- 8 Brewster DH, Bhatti LA, Inglis JHC *et al.* Recent trends in incidence of nonmelanoma skin cancers in the East of Scotland, 1992–2003. *Br J Dermatol* 2007; **156**:1295–300.
- 9 Office for National Statistics. KS201EW – ethnic group. Available at: <https://www.nomisweb.co.uk/census/2011/KS201EW/view/2092957700?cols=measures> (last accessed on 5 December 2020).
- 10 Holme SA, Malinovsky K, Roberts DL. Changing trends in non-melanoma skin cancer in South Wales, 1988–98. *Br J Dermatol* 2000; **143**:1224–9.
- 11 Goon PKC, Greenberg DC, Igal L, Levell NJ. Predicted cases of U.K. skin squamous cell carcinoma and basal cell carcinoma in 2020 and 2025: horizon planning for National Health Service dermatology and dermatopathology. *Br J Dermatol* 2017; **176**:1351–3.
- 12 Benchimol EI, Smeeth L, Guttman A, *et al.* The REporting of studies Conducted using Observational Routinely-collected health Data (RECORD) Statement. *PLOS Med* 2015; **12**:e1001885.
- 13 Ford DV, Jones KH, Verplancke J-P *et al.* The SAIL Databank: building a national architecture for e-health research and evaluation. *BMC Health Serv Res* 2009; **9**:157.
- 14 Jones KH, Ford DV, Jones C *et al.* A case study of the Secure Anonymous Information Linkage (SAIL) Gateway: a privacy-protecting remote access system for health-related research and evaluation. *J Biomed Inform* 2014; **50**:196–204.
- 15 Lyons RA, Jones KH, John G *et al.* The SAIL databank: linking multiple health and social care datasets. *BMC Med Inform Decis Mak* 2009; **9**:3.
- 16 Welsh Government. StatsWales – Welsh Index of Multiple Deprivation. Available at: <https://statswales.gov.wales/Catalogue/Community-Safety-and-Social-Inclusion/Welsh-Index-of-Multiple-Deprivation> (last accessed 6 April 2021).
- 17 Eurostat. Revision of the European Standard Population – report of Eurostat’s task force – 2013 edition. Available at: <https://ec.europa.eu/eurostat/web/products-manuals-and-guidelines/-/ks-ra-13-028> (last accessed 5 November 2022).
- 18 Waterhouse J, Muir CS, Correa P, Powell J. *Cancer Incidence in Five Continents*, vol. III. Lyon: IARC Scientific Publications, 1976.
- 19 Ahmad OB, Boschi-Pinto C, Lopez AD *et al.* Age standardization of rates: a new WHO standard. Available at: https://cdn.who.int/media/docs/default-source/gho-documents/global-health-estimates/gpe_discussion_paper_series_paper31_2001_age_standardization_rates.pdf (last accessed 5 November 2022).
- 20 Vallejo-Torres L, Morris S, Kinge JM *et al.* Measuring current and future cost of skin cancer in England. *J Public Health (Oxf)* 2014; **36**:140–8.
- 21 Welsh Government. National standards for skin cancer services 2005. Available at http://www.wales.nhs.uk/sites3/documents/322/National_Standards_for_Skin_Cancer_Services_2005_English.pdf (last accessed 5 November 2022)
- 22 European Network of Cancer Registries. ENCR recommendations. Non melanoma skin cancer. Available at: <https://www.encl.eu/sites/default/files/pdf/skinrecs.pdf> (last accessed 5 December 2020).
- 23 Marcil I, Stern RS. Risk of developing a subsequent nonmelanoma skin cancer in patients with a history of nonmelanoma skin cancer: a critical review of the literature and meta-analysis. *Arch Dermatol* 2000; **136**:1524–30.
- 24 Musah A, Gibson JE, Leonardi-Bee J *et al.* Regional variations of basal cell carcinoma incidence in the U.K. using The Health Improvement Network database (2004–10). *Br J Dermatol* 2013; **169**:1093–9.
- 25 Schreuder K, Hollestein L, Nijsten TEC *et al.* A nationwide study of the incidence and trends of first and multiple basal cell carcinomas in the Netherlands and prediction of future incidence. *Br J Dermatol* 2022; **186**:476–84.
- 26 Cancer Australia. The 2002 national non-melanoma skin cancer survey. Available at: <https://www.canceraustralia.gov.au/publications-and-resources/cancer-australia-publications/2002-national-non-melanoma-skin-cancer-survey> (last accessed 5 November 2022).
- 27 Australian Institute of Health and Welfare. Health expenditure Australia 2019–20. Available at: <https://www.aihw.gov.au/reports/health-welfare-expenditure/health-expenditure-australia-2019-20/contents/about> (last accessed 31 January 2022).
- 28 Gallagher RP, Hill GB, Bajdik CD *et al.* Sunlight exposure, pigmentary factors, and risk of nonmelanocytic skin cancer: I. Basal cell carcinoma. *Arch Dermatol* 1995; **131**:157–63.
- 29 Kricke A, Armstrong BK, English DR, Heenan PJ. Does intermittent sun exposure cause basal cell carcinoma? A case-control study in Western Australia. *Int J Cancer* 1995; **60**:489–94.
- 30 Rosso S, Zanetti R, Martinez C *et al.* The multicentre south European study ‘Helios’. II: Different sun exposure patterns in the aetiology of basal cell and squamous cell carcinomas of the skin. *Br J Cancer* 1996; **73**:1447–54.
- 31 van Doorslaer E, Masseria C, Koolman X *et al.* Inequalities in access to medical care by income in developed countries. *Can Med Assoc J* 2006; **174**:177–83.
- 32 SatsWales. Welsh Index of Multiple Deprivation (WIMD) 2019. Available at: <https://statswales.gov.wales/Catalogue/Community-Safety-and-Social-Inclusion/Welsh-Index-of-Multiple-Deprivation> (last accessed 5 November 2022).
- 33 Welsh Government. The sunbeds (regulation) 2010 (Wales) Regulations 2011. Available at: <https://gov.wales/sites/default/files/publications/2019-04/the-sunbeds-regulation-act-2010-wales-regulations-2011.pdf> (last accessed 5 November 2022).
- 34 Welsh Government. Survey of sunbed use in Wales: summary briefing. Available at: <https://gov.wales/sites/default/files/statistics-and-research/2019-07/170927-survey-sunbed-use-summary-briefing-en.pdf> (last accessed 5 November 2022).
- 35 Wehner MR, Shive ML, Chren M-M *et al.* Indoor tanning and non-melanoma skin cancer: systematic review and meta-analysis. *BMJ* 2012; **345**:e5909.
- 36 Kelly C, Hulme C, Farragher T, Clarke G. Are differences in travel time or distance to healthcare for adults in global north countries associated with an impact on health outcomes? A systematic review. *BMJ Open* 2016; **6**:e013059.
- 37 Basra MKA, Finlay AY. The family impact of skin diseases: the Greater Patient concept. *Br J Dermatol* 2007; **156**:929–37.
- 38 Openshaw S. Ecological fallacies and the analysis of areal census data. *Environ Plan A* 1984; **16**:17–31.
- 39 Gehlke CE, Biehl K. Certain effects of grouping upon the size of the correlation coefficient in census tract material. *J Am Stat Assoc* 1934; **29**:169–70.

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